Ossification of the posterior longitudinal ligament in non-Asians: demographic, clinical, and radiographic findings in 43 patients

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Object. Ossification of the posterior longitudinal ligament (OPLL) is a disorder afflicting as many as 2% of East Asians. However, reports of OPLL in non-Asians have been sporadic in the medical literature. This study describes clinical and radiographic findings with OPLL in non-Asians at a tertiary care center treating a diverse multiethnic population.

Methods. Over a 6-year period, 43 patients not of East Asian descent presented to an urban tertiary medical center with OPLL. Patient data, including ethnicity, spinal cord function, Nurick grade, radiographic findings, OPLL subtype, and degree of cervical stenosis, were recorded.

Results. The average patient age was 59 years (range 32–92 years) with 18 women and 25 men. There were 22 Caucasian patients, 17 Hispanic patients, and 4 Black patients. With respect to the radiographic findings, OPLL morphology was continuous in 19, segmental in 17, mixed in 6, and other in 1. Average canal diameter was 7.6 mm (range 4.2–9.0 mm) at the most stenotic points. The mean Nurick grade was 2.95 at presentation, but 7 of the patients had OPLL identified incidentally and with early or minimal symptoms and signs of myelopathy.

Conclusions. Ossification of the posterior longitudinal ligament in non-Asians demonstrates similar demographic and radiographic characteristics as in East Asians. The representation of different ethnic groups mirrors the demographics of the medical center population in general, showing no specific predilection for particular ethnic groups. Surgical decompression in appropriately selected patients results in similar rates of improvement when compared with the Japanese literature. (DOI: 10.3171/2010.12.FOCUS10277)

Key Words • ossification of the posterior longitudinal ligament • Asian • demographics • ethnicity

Ossification of the posterior longitudinal ligament is characterized by growth of the posterior longitudinal ligament followed by ossification and the growth of this ectopic bone formation. While relatively uncommon, this disease results in progressive degeneration and increasing spinal stenosis. The final result of disease progression is severe cervical spinal cord dysfunction with its attendant neurological sequelae.

Ossification of the posterior longitudinal ligament was first reported in 1838 in Europe, but the disorder received recognition in the 1960s by Japanese spine surgeons. As disease reports proliferated, it became apparent that this disease was significantly different from cervical spondylotic myelopathy, a disease more common in non-Asians. It is now generally believed that OPLL has a prevalence of 1.9%–4.3% in the Japanese population. As such, it became associated with a higher frequency of occurrence in Asian populations, and it is now recognized that OPLL represents a significant public health problem in Japan.

Abbreviation used in this paper: OPLL = ossification of the posterior longitudinal ligament.

Whereas the exact causes of OPLL remain elusive, specific genetic and environmental factors have been associated with this disease. Mutations in the gene for nucleotide pyrophosphatase have been associated with an OPLL variant in rodents (tiptoe walking mouse), as well as with a higher frequency in patients with OPLL. Environmental factors such as exposure to high concentrations of fluoride have also been implicated in the pathogenesis of OPLL, but genetic links to the disease are concordant with what appears to be a propensity for afflicting specific ethnic groups.

In the last half-century the bulk of scientific and clinical reports on OPLL have originated from Japan. This increased attention to OPLL in East Asia was suspected as a potential contributor to higher reported incidences in East Asian populations. In 1980, Izawa reported on a radiographic survey of patients in Japan, Korea, Hawaii, Minnesota, and Germany. Using plain radiographs to make the diagnosis, thousands of patients were studied. While the incidence of degenerative disease was similar between groups, OPLL among asymptomatic Japanese adults was found to be 2.0%, compared with 0.95% in...
Koreans and 0.17%–0.20% in Caucasians. Corroborative evidence emerged from a US study of 1000 plain cervical radiographs by Firooznia et al.; in that report, OPLL was identified in 0.7% of New Yorkers in general.

