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Original Investigation

Clinical and Radiological Characteristics of Ossification of the Posterior Longitudinal Ligament of the Cervical Spine in Patients without Myelopathy: Results of a 1-year Pilot Study

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ABSTRACT

AIM: We analyzed the demographics, clinical and radiological characteristics of non-myelopathic cervical ossification of the posterior longitudinal ligament (OPLL) patients.

MATERIAL and METHODS: This study included 115 patients with cervical OPLL but without myelopathy. Demographic features of age, sex, and the nature of the patient's symptoms were evaluated. Clinically, visual analog scale (VAS) scores for the neck and arm, and Japanese Orthopaedic Association (JOA) scores were evaluated. Radiologically, the number of involved segments, type of OPLL, and the maximal compression ratio were analyzed using a computed tomography (CT) scan. The relationship between clinical scores and radiological parameters was analyzed.

RESULTS: At the time of diagnosis, there was absence of symptoms in 23, axial neck pain in 44, radiculopathy in 40, and tingling sensation of fingers in 8. VAS score for the neck was 4.42 and that for the arm was 3.64. The mean JOA score was 16.13. Radiologically, the mean number of involved segments was 3.55. The type of OPLL mass was continuous, mixed, segmental, and local in 10, 43, 42, and 20 cases, respectively. The maximal compression ratio was 0.38. There was a significant relationship between the maximal compression ratio and the number of involved segments ($p<0.001$). No relationship was found between clinical symptoms, clinical scores, and radiological findings.

CONCLUSION: Some non-myelopathic cervical OPLL patients showed no symptoms, some presented axial neck pain and radiculopathy. Radiologically, a significant relationship between the maximal compression ratio and the number of involved segments was found. However, there was no relationship between clinical symptoms and radiological findings in neurologically intact patients.

KEYWORDS: Cervical spine, OPLL, Asymptomatic, Non-myelopathic

INTRODUCTION

Ossification of the posterior longitudinal ligament (OPLL) involves the abnormal calcification of the posterior longitudinal ligament that mostly occurs at the cervical spine. OPLL-induced spinal compression may lead to neurologic symptoms and surgery is required in cases with severe neurologic deficit. Although the literature is rich regarding various surgical treatments and outcomes, there are few studies reporting data regarding conservative management of these patients (3,9,13). While early diagnosis and careful observation of OPLL patients is important, the nature of the symptoms and that of asymptomatic OPLL have been unclear. Also, patients with OPLL may be asymptomatic or may have simple neck pain, radiculopathy, or myelopathy, regardless of radiological findings. Previous reports that examined the natural course of OPLL included a simple analysis of the development of new neurological symptoms during follow-up. Furthermore, most of the studies evaluated

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only operated cases. To our knowledge, no study has reported on the analysis of OPLL patients without myelopathy.

The purpose of this study was to analyze the demographic, clinical, and radiological characteristics of cervical OPLL in patients without myelopathy and to serve as a basic reference for evaluating the treatment strategy. Moreover, the effect of OPLL on their symptoms was examined especially in patients without myelopathy.

MATERIAL and METHODS

Between March 2010 and December 2012, a total of 115 consecutive patients who were diagnosed with cervical OPLL and had no neurological deterioration at the initial period were examined retrospectively. Institutional Review Board approval was obtained for this study. Neurological deterioration was defined as decreased motor power or sensory tone related to the cervical compression level. Among patients who suspicious of myelopathy, those with a grip and release test result of greater than 20/10 seconds, negative results in the Hoffman test, and those with no objective gait deterioration were considered neurologically unaffected and were included in this study. Patients, who initially showed neurological symptoms were followed up for less than 1 year, and/or lacked radiological evaluation with computed tomography (CT) were excluded from this study. Conservative management was provided to patients with axial neck pain or radiculopathy, but who did not have neurological impairment. Surgery was performed in patients with myelopathy, radiculopathy with neurological deterioration, or sustained radicular pain despite aggressive medical treatment for more than 6 weeks. Three hundred sixty-five patients were diagnosed with OPLL, and 151 underwent operative treatment (Figure 1). Among the 214 patients who received conservative management, 53 were excluded because they lacked radiological evaluation or had been followed up for less than 1 year. Another 46 patients were excluded because of neurological deterioration at initial visit.

