scoring system such that the presence of DISH could be assessed reproducibly. This system scores individuals who fulfill the Resnick criteria by numerically classifying each vertebral level based on the amount of ossification and whether partial or complete bridging of the disc space is present [3].

Although some reports have indicated a significant association between DISH and ossification of the posterior longitudinal ligament (OPLL) [4–7], DISH is thought to be an asymptomatic condition in many affected individuals; however, several clinical symptoms have been described including pain, limited range of spinal motion, and increased susceptibility to unstable spinal fractures after trivial trauma [8]. In addition, dysphagia and airway obstruction at the cervical levels [8, 9], as well as radiculopathy and spinal injury after spinal fracture [10–12], have been reported as clinical manifestations of DISH.

Although the condition is recognised in many parts of the world [13–20], there are relatively few population-based studies concerning its prevalence. Such data are important in order to characterise the burden of the disease. In addition, regarding its characteristics, several epidemiologic studies have reported that DISH is observed mainly in the elderly, and that prevalence increases with age [18, 19]. Men are affected by DISH much more frequently than women [20]. Although metabolic disturbance is hypothesised to be a factor [21, 22], the aetiology of the condition remains unknown.

Based on the definition of DISH as the radiographic finding of calcification or ossification, it appears that the condition might be associated with osteoarthritis (OA) of the spine. The severity of OA, as observed on radiography, was determined according to Kellgren-Lawrence (KL) grading as follows [23]: KL0, normal; KL1, slight osteophytes; KL2, definite osteophytes; KL3, joint or intervertebral space narrowing with large osteophytes; and KL4, bone sclerosis, joint or intervertebral space narrowing, and large osteophytes. KL2 is commonly used as the diagnostic criterion for lumbar spondylosis (LS) or OA at other sites. Thus, LS—defined as KL2 (defined as the definite presence of osteophytes)—could easily be associated with DISH. However, there are few reports to confirm the association between DISH and severe LS with the criterion of KL3 (defined as the presence of intervertebral space narrowing) or KL4 (defined as the presence of bone sclerosis). In addition, there are few reports to clarify the association between DISH and OA at other sites, such as the knees.

We conducted a survey, known as the Research on Osteoarthritis/osteoporosis Against Disability (ROAD) study, using a population-based cohortto determine the prevalence of DISH using lateral whole-spine radiography in recently examined subjects, which included men and women in Japan. Another aim of our study was to clarify the association of DISH with LS and knee osteoarthritis (KOA) based on KL grade.

Materials and methods

Outline of the ROAD study

We conducted the present study using the cohorts established in 2005 for the ROAD study—a nationwide, prospective study of OA comprising population-based cohorts in several communities in Japan. Details of the cohort profile have been reported elsewhere [24, 25]. Briefly, from 2005 to 2007, we developed a baseline database that included clinical and genetic information of 3,040 residents of Japan (1,061 men, 1,979 women) with a mean age of 70.3 (SD, 11) years [men: 71 (SD, 10.7) years, women: 69.9 (SD, 11.2) years]. Subjects were recruited from resident registration listings in three communities with different characteristics: 1,350 subjects (465 men, 885 women) from an urban region in Itabashi, Tokyo; 864 (319 men, 545 women) from a mountainous region in Hidakagawa, Wakayama; and 826 (277 men, 549 women) from a coastal region in Taiji, Wakayama.

Participants completed an interviewer-administered questionnaire of 400 items that included lifestyle information, such as occupation, smoking habits, alcohol consumption, family history, medical history, physical activity, reproductive variables, and health-related quality of life. The questionnaire was prepared by modifying the questionnaire used in the Osteoporotic Fractures in Men Study (MrOS) [26]; some new items also were added to the modified questionnaire. Participants were asked whether they took prescription medication daily or nearly every day (no = 0, yes = 1). If the participants did not know the reason for the prescribed medication, they were asked to bring their medication to the medical doctor (NY).

Anthropometric measurements, including height (cm), body weight (kg), arm span (cm), bilateral grip strength (kg), and body mass index (BMI, kg/m^2) were recorded for each patient. Medical information was recorded by experienced orthopaedic surgeons on systematic, local, and mental status, including information on back, knee, and hip pain; swelling and range of motion of the joints; and patellar and Achilles tendon reflexes.

Eligible subjects of the present study

In the ROAD study, radiographic examination of the thoracic spine was performed only in subjects in mountainous and coastal regions. These subjects also underwent blood and urinary examinations. In the present study, among 1,690 subjects (596 men, 1,094 women) in mountainous and coastal regions in the ROAD study, we excluded 43 whose radiograph quality was so poor that it was difficult to observe the sites of thoracic–lumbar junction and lumbosacral junction; thus, we analysed 1,647 participants (573 men, 1,074 women) ranging in age from 23 to 94 years (mean: 65.3 years, men: 66.3 years, women: 64.7 years).

Study participants provided written informed consent, and the study was approved by the ethics committees of the University of Wakayama Medical University (No. 373) and the University of Tokyo (No. 1264 and No. 1326).

Radiographic assessment

Plain radiographs of the cervical, thoracic, and lumbar spine in the anteroposterior and lateral views, and bilateral knees in the anteroposterior view with weight-bearing and foot-map positioning were obtained. DISH was diagnosed according to the following criteria, defined by Resnick and Niwayama [2]: (1) flowing ossification along the lateral aspect of at least 4 contiguous vertebral bodies, (2) relative preservation of intervertebral disc height in the involved segments, and (3) absence of epiphyseal joint bony enclosing and sacroiliac joint erosion. In the assessment of lateral radiographs, since it was difficult to read the C7/Th1 to T3/4 vertebral levels, 'whole spine' in the present study implies radiographs assessed from the C0/1 to C6/7, Th4/5 to Th12/L1, and L1/L2 to L5/S1 levels.