While most North American spine surgeons have observed sporadic cases of OPLL in non-Asians, there have been few reports of OPLL in non-Asian populations, and these have generally been limited to small case series or pooled populations from multiple centers. This report from a tertiary academic medical center serving a diverse ethnic population was directed at elucidating the presentation, clinical findings, and radiographic characteristics in non-Asian North American patients with OPLL.

Methods

Study Population

Over a 6-year period, 43 patients with OPLL who were not of East Asian descent presented to an urban tertiary medical center in Southern California. The center services a diverse population composed of approximately 50% Hispanic, 15% Black, 15% Caucasian, and 10% Asian patients. Patients presented through either the outpatient clinic or the emergency department. Patient data, including ethnicity, spinal cord function, Nurick grade, radiographic findings, OPLL subtype, and degree of cervical stenosis were recorded. Ethnicity was determined by patient self-report.

Imaging Protocol

All patients underwent cervical imaging, including flexion-extension radiographs, CT scanning, and MR imaging, which was the standard protocol evaluation process for these patients. The degree of stenosis was determined by measuring the sagittal canal diameter at the most stenotic cervical level on high-resolution axial MR images. Measurements in millimeters were determined at the midline.

Diagnosis of OPLL

The diagnosis of OPLL was made if 3 conditions were met: 1) clinical symptoms consistent with cervical spinal cord compression were responsible for presentation to the clinician; 2) MR imaging evidence of cervical stenosis as defined by a minimal midsagittal canal measurement of 9 mm or less; and 3) CT demonstrated that the stenosis was due primarily to a calcified mass consistent with the morphology of OPLL. Ossification of the posterior longitudinal ligament morphology was classified according to the scheme by Hirabayashi et al. (Fig. 1).

Results

Patient Demographics

Patient demographic data are shown in Table 1. The average patient age was 59 years, with a range of 32 to 92 years. There was a slight male predominance with 18 women and 25 men. Thirty-six patients presented with signs and symptoms of myelopathy, and the mean Nurick grade was 2.95 at presentation (Figs. 2 and 3). However, 7 patients had OPLL identified in their early stages with minimal symptoms and no reflex changes. These patients typically had upper extremity numbness or presenting symptoms of radiculopathy due to cervical nerve root compression. With respect to ethnicity there were 22 Caucasian patients, 17 Hispanic patients, and 4 Black patients. The mean follow-up was 13

![Fig. 1. Illustration of the 4 types of OPLL morphology as described by Hirabayashi et al.](image-url)
± 4.7 months for those undergoing operative intervention. For the 8 patients who did not undergo operative intervention (at our institution), mean follow-up was 3.2 months. Four of these patients had only 1 clinic visit.

Characteristics of OPLL

Ossification of the posterior longitudinal ligament morphology was determined based on axial CT and sagittal reconstruction images. The morphology of OPLL was continuous in 19 patients, segmental in 17, mixed in 6, and other in 1. The average canal diameter was 7.6 mm (range 4.2–9.0 mm) at the most stenotic points. The ossification spanned a mean of 2.2 spinal segments (range 1–6 segments). There was no association between the OPLL morphology and patient ethnicity. Two cases had an associated thoracic involvement with OPLL.

Surgical Intervention

With respect to surgical treatment, 35 patients underwent decompression. An anterior approach for corpectomy, fusion, and plating was performed in 7 patients, laminectomy with instrumented fusion in 15, laminoplasty in 9, nonoperative management in 24, and combined anterior-posterior surgery in 4. Expectant nonoperative management was applied in 8 patients. These patients had minimal symptoms and did not desire operative treatment given the risks of surgery. Of the 35 patients treated operatively, 29 showed an improvement in their Nurick grades, resulting in a mean improvement of 1.3 points for the entire surgical cohort.