In all patients, medical records, questionnaires, and CT scans were evaluated. The demographic analysis included the nature of the patient's symptoms, concomitant medical disease, familial history of OPLL, and the progression or development of symptoms. The nature of patients' symptoms was categorized as 1) absence of any symptom, 2) axial neck pain, 3) pain radiating to the upper extremity, 4) tingling sensation of the fingers, 5) difficulty in gait or clumsiness of the hand, and 6) others. Clinically, the visual analog scale (VAS) scores of the neck and arm, and Japanese Orthopaedic Association (JOA) scores were evaluated. Radiological analysis, including the number of involved segments, type of OPLL, and maximal compression ratio using a CT scan, was performed. The maximal compression ratio (the ratio of the length occupied by the OPLL to the normal spinal canal length) was determined by measuring the space available for the spinal cord (SAC) and the length of the unaffected spinal canal on sagittal view on the CT scan in the maximal compression area (Figure 2). The relationship between the clinical scores and radiological parameters was analyzed.

CT data for all participants was analyzed by 2 orthopedic spine surgeons based on the number of involved segments, types of ossification, and maximal compression ratio in the sagittal view (10). The CT images (Discovery CT 750 HD, GE, USA) were 1-mm thick in the disk spaces and 2-mm thick in the vertebral body and sagittal planes. The patients who showed spondylotic or cord compression lesion in other parts outside OPLL were excluded from this analysis. The OPLL types were categorized as local, segmental, continuous, and mixed, based on the classification determined by the

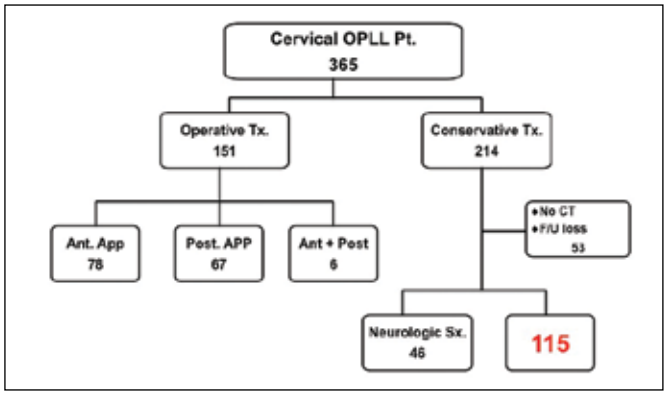

Figure 1: Flow diagram showing patient allocation.

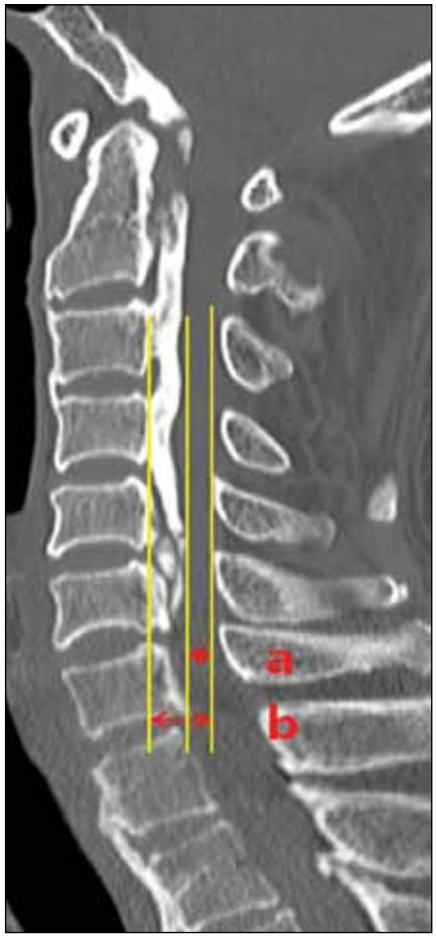

Figure 2: Maximal compression ratio. **a)** Sagittal SAC, **b)** Sagittal canal length, **(b-a)/b:** maximal compression ratio.

Investigation Committee on OPLL of the Japanese Ministry of Public Health and Welfare (12). The local type of OPLL is observed at the posterior rim of the intervertebral disc; the segmental type is observed at the posterior rim of each vertebral body; the continuous type is observed continuously over many segments; and the mixed type is a combination of the above types.

All CT measurements were performed by the picture archiving and communication system (PACS) (m-view™, MaroTech, Seoul, Korea) system. Two blinded observers independently interpreted each of the radiological findings, and the average measurements were used in the analysis. When there was a difference in the interpretation, the 2 observers drew a mutual conclusion, which was used for analysis. To verify the reliability of measured values, intra- and inter-observer consistencies were checked using the kappa coefficient test.