The radiographic severity of OA was determined according to the above-mentioned KL grade [20]. Radiographs of each site (i.e., vertebrae and knees) were examined by a single experienced orthopaedic surgeon (SM) who was blinded to the participants' clinical status. In the present study, the maximum grade, diagnosed in at least 1 intervertebral level of the lumbar spine or at least 1 knee joint, was regarded as the subject's KL grade.

Statistical analysis

All statistical analyses were performed using STATA statistical software (STATA Corp., College Station, TX, USA). Differences in proportions were compared using the Chi-square test. Differences in continuous variables were tested for significance using analysis of variance for comparisons among multiple groups or Scheffe's least significant difference test for pairs of groups.

To test the association between the presence of DISH and LS and/or KOA, we used logistic regression analysis. In the analysis, we used presence of DISH as the objective variable (absence = 0, presence = 1), and severity of prevalent LS (KL0, 1 = 0 vs. KL2 = 1; KL0, 1 = 0 vs. KL3 or 4 = 2) and KOA (KL0, 1 = 0 vs. KL2 = 1; KL0, 1 = 0 vs. KL3 or 4 = 2) as explanatory variables, in addition to basic characteristics such as age (+1 year), sex (men = 1, women = 0), BMI (+1 kg/m^2), and regional differences (mountainous area = 0, coastal area = 1). Other potential associated factors were selected with significant or marginal ($p < 0.1$) association with DISH status in a simple linear analysis. The selected explanatory variables for logistic regression analysis are described in the Results section.

Results

Prevalence of DISH was 10.8 % (men: 22.0 %, women: 4.8 %), and was significantly higher in men than in women. Figure 1 shows the prevalence of DISH according to age and sex. Prevalence increased with age in both men and women. Prevalence in subjects classified by age-strata—<50, 50–59, 60–69, 70–79, and ≥80 years—was 1.8, 11.7, 15.4, 32.6, and 39.6 % in men, and 0.7, 1.5, 3.5, 7.6, and 11.8 % in women, respectively.

Table 1 shows the baseline characteristics of the 1,647 participants with and without DISH. In total, subjects with DISH tended to be older, taller, heavier, and have higher BMI than those without DISH ($p < 0.0001$). In the comparison classified by sex, age was significantly higher in those with DISH in both men and women ($p < 0.0001$). In women, mean weight and BMI were significantly higher in those with DISH than in those without DISH (weight: $p < 0.05$, BMI: $p < 0.0001$).

Prevalence of DISH was lower in individuals residing in a coastal area. Individuals with DISH had a higher frequency of smoking and alcohol consumption ($p < 0.05$). The difference in the residing area was significantly observed in men. However, in the comparison classified by sex, differences in smoking and drinking were diluted (Table 1).

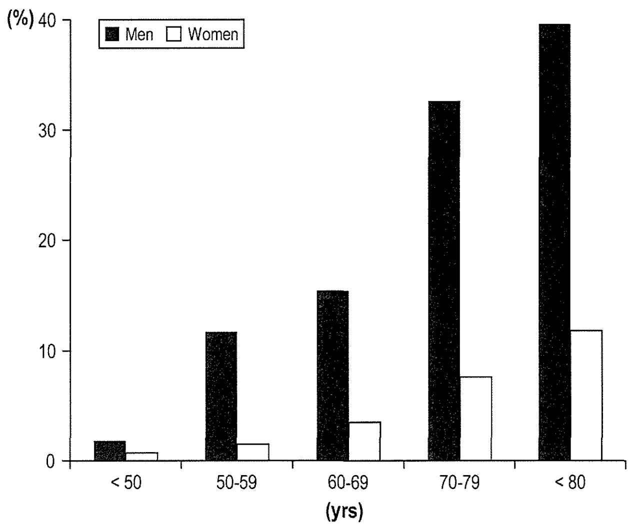

Fig. 1 Prevalence of diffuse idiopathic skeletal hyperostosis (DISH) according to sex and age

Table 1 Mean values (standard deviations) of the anthropometric measurements and the prevalence of lifestyle factors for the participants classified by presence or absence of DISH