There was no significant difference in neurological improvement between the anterior, posterior, and anterior-posterior surgical cohorts; the mean improvement in Nurick grade for these groups was 1.2, 1.4, and 0.8 points, respectively. However, 2 patients experienced neurological worsening. Both of these patients had severe preoperative stenosis and were not ambulating independently prior to intervention, and both patients underwent anterior decompressive surgery (Fig. 4). Surgical complications included dural tears in 5 patients who underwent anterior surgery, with 3 requiring additional surgical intervention; C-5 nerve root palsy in 4 patients (75% from posterior surgery); persistent dysphagia in 3 patients (100% from anterior or combined anterior/posterior surgery); and wound infection or breakdown in 2 patients (100% from posterior surgery).

Discussion

In this report we describe our experience with a series of 43 non-Asian patients with OPLL. In our series the clinical and radiographic presentation of OPLL was similar to previous series from East Asia. We found a varied distribution of OPLL morphologies, with a predominance of continuous and segmental forms, similar to that reported in series with Japanese patients. This study builds on several previous reports of OPLL in non-Asians. In Trojan et al.’s review23 of 73 cases of OPLL in non-Asians, several similarities were found with reports from the Japanese literature. These findings included: 1) male predominance, 2) peak age of symptoms in the 6th decade of life, 3) varied clinical presentations, 4) predominance for the cervical spine, and 5) association with other enthesopathies such as diffuse idiopathic skeletal hyperostosis. In the series of Jayakumar et al.,13 47 symptomatic Asian Indian patients of Caucasoid origin were studied. Sixty-five percent of these patients were found to have continuous-
type OPLL, and the disease predominantly affected the upper cervical spine. Epstein\textsuperscript{2–4} has reported the largest series of patients with OPLL in the US, but the reports have provided little information on patient ethnicity.

**Surgical Interventions**

Our selection of surgical approach was based upon the guidelines of Hirabayashi et al.,\textsuperscript{9} favoring posterior indirect decompressive approaches (laminectomy with instrumented fusion and laminoplasty) for longer segment and continuous-type OPLL, and anterior approaches for younger patients with more focal lesions. Furthermore, anterior corpectomies were favored when more that 60% of the spinal canal was compromised, given the higher rates of improvement compared with indirect through posterior approaches.\textsuperscript{11,16} Thus, there were preoperative differences between the patients who underwent different surgical treatments, with a bias toward worsened clinical status for anterior surgery.

Both of the cases with neurological worsening also occurred with anterior or combined anterior/posterior surgery. This may have been due to this selection bias as these patients had more severe radiographic spinal cord compression and worse neurological status at presentation. However, all cases of dural penetration were also associated with anterior surgery, and these events may have been correlated with direct iatrogenic mechanical injury to the spinal cord. Other surgical complications were as expected from previous patient series, with higher rates of dysphagia following anterior surgery and C-5 nerve root palsy from posterior decompression.

**Complications**

The higher incidence of complications in patients with OPLL compared with cervical spondylotic myelopathy has been well described. Dural tears, unusual associated with anterior surgery for cervical spondylosis, are common in patients with OPLL given the adherence and/or penetration of the dura mater by the osseous overgrowth. In many instances of advanced disease it may be impossible to separate the dura from the offending lesion. In our more recent practice we have adopted the “floating corpectomy” technique advocated by Japanese authors.\textsuperscript{25} This approach leaves a thin shell of OPLL adherent to the dura, but frees it from the vertebral body, allowing it to migrate en bloc into the ventral corpectomy defect. This maneuver can save time and blood loss by avoiding much of the microdissection when stripping the OPLL away, and offers the additional advantage of keeping the drilling at the lateral margins of the spinal canal, where the ventral epidural space is commonly patent.

Higher rates of neurological worsening can be due to numerous factors, including: 1) iatrogenic spinal cord or anterior spinal artery injury due to the absence of dura; 2) the profound compromise of the spinal canal space, rendering drilling and bone removal more treacherous; and 3) a compromised spinal cord more susceptible to spinal cord injury from ischemia and hypotension. We pay particular attention to the vulnerability of the spinal cord in patients with OPLL, ensuring fiberoptic intubation, maintenance of a mean systolic pressure greater than 85 mm Hg, and using motor evoked potential monitoring.