SPSS 18.0 (SPSS Inc., Chicago, IL, USA) was used for statistical analysis and to determine the relationship between radiological parameters. The relationship between the patient's symptoms, clinical scores, and radiological findings was also analyzed using correlation analysis or chi-square analysis. The results were deemed statistically significant at $p < 0.05$.

RESULTS

Clinical Features

The mean age of the 115 recruited patients was 57.3 ± 9.16 (range, 32–86) years, and 56 men and 59 women were included. The mean follow-up period was 15.2 ± 7.9 (range, 13–37) months. At the time of diagnosis, there were no symptoms in 23 patients, axial neck pain in 44, radiculopathy in 40, and tingling sensation of the fingers in eight. Six patients complained of difficulty in gait or clumsiness of the hand without objective neurological abnormality. Two patient underwent operative treatment during the follow up. One was due to aggravation of radicular pain and the other due to the development of objective gait disturbance.

Fifty-nine patients had no concomitant medical disease, and 29, 14, and six patients had hypertension, diabetes mellitus, and thyroid disease, respectively. There were only two patients with a family history of OPLL (Table I).

The mean neck and arm VAS scores were 4.42 (range, 0–10) and 3.64 (range, 0–10), respectively. The mean JOA score was 16.13. A significant relationship was found between the neck and arm VAS scores ($p < 0.01$); the arm VAS scores also showed a significant relationship with JOA scores ($p = 0.002$) (Table II).

Radiological Features

Inter-(K1) and intra-(K2) observer consistencies of radiologically measured values indicated high levels of reliability (K1 = 0.81, K2 = 0.84).

The mean number of involved segments of OPLL was 3.55. The type of OPLL mass was continuous, mixed, segmental, and local in 10, 43, 42, and 20 cases, respectively (Figure 3A-D).

The mean maximal compression ratio was 0.38. There was a significant relationship between the maximal compression ratio and the number of involved segments ($r = 0.471$, $p < 0.001$). The continuous and mixed type showed many more involved segments ($p < 0.001$), and the maximal compression ratio was high ($p < 0.001$) (Table III).

Relationship Between Clinical and Radiological Features

There was no significant relationship between clinical symptoms and the number of involved segments ($p = 0.83$), type of OPLL ($p = 0.88$), and the maximal compression ratio ($p = 0.167$). Neck VAS, arm VAS, and JOA scores were not related to radiological parameters.

DISCUSSION

The investigation Committee on Ossification of the Spinal Ligaments of the Japanese Ministry of Public Health, Labour, and Welfare performed the first national survey of OPLL in

Table I: Summary of Demographic Data of the Patients

Factor	Number of Cases (%)
Sex	
Male	56 (48.69)
Female	59 (51.3)
Symptoms at time of diagnosis	
No symptoms	23 (20.1)
Axial neck pain	44 (38.26)
Radiculopathy	40 (34.78)
Tingling sensation of fingers	8 (6.95)
Concomitant medical disease	
No	59 (51.30)
Hypertension	29 (25.21)
Diabetes Mellitus	14 (12.17)
Thyroid disease	6 (5.21)

Table II: Relationship between Clinical Scores

	Neck VAS*	Arm VAS	JOA**
Neck VAS Correlation ratio p value	1	0.488 <0.01	0.07 0.458
Arm VAS Correlation ratio p value		1	0.28 **0.002**
JOA			1

**: visual analog scale.*
***: Japanese Orthopaedic Association score.*

1975, and 2142 OPLL patients were registered. Ninety-five percent of the patients had clinical symptoms, but 5% of these patients were asymptomatic. As many as 16.8% of the patients needed assistance in their activities of daily living; 5.4% of patients showed a rapid aggravation of symptoms, and 11.4% of patients showed chronic aggravation. These data were meaningful. However, most of the patients had clinical symptoms at the time of diagnosis, and there were some limitations in the evaluation of the natural course of OPLL. In our study, 168 of the 365 cervical OPLL patients (46%) were neurologically intact. Even though there is some possibility of selection bias, we can evaluate a large number of asymptomatic OPLL cases.