	Total ($n = 1647$)			Men ($n = 573$)			Women ($n = 1074$)		
	DISH (−) $n = 1470$	DISH (+) $n = 177$	p	DISH (−) $n = 447$	DISH (+) $n = 126$	p	DISH (−) $n = 1023$	DISH (+) $n = 51$	p
Age (years)	64.4 (12.1)	72.3 (8.4)	<0.0001***	64.6 (12.1)	72.4 (8.2)	<0.0001***	64.3 (12.2)	71.9 (8.8)	<0.0001***
Height (cm)	154.7 (9.2)	158.6 (8.8)	<0.0001***	163.7 (7.3)	162.5 (6.7)	0.0918	150.8 (7.0)	148.9 (5.5)	0.0589
Weight (kg)	55.9 (10.6)	60.1 (10.5)	<0.0001***	62.3 (11.0)	62.1 (10.0)	0.8806	51.9 (8.8)	55.0 (10.3)	0.0126*
BMI (kg/m^2)	22.9 (3.4)	23.8 (3.3)	0.0005***	23.2 (3.2)	23.5 (2.9)	0.3378	22.8 (3.4)	24.7 (3.9)	0.0001***
Residing in the coastal area (%)	50.48	40.11	0.009**	50.3	35.7	0.004**	50.5	51.0	0.951
Current smoking habit (regularly, ≥1 month) (%)	11.9	21.3	<0.001***	29.9	29.0	0.858	3.8	2.0	0.506
Current alcohol consumption (regularly, ≥1 month) (%)	38.7	48.0	0.017*	68.5	61.1	0.122	25.7	15.7	0.108
Presence of LS (KL grade ≥2) (%)	59.1	93.8	<0.001***	72.0	94.4	<0.001***	53.4	92.2	<0.001***
Presence of LS (KL grade ≥3) (%)	35.6	48.0	0.001**	35.4	45.2	0.043*	35.7	54.9	0.005**
Presence of KOA (KL grade ≥2) (%)	48.2	65.5	<0.001***	35.5	58.7	<0.001***	53.8	83.3	<0.001***
Presence of KOA (KL grade ≥3) (%)	18.4	34.5	<0.001***	11.0	27.0	<0.001***	21.7	54.2	<0.001***

DISH diffuse idiopathic skeletal hyperostosis, *BMI* body mass index, *LS* lumbar spondylosis, *KOA* knee osteoarthritis, *KL grade* Kellgren-Lawrence grade

DISH (−) absence of DISH, *DISH (+)* presence of DISH

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

Table 1 also shows the prevalence of LS and KOA defined by KL grade ≥2 and grade ≥3, according to DISH status. In total, the prevalence of LS was higher in those with DISH than in those without DISH ($p = 0.001$). A similar tendency was observed in the prevalence of KOA ($p < 0.001$). This tendency also was noted in the comparison classified by sex.

We classified subjects with DISH into 4 types: (1) cervical, ossification along the lateral aspect of at least 4 contiguous vertebral bodies only in the cervical region (C0/1–C6/7); (2) thoracic, ossification along the lateral aspect of at least 4 contiguous vertebral bodies only in the thoracic region (Th4/5–Th12/L1); (3) lumbar, ossification along the lateral aspect of at least 4 contiguous vertebral bodies only in the lumbar region (L1/2–L5/S1); and (4) diffuse, ossification along the lateral aspect of at least 4 contiguous vertebral bodies in more than 2 regions or through more than 2 regions. Table 2 shows the prevalence of DISH classified by location in the spine. A total of 89 % was shown to be thoracic, whereas the remaining was diffuse; there were no subjects with cervical-type or lumbar-type DISH.

Figure 2 shows the distribution of DISH classified by vertebral level (Th4/5–LS/S1). Among diffuse-type DISH, although 2 subjects had ossification in the cervical region, the cervical site is excluded from the figure. Figure 2 shows that ossification was observed mainly in the middle-lower thoracic sites (Th7/8–Th9/10).

Logistic regression analysis was performed with DISH as the objective variable, LS and KOA as explanatory variables, and patient characteristics including age, sex, BMI, regional differences, smoking, and alcohol consumption as potential risk factors. Presence of DISH was significantly associated with presence of LS (KL2 vs KL0: 1, KL ≥3 vs KL0: 1) and KOA (KL ≥3 vs KL0: 1). Among other potential associated factors, older age, male sex, and higher BMI remained as significantly associated with the presence of DISH (Table 3).

Table 2 Number (proportion, %) of DISH (+) patients classified by spinal ossification site

Type of DISH	Total	Men	Women
Cervical type	0 (0.0 %)	0 (0.0 %)	0 (0.0 %)
Thoracic type	157 (88.7 %)	111 (88.1 %)	46 (90.2 %)
Lumbar type	0 (0.0 %)	0 (0.0 %)	0 (0.0 %)
Diffuse type	20 (11.3 %)	15 (11.9 %)	5 (9.8 %)
Total	177 (100.0 %)	126 (100.0 %)	51 (100.0 %)

Cervical type: Ossification along the lateral aspect of at least four contiguous vertebral bodies existing only in the cervical region (C0/1–C6/7)

Thoracic type: Ossification along the lateral aspect of at least four contiguous vertebral bodies existing only in the thoracic region (Th4/5–Th12/L1)

Lumbar type: Ossification along the lateral aspect of at least four contiguous vertebral bodies existing only in the lumbar region (L1/2–L5/S1)

Diffuse type: Ossification along the lateral aspect of at least four contiguous vertebral bodies existing in more than 2 regions or through more than 2 regions

Finally, to clarify the association of DISH with LS and KOA, we performed logistic regression analysis using DISH as an objective variable, LS and KOA as explanatory variables, and patient characteristics including age, sex, BMI, regional differences, smoking, and alcohol consumption as potential risk factors. Presence of DISH was significantly associated with presence of LS (KL2 vs KL0: 1, KL ≥3 vs KL0: 1) and KOA (KL ≥3 vs KL0: 1) independently (Table 4).