Postoperative dysphagia and dysphonia are also more common in this population given the presence of anterior vertebral osteophytes and diffuse idiopathic skeletal hyperostosis. In addition, the need for anterior exposures at C-2 and C-3 are more likely to jeopardize normal swallowing function. In our practice we minimize this risk by careful, sharp, anterior neck dissection so that retractor pressure and stretching of soft tissues are minimized. Prolonged surgeries are also managed with periodic retractor loosening.

Nerve root palsies at C-5 are also more common given the severity of spinal cord compression. This risk is managed by careful electromyographic monitoring of the biceps muscle during surgery. Any aberrant electromyographic activity may be indicative of C-5 nerve root irritation from stretching. This is managed with the addition of a C4–5 foraminotomy if a posterior decompression is used.

**Classification by Ethnicity**

This report is the largest series of OPLL cases from a single institution focused on ethnicity. The case series is derived from the diverse Southern California population, which has a high representation of Caucasians, Asians, Blacks, and White and Black Hispanics. In our series we were unable to identify a predominance of OPLL in any particular non-Asian group. However, classification of patients by ethnicity remains problematic and artificial. While it remains undisputed that there are distinct phenotypic

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**Fig. 4.** Images obtained in a 63-year-old Caucasian man who presented confined to a wheelchair with severe tetraparesis. Sagittal MR imaging (A) and axial CT (B) demonstrated severe stenosis to less than 4 mm at the widest (sagittal diameter) point. The patient underwent a combined anterior-posterior surgery for decompression and stabilization (C). An intraoperative dural tear resulted in an anterior neck pseudomeningocele noted on CT (D), which required subsequent surgery involving patching and the application of a fibrin sealant.
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characteristics within populations that can be attributed to the concept of race or ethnicity, the number and types of subcategories can be quite varied and disputed. The typical Linnaean variety of classification relies on a partitioning of individual patients into distinct ethnic groups. Other methods of classification, such as a cladistic or clinal approach, may eventually prove more useful for ethnic designations, avoiding the need for discrete and separate groups. In addition, this methodology more closely models the genetic variations found within human populations and likely provides more reliable predictive information. In this study we classified patients according to self-report, which is problematic as the patient’s self-assigned identity may bear no relation to physical traits or genetic constitution.

Another major drawback of this study is that our patient cohort was not population-based. Whereas our patients were drawn from a cohort of approximately 2.5 million patients, complete capture of all patients with OPLL was similar to that of East Asian patients, it is important for neurosurgeons working with less diverse populations to recognize that OPLL occurs with some frequency in non-Asian ethnic groups. Preoperative CT scanning can be useful when OPLL is suspected to assist in surgical planning and reduce the risk of complications.

Conclusions

This report describes our experience treating cervical OPLL in a diverse US population. While the clinical presentation, radiographic characteristics, and treatment were similar to that of East Asian patients, it is important for neurosurgeons working with less diverse populations to recognize that OPLL occurs with some frequency in non-Asian ethnic groups. Preoperative CT scanning can be useful when OPLL is suspected to assist in surgical planning and reduce the risk of complications.

Disclosure

Dr. Wang serves as a consultant for DePuy Spine, Biomet Spine, Aesculap Spine, and Globus Spine. He also holds a patent with DePuy Spine.

Author contributions to the study and manuscript preparation include the following. Conception and design: Wang. Acquisition of data: Wang. Analysis and interpretation of data: Wang. Drafting the article: both authors. Critically revising the article: Thambuswamy. Included in the article: both authors. Critically revising the article: Thambuswamy. Reviewed final version of the manuscript and approved it for submission: both authors.

References


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