Table III: Relationship Between Type of OPLL and Radiological Features

	No. of involved segment	Maximal compression ratio
Continuous type	3.54	0.47
Mixed type	4.76	0.47
Segmental type	3.11	0.29
Local type	1.95	0.35
p value	**<0.001**	**<0.001**

OPLL is formed mainly through enchondral ossification, and part of it develops through membranous ossification. McAfee et al. reported that the pathology of most OPLL cases is composed of a lamellar bone with mature Haversian canals, in addition to fibrous cartilages and woven bones wrapped with calcified cartilage (6). Ossification begins at the connection area of lateral limbus of vertebral body and posterior longitudinal ligament, and progresses up and down. OPLL progresses slowly, and shows a wide variety of neuronal

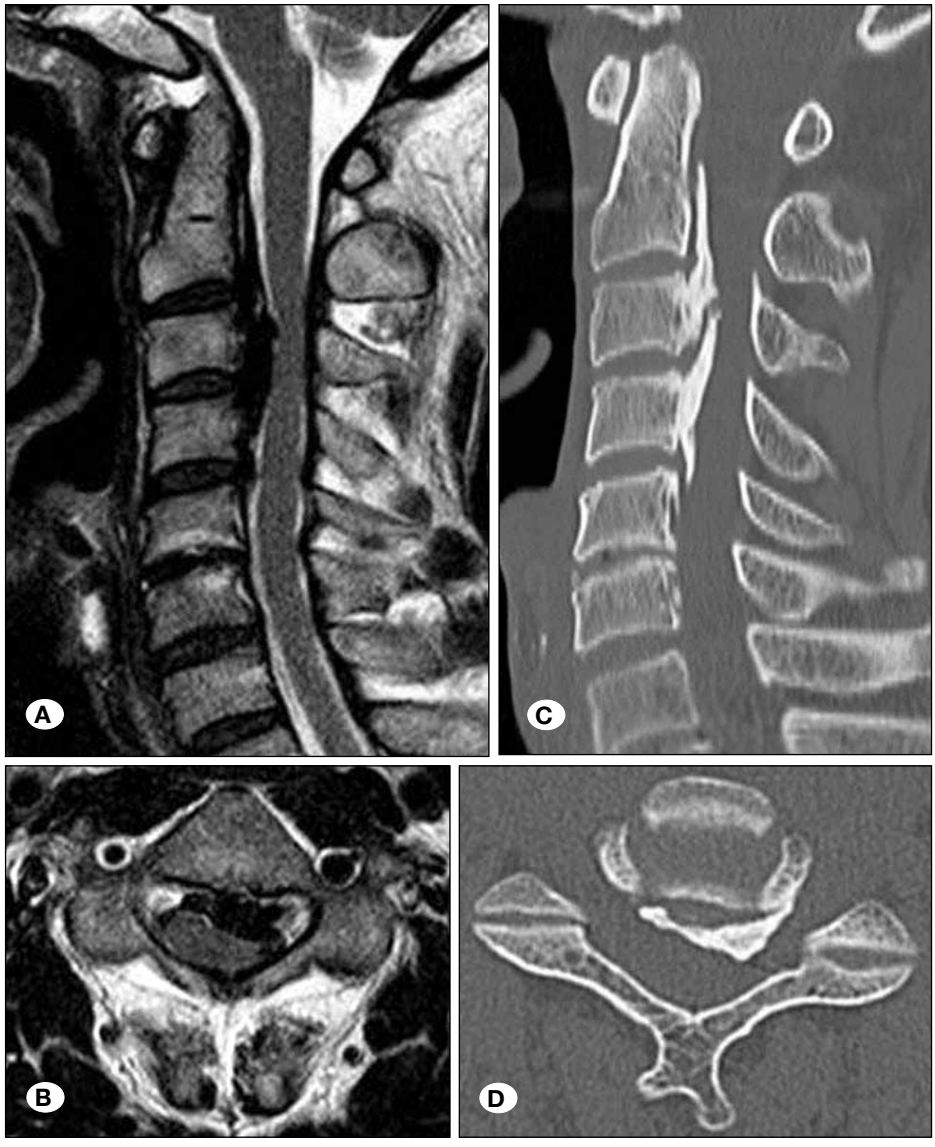

Figure 3: A 42-year-old man complained of neck pain with no myelopathic symptoms. Magnetic resonance imaging and computed tomography scan showing ossification of the posterior longitudinal ligament mixed type in C2–6, resulting in spinal canal stenosis. The Japanese Orthopaedic Association (JOA) score, neck visual analog scale (VAS) score, and arm VAS score were 17, 5, and 3, respectively. **A, B)** Sagittal and axial view in MRI. **C, D)** Sagittal and axial view in CT.

degeneration by compressing the spinal cord and nerve roots. Jeon et al. assume that segmental OPLL progresses to mixed OPLL or continuous OPLL and that mixed OPLL progresses to continuous OPLL (2). When they evaluated the age of each type of OPLL, localized and segmental OPLL types were more often identified in younger patients, whereas mixed and continuous types of OPLL were more often identified in older patients. Our result also showed a significant relationship between the maximal compression ratio and the number of involved segments. The continuous and mixed types showed much more involved segments, and the maximal compression ratio was high. In the cases of mixed or continuous type, OPLL has progressed and can involve multiple levels simultaneously due to the growth of the OPLL mass.