Discussion

In the present study, using lateral whole-spine radiographs of recently examined population-based samples, we estimated that the prevalence of DISH was one-tenth of the population, which consisted of participants from the ROAD study. The subjects with DISH tended to be older and had bigger body build than those without DISH. In addition, DISH was observed more frequently in men than

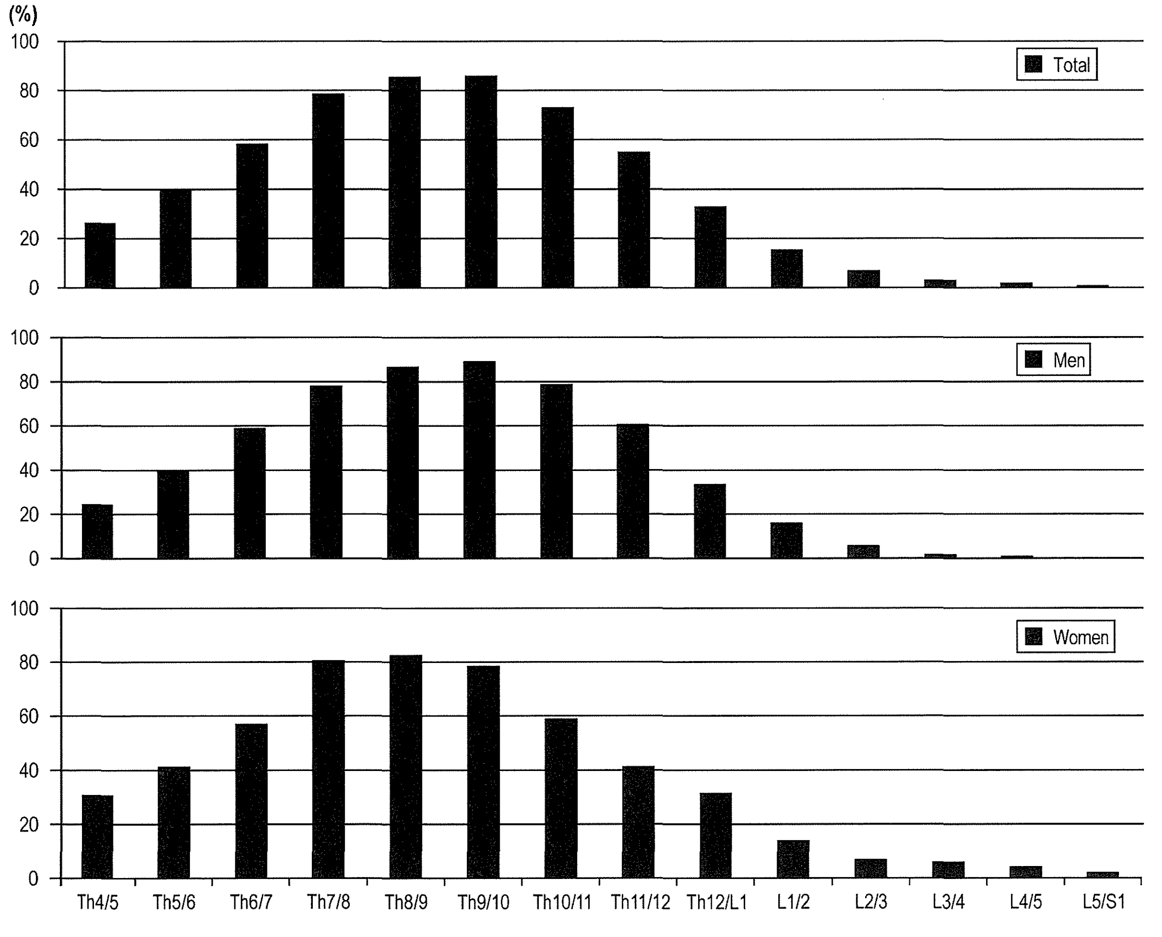

Fig. 2 Prevalence of diffuse idiopathic skeletal hyperostosis (DISH) in each vertebral level, classified by sex

Table 3 Odds ratios of lumbar spondylosis or knee osteoarthritis, and potentially associated factors for the presence of DISH vs. absence of DISH

Explanatory variables	Category	OR	95 % CI	p
Lumbar spondylosis				
Presence of LS	0: KL grade = 0, 1, 1: KL grade = 2	5.80	2.97–11.3	<0.001***
	0: KL grade = 0, 1, 2: KL grade ≥3	4.54	2.34–8.84	<0.001***
Age (years)	+1 year	1.07	1.05–1.09	<0.001***
Gender	1: men, 0: women	4.61	3.05–6.99	<0.001***
Region	0: mountainous area, 1: coastal area	0.88	0.61–1.26	0.475
BMI (kg/m^2)	+1 kg/m^2	1.11	1.05–1.17	<0.001***
Smoking	0: ex or never smoker, 1: current smoker	1.65	1.04–2.63	0.034*
Alcohol consumption	0: ex or never drinker, 1: current drinker	0.82	0.56–1.22	0.329
Knee osteoarthritis				
Presence of KOA	0: KL grade = 0, 1, 1: KL grade = 2	1.34	0.85–2.10	0.211
	0: KL grade = 0, 1, 2: KL grade ≥3	2.15	1.32–3.52	0.002**
Age (years)	+1 year	1.07	1.04–1.09	<0.001***
Gender	1: men, 0: women	6.90	4.48–10.6	<0.001***
Region	0: mountainous area, 1: coastal area	0.95	0.65–1.37	0.771
BMI (kg/m^2)	+1 kg/m^2	1.09	1.03–1.15	0.002**
Smoking	0: ex or never smoker, 1: current smoker	1.52	0.95–2.42	0.079
Alcohol consumption	0: ex or never drinker, 1: current drinker	0.85	0.58–1.26	0.431

DISH diffuse idiopathic skeletal hyperostosis, *BMI* body mass index, *LS* lumbar spondylosis, *KOA* knee osteoarthritis, *KL grade* Kellgren-Lawrence grade

OR odds ratios, *95 % CI* 95 % confidence interval

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

Table 4 Odds ratios of lumbar spondylosis and knee osteoarthritis, and potentially associated factors for the presence of DISH vs. absence of DISH

Explanatory variables	Category	OR	95 % CI	p
Presence of LS (KL grade = 2)	vs. KL grade = 0, 1	5.50	2.81–10.8	<0.001***
Presence of LS (KL grade ≥3)	vs. KL grade = 0, 1	4.09	2.08–8.03	<0.001***
Presence of KOA (KL grade = 2)	vs. KL grade = 0, 1	1.22	0.77–1.92	0.404
Presence of KOA (KL grade≥ 3)	vs. KL grade = 0, 1	1.89	1.14–3.10	0.013**
Age (years)	+1 year	1.06	1.03–1.14	<0.001***
Gender	1: men, 0: women	5.55	3.57–8.63	<0.001***
Region	0: mountainous area, 1: coastal area	0.88	0.60–1.29	0.522
BMI (kg/m^2)	+1 kg/m^2	1.08	1.02–1.14	0.008**
Smoking	0: ex or never smoker, 1: current smoker	1.59	1.00–2.55	0.052
Alcohol consumption	0: ex or never drinker, 1: current drinker	0.81	0.54–1.21	0.298

DISH diffuse idiopathic skeletal hyperostosis, *BMI* body mass index, *LS* lumbar spondylosis, *KOA* knee osteoarthritis, *KL grade* Kellgren-Lawrence grade

OR odds ratios, *95 % CI* 95 % confidence interval

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

in women, and the most common site was the thoracic vertebrae. Presence of DISH was significantly associated with the presence of KOA and LS, after adjusting for potential associated factors.

There have been several epidemiologic studies on DISH in many parts of the world [12–19]. The results indicate that DISH is observed mainly in men and the elderly; prevalence increases with age, and it is distributed mostly in the thoracic spine. These results are supported by the results of the present study. However, there are considerable differences in the prevalence. Weinfeld et al. [20] reported that genetic or hereditary differences are important predisposing factors for DISH. Their previous study involved patients from ethnic populations, including 667 white, 144 black, 72 Native American, 11 Hispanic, and 30 Asian patients. They showed that the Asian, black, and Native American populations had a remarkably lower prevalence of DISH; however, their study population was small. In a recent study, Kim et al. [18] reported that race influences the prevalence of DISH. Their prevalence of DISH was 5.4 % in men and 0.8 % in women aged over 80 years in a Korean population, which is remarkably lower than the prevalence in our study, despite the similar race. Our prevalence was similarly high as the white population in Weinfield's report. Therefore, it is believed that genetic factors influence the prevalence of DISH more than race.

The present study clarified that most cases of DISH were observed in the thoracic vertebrae. There were no cases of DISH located in only the cervical or lumbar region. All cases of DISH in the cervical region were categorised as diffuse-type. Even if subjects were categorised into diffuse-type DISH, thoracic vertebrae were found to be the most affected. In addition, among the thoracic vertebrae, we found the predilection site to be the middle thoracic vertebrae (Th7–Th9). Holton et al. [27] reported that the distribution of the lowest level of DISH in 298 male subjects aged $\geq$65 years was 38 % in the thoracic region, 49 % in the thoracolumbar region, and 13 % in the lumbar region. It is interesting that DISH has predilection sites, which might be due to anatomic alignment of the vertebrae. For example, the middle thoracic vertebrae are likely to be affected by compressive mechanical stress because the Th8 is located nearly at the top in physiologic kyphosis. DISH originates mainly from the thoracic spine and extends to the cervical and/or lumbar spine by mechanical stress. In the present cross-sectional study, we could not evaluate whether DISH tends to occur in the thoracic vertebrae and then forms in the lumbar spine secondarily; however, we were able to follow-up on the ROAD study and clarify the disease course of thoracic DISH.

Regarding the definition of DISH, it might be easy to imagine that LS, defined by KL2 (defined as radiographically definite osteophytes), is associated with DISH. However, there are few reports to confirm the association between DISH and severe LS with the criterion of KL3 or 4. In the present study, we confirmed the significant association between DISH and LS, not only with the criterion of KL2, but also with KL $\geq$3. In addition, there are few reports to clarify the association between DISH and OA of other sites. In the present study, we also confirmed the significant association between DISH and KOA. In fact, the OR of the presence of DISH for KOA significantly increased according to the severity of KOA. The effects of LS and KOA coexisted independently. This result suggests that DISH and OA might be in a similar vein of disease, for example, the so-called 'bone proliferative group'. There have been several reports regarding the association between DISH and OPLL [4–7]. Resnick et al. [4] described 4 patients with coexisting DISH and cervical OPLL, and found OPLL in 50 % of 74 additional patients with DISH after reviewing their cervical spine radiographs. However, there has been no report on the association of DISH and OA; thus the etiology of ossification might not be similar to that of OA. Therefore, with only the results of the present study, we cannot definitely claim that DISH and OA are in a similar disease group, even though DISH tends to have similar associated factors, such as age, overweight (bigger BMI), and mechanical stress, as OA.