Matsunaga et al. reported that only 17% of the patients without myelopathy at the first visit developed myelopathy during the follow-up (5). Pham et al. (9) also showed that patients without myelopathy had a 16.7% chance of progressive myelopathy over an aggregate mean follow-up of 14.6 years. However, there was no report about the clinical pattern or radiological features of asymptomatic OPLL patients. Our study showed that 197 patients of the 365 patients who were diagnosed with cervical OPLL required surgery because of neurological deterioration, and 46% of the OPLL patients did not need surgical intervention. Neurologically unaffected patients did not have any symptom (20%), or had neck pain (38%), or radicular pain (34%). Sasaki et al. reported that 86.7% of the OPLL patients had symptoms, while 77% of the non-OPLL patients had symptoms (11). Their result showed many OPLL patients did not have severe symptoms compared with that in the non-OPLL group. We can draw two important conclusions from these results. While most of the subjects with OPLL had pain between the neck and arm, a number of patients with OPLL did not receive treatment (11). Another point is that OPLL patients do not show severe symptoms but rather radiological findings, and surgical intervention should be considered according to the patients' symptoms, especially myelopathy.

Our result showed no significant relationship between clinical symptoms and radiological findings in neurologically intact cervical OPLL patients. The mean JOA score was 16.13. Conservative treatment is required for patients with a JOA score of at least 14 points (7). Morio et al. reported a mean JOA score of 12.9 and 16.1 in patients with and without myelopathy, respectively (8). Based on the review of four studies by Pham et al., the mean aggregate JOA score of 111 OPLL patients was 15.3 on presentation; these patients were subsequently managed with conservative treatment (9).

In order to establish radiological predictors for the development of myelopathy in patients with OPLL, Matsunaga et al. performed a multicenter cohort study analyzing 156 patients with a mean follow-up of 10.3 years (4). They found that 60% or greater spinal canal stenosis; large ROM of the cervical spine and laterally deviated-type OPLL were risk factors for developing myelopathy. The maximal compression was less than 40% in asymptomatic OPLL cases in our study. Chang et al. evaluated the factors related to the manifestation of myelopathic symptoms in patients with OPLL (1). In patients with OPLL of the cervical spine, the manifestation of myelopathic symptoms was not related to the range of motion of the cervical spine or the number of involved segments. However, age at the time of diagnosis and the maximum compression of the spinal canal were related factors. However, there is controversy surrounding the management of asymptomatic patients. In a survey of 774 spine surgeons, the majority deemed the presence of clinically symptomatic radiculopathy to predict progression to myelopathy in nonmyelopathic patients with cervical stenosis (14).

This was a pilot study to evaluate the clinical and radiological features of asymptomatic OPLL patients. Our study is limited in that it included a short-term follow-up period, and the data obtained were not sufficient to determine the clinical application for predicting the clinical course of asymptomatic OPLL patients. Our center has renown for OPLL and many patients were consulted from other institutions. That is the reason we can collect many asymptomatic OPLL patients. This can be another limitation as selection bias. However, we evaluated a relatively large number of asymptomatic OPLL patients. With serial follow up and further evaluation, these data can be beneficial to understand the natural history of OPLL.

CONCLUSION

The demographic characteristics, clinical presentation, and radiography findings of OPLL in neurologically intact cervical OPLL patients were analyzed, which could serve as a basis for further study on and treatment of OPLL. Some neurologically intact cervical OPLL patients showed no symptoms; however, some presented with axial neck pain and radiculopathy. Most patients had no family history of OPLL.

Radiologically, a significant relationship between the maximal compression ratio and the number of involved segments was found. However, there was no relationship between clinical symptoms and radiological findings in neurologically intact patients.

ACKNOWLEDGMENT

This study was conducted in accordance with the declaration of Helsinki. This study was conducted with approval from the ethics committee of Inje University Haeundae Paik Hospital (129792-2014-038).

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