Another hypothesis is that there might be hidden associated factors that might affect both DISH and OA. We considered risk factors for metabolic syndrome as potential confounders. Several constitutional and metabolic abnormalities have been reported to be associated with DISH including obesity, large waist circumference, hypertension, diabetes mellitus, hyperinsulinemia, dyslipidemia, and hyperuricemia [21, 28–30]. In addition, both LS and KOA are well known to be associated with obesity [31]. We have already reported on the presence of hypertension and impaired glucose tolerance, and shown that the accumulation of metabolic risk factors is associated with the presence and occurrence of KOA [32, 33]. In addition, we found that current smoking, a known risk factor for cardiovascular disease as well as metabolic risk factors, was significantly associated with DISH. These findings may indicate that DISH is a candidate surrogate index for metabolic risk factors as a predictor of OA, or vice versa. We could not evaluate this hypothesis at present, but we would clarify the association including the causal relationships between DISH, OA, and metabolic risk factors in a further study.

Alternatively, we considered associated factors for inflammation or cartilage metabolic turnover as potential confounders between DISH and OA. These factors might coexist as risk factors for DISH and OA. Thus, there might be a direct or indirect pathway between DISH and OA via hidden associated factors, which should be investigated in a further study.

This study has several limitations. First, although the ROAD study includes a large number of participants, these subjects may not truly represent the general population. To address this, we compared the anthropometric measurements and frequencies of smoking and alcohol consumption between study participants and the general Japanese population; no significant differences were found, with the exception that male ROAD study participants aged 70–74 years were significantly smaller in terms of body structure than the overall Japanese population ($p < 0.05$)

[25]. This difference should be considered when evaluating potential risk factors in men aged 70–74 years; factors such as body build, particularly greater weight, are known to be associated with LS and KOA. Therefore, our results may be an underestimation of the prevalence of these conditions. Second, in the present study, we used only the data of the baseline study. Thus, we were not able to confirm a causal relationship between DISH status and other associated factors, as mentioned above. Nevertheless, we have performed a follow-up study, so we will be able to clarify the causal relationship between DISH status and OA in the near future. Third, this study could not evaluate the cervicothoracic junction (C7–Th4) because we assessed only radiographs. Although most cases of DISH existed in the inferior thoracic spine, as Fig. 2 shows, the lack of findings in the C7/C1–Th3/Th4 levels might have underestimated the prevalence of DISH. To evaluate the cervicothoracic junction, it would be necessary to use computed tomography or magnetic resonance imaging of the whole spine, which appeared impossible to perform on more than 1,600 subjects. Fourth, LS defined by KL2 may have been included in cases of DISH, but there is no method to confirm the overlap of the presence of DISH and LS of KL2 using the radiographic diagnostic criteria. DISH is observed mainly in the thoracic region, and only the diffuse type expands partly into the lumbar region. Therefore, there is a small possibility that LS of KL2 might be contaminated into DISH. Finally, in the present study, we could not evaluate other sites of OA besides the knee and lumbar spine, such as the hands or hip. To evaluate DISH and other sites of OA, we should evaluate the presence or occurrence of OA at other sites in a further study.

In conclusion, in the present population-based study, we found that the prevalence of DISH was 10.8 % in the overall population. Prevalence was significantly higher in older subjects, and mainly distributed at the thoracic spine. Logistic regression analysis revealed that the presence of DISH was significantly associated with older age, male sex, higher BMI, and presence of severe KOA.

Acknowledgments This work was supported by Grants-in-Aid for Scientific Research B23390172 and B20390182 to NY, C20591737 to TA, and C20591774 to SM, for Young Scientists A18689031 to HO, and Collaborating Research with NSF 08033011-00262 (Director, NY) from the Ministry of Education, Culture, Sports, Science and Technology; and H17-Men-eki-009 (Director, KN), H18-Choujyu-037 (Director, TN), H20-Choujyu-009 (Director, NY), H23-Chojyu-002 (Director, TA), and H25-Chojyu-007 (Director, NY) from the Ministry of Health, Labour and Welfare in Japan. This study also was supported by grants from the Japan Osteoporosis Society (NY, SM, HO, and TA) and research aid from the Japanese Orthopaedic Association (JOA-Subsidized Science Project Research 2006-1 and 2010-2 [Director, HK]). The authors wish to thank Dr. Takako Nojiri and Mr. Kazuhiro Hatanaka of the Gobo Public Health Centre; Dr. Naoki Hirabayashi of the Kawakami Clinic, Hidakagawa Town; Mrs. Tomoko Takijiri, Mrs. Kumiko Shinou, Mrs. Rie Takiguchi, Mrs. Kyoko Maeda, Ms. Ikuyo Ueyama, Mrs. Michiko Mori, Mrs. Hisayo Sugimoto, and other members of the public office in Hidakagawa Town; Dr. Shinji Matsuda of the Shingu Public Health Centre; and Mrs. Tamako Tsutsumi, Mrs. Kanami Maeda, Mr. Shoichi Shimoichi, Mrs. Megumi Takino, Mrs. Shuko Okada, Mrs. Kazuyo Setoh, Mrs. Chise Ryouno, Mrs. Miki Shimosaki, Mrs. Chika Yamaguchi, Mrs. Yuki Shimoji, and other members of the public office in Taiji Town for their assistance in locating and scheduling participants for examinations. We also thank Ms. Kyoko Yoshimura, Mrs. Toki Sakurai, and Mrs. Saeko Sahara for their assistance with data reduction and administration.

Conflict of interest All authors declare that (1) no author has received corporate support for the submitted work; (2) the authors have no relationships with companies that might have an interest in the submitted work in the previous 3 years; (3) the authors' spouses, partners, or children do not have financial relationships that may be relevant to the submitted work; and (4) the authors have no nonfinancial interests that may be relevant to the submitted work.

References

1. Mader R, Sarzi-Puttini P, Atzeni F, Olivieri I, Pappone N, Verlaan JJ, Buskila D (2009) Extraspinal manifestations of diffuse idiopathic skeletal hyperostosis. Rheumatology (Oxford) 48:1478–1481
2. Resnick D, Niwayama G (1976) Radiographic and pathologic features of spinal involvement in diffuse idiopathic skeletal hyperostosis (DISH). Radiology 119:559–568
3. Mata S, Chhem RK, Fortin PR, Joseph L, Esdaile JM (1998) Comprehensive radiographic evaluation of diffuse idiopathic skeletal hyperostosis: development and interrater reliability of a scoring system. Semin Arthr Rheum 28:88–96
4. Resnick D, Guerra J Jr, Robinson CA, Vint VC (1978) Association of diffuse idiopathic skeletal hyperostosis (DISH) and calcification and ossification of the posterior longitudinal ligament. AJR 131:1049–1053
5. Ono M, Russell WJ, Kudo S, Kuroiwa Y, Takamori M, Motomura S, Murakami J (1982) Ossification of the thoracic posterior longitudinal ligament in a fixed population. Radiological and neurological manifestations. Radiology 143:469–474
6. Ehara S, Shimamura T, Nakamura R, Yamazaki K (1998) Paravertebral ligamentous ossification: DISH, OPLL and OLF. Eur J Radiol 27:196–205
7. Yoshimura N, Nagata K, Muraki S, Oka H, Yoshida M, Enyo Y, Kagotani R, Hashizume H, Yamada H, Ishimoto Y, Teraguchi M, Tanaka S, Kawaguchi H, Toyama Y, Nakamura K, Akune T (2013) Prevalence and progression of radiographic ossification of the posterior longitudinal ligament and its associated factors in the Japanese population: a 3-year follow-up of the ROAD study. Osteoporos Int [E-pub ahead of print]
8. Hannallah D, White AP, Goldberg G (2007) Diffuse idiopathic skeletal hyperostosis. Oper Tech Orthop 17:174–177
9. Albert TJ, Mader R (2002) Clinical manifestations of diffuse idiopathic skeletal hyperostosis of the cervical spine. Semin Arthr Rheum 32:130–135
10. Meyer PR Jr (1999) Diffuse idiopathic skeletal hyperostosis in the cervical spine. Clin Orthop Relat Res 359:49–57
11. Westerveld LA, Verlaan JJ, Oner FC (2009) Spinal fractures in patients with ankylosing spinal disorders: a systematic review of the literature on treatment, neurological status and complications. Eur Spine J 18:145–156
12. Hendrix RW, Melany M, Miller F, Rogers LF (1994) Fracture of the spine in patients with ankylosis due to diffuse skeletal hyperostosis: clinical and imaging findings. AJR 162:899–904

13. Julkunen H, Heinonen OP, Knekt P, Maatela J (1975) The epidemiology of hyperostosis of the spine together with its symptoms and related mortality in a general population. Scand J Rheumatol 4:23–27
14. Tsukamoto Y, Onitsuka H, Lee K (1977) Radiological aspects of DISH in the spine. AJR 129:913–918
15. Bloom RA (1984) The prevalence of ankylosing hyperostosis in a Jerusalem population–with description of a method of grading the extent of the disease. Scand J Rheumatol 13:181–189
16. Cassim B, Mody G, Rubin D (1990) The prevalence of diffuse idiopathic skeletal hyperostosis in African blacks. Br J Rheumatol 29:131–132
17. Kiss C, O'Neill TW, Mituszova M, Szilagyi M, Donath J, Poor GY (2002) Prevalence of diffuse idiopathic skeletal hyperostosis in Budapest, Hungary. Rheumatology 41:1335–1336
18. Kim SK, Choi BR, Kim CG, Chung SH, Choe JY, Joo KB, Bae SC, Yoo DH, Jun JB (2004) The prevalence of diffuse idiopathic skeletal hyperostosis in Korea. J Rheumatol 31:2032–2035
19. Kiss C, O'Neill TW, Mituszova M, Szilagyi M, Poor G (2002) The prevalence of diffuse idiopathic skeletal hyperostosis in a population-based study in Hungary. Scand J Rheumatol 31:226–229
20. Weinfeld RM, Olson PN, Maki DD, Griffiths HJ (1997) The prevalence of diffuse idiopathic skeletal hyperostosis (DISH) in two large American Midwest metropolitan hospital populations. Skeletal Radiol 26:222–225
21. Kiss C, Szilagyi M, Paksy A, Poor G (2002) Risk factors for diffuse idiopathic skeletal hyperostosis: a case control study. Rheumatology (Oxford) 41:27–30
22. Mader R, Novofestovski I, Adawi M, Lavi I (2009) Metabolic syndrome and cardiovascular risk in patients with diffuse idiopathic skeletal hyperostosis. Semin Arthr Rheum 38:361–365
23. Kellgren JH, Lawrence LS (1957) Radiological assessment of osteoarthrosis. Ann Rheum Dis 16:494–502
24. Yoshimura N, Muraki S, Oka H, Kawaguchi H, Nakamura K, Akune T (2010) Cohort profile: research on osteoarthritis/osteoporosis against disability (ROAD) study. Int J Epidemiol 39:988–995
25. Yoshimura N, Muraki S, Oka H, Mabuchi A, En-yo Y, Yoshida M, Saika A, Yoshida H, Suzuki T, Yamamoto S, Ishibashi H, Kawaguchi H, Nakamura K, Akune T (2009) Prevalence of knee osteoarthritis, lumbar spondylosis, and osteoporosis in Japanese men and women: the research on osteoarthritis/osteoporosis against disability study. J Bone Miner Metab 27:620–628
26. Orwoll E, Blank JB, Barrett-Connor E, Cauley J, Cummings S, Ensrud K, Lewis C, Cawthon PM, Marcus R, Marshall LM, McGowan J, Phipps K, Sherman S, Stefanick ML, Stone K (2005) Design and baseline characteristics of the osteoporotic fractures in men (MrOS) study–a large observational study of the determinants of fracture in older men. Contemp Clin Trials 26:569–585
27. Holton KF, Denard PJ, Yoo JU, Kado DM, Barrett-Connor E, Marshall LM, Osteoporotic Fractures in Men (MrOS) Study Group (2011) Diffuse idiopathic skeletal hyperostosis and its relation to back pain among older men: the MrOS Study. Semin Arthr Rheum 41:131–138
28. Littlejohn GO (1985) Insulin and new bone formation in diffuse idiopathic skeletal hyperostosis. Clin Rheumatol 4:294–300
29. Denko CW, Boja B, Moskowitz RW (1994) Growth promoting peptides in osteoarthritis and diffuse idiopathic skeletal hyperostosis–insulin, insulin-like growth factor-I, growth hormone. J Rheumatol 21:1725–1730
30. Vezyroglou G, Mitropoulos A, Kyriazis N, Antoniadis C (1996) A metabolic syndrome in diffuse idiopathic skeletal hyperostosis: a controlled study. J Rheumatol 23:672–676
31. Muraki S, Akune T, Oka H, Mabuchi A, En-yo Y, Yoshida M, Saika A, Nakamura K, Kawaguchi H, Yoshimura N (2009) Association of occupational activity with radiographic knee osteoarthritis and lumbar spondylosis in elderly patients of population-based cohorts: a large-scale population-based study. Arthr Care Res 61:779–786
32. Yoshimura N, Muraki S, Oka H, Kawaguchi H, Nakamura K, Akune T (2011) Association of knee osteoarthritis with the accumulation of metabolic risk factors such as overweight, hypertension, dyslipidemia, and impaired glucose tolerance in Japanese men and women: the ROAD study. J Rheumtol 38:921–930
33. Yoshimura N, Muraki S, Oka H, Tanaka S, Kawaguchi H, Nakamura K, Akune T (2012) Accumulation of metabolic risk factors such as overweight, hypertension, dyslipidemia, and impaired glucose tolerance raises the risk of occurrence and progression of knee osteoarthritis: a 3-year follow-up of the ROAD study. Osteoarthr Cartil 20:1217–1226

Gradual spinal cord decompression through migration of floated plaques after anterior decompression via a posterolateral approach for OPLL in the thoracic spine

Satoshi Kato, MD, Hideki Murakami, MD, Satoru Demura, MD, Katsuhito Yoshioka, MD, Hiroyuki Hayashi, MD, Noriaki Yokogawa, MD, Xiang Fang, MD, and Hiroyuki Tsuchiya, MD

Department of Orthopaedic Surgery, Kanazawa University School of Medicine, Kanazawa, Japan

OBJECT Several surgical procedures have been developed to treat thoracic OPLL (ossification of the posterior longitudinal ligament). However, favorable surgical results are not always achieved, and consistent protocols and procedures for surgical treatment of OPLL in this region have not been established. Beak-type OPLL in the thoracic spine is known to be the most complicated form of OPLL to treat surgically. In this study, the authors examine the clinical outcomes after anterior decompression via a posterolateral approach for beak-type OPLL in the thoracic spine and address the gradual spinal cord decompression caused by migration of the floated plaques after surgery.

METHODS Between 2011 and 2013, a total of 12 patients with thoracic myelopathy due to OPLL were surgically treated at the authors' institute. The study group for this paper comprises 6 of those 12 patients. These 6 patients, who had beak-type OPLL, underwent with anterior decompression and instrumented fusion via the authors' posterolateral approach–based surgical technique. The other 6 patients, who exhibited other types of OPLL, underwent posterior decompression and instrumented fusion. In the study group (the 6 patients with beak-type OPLL), half of the patients (the 3 patients who were treated first) were treated with removal of the ossified ligament. These patients are referred to as the removal group. The other 3 patients were treated by means of "floating" the OPLL plaques and are referred to as the floating group. Clinical and radiographic outcomes were evaluated in these 6 cases.

RESULTS The recovery rates were 52.4% in the removal group and 60.0% in the floating group. Two patients in the removal group had operative complications, including a dural tear and temporary neurological deterioration. No operative complications were encountered in the floating group. In all 3 cases in the floating group, floating of the ossified ligament was completely achieved, and the floated plaque gradually migrated into the ventral bone resection areas. The mean migration distances of the floated plaque were 2.4 mm, 4.3 mm, 4.7 mm, and 4.8 mm at 1, 3, 6, and 12 months after surgery.

CONCLUSIONS Treatment of beak-type OPLL in the thoracic spine via the posterolateral approach–based floating plaque technique was safe and effective in this small case series. Gradual migration of the floated plaques provided additional spinal cord decompression during the postoperative course.

http://thejns.org/doi/abs/10.3171/2015.1.SPINE14960

KEY WORDS ossification of the posterior longitudinal ligament; thoracic spine; anterior decompression; posterolateral approach; gradual spinal cord decompression; migrating floated plaques

Thoracic myelopathy consequent to ossification of the posterior longitudinal ligament (OPLL) in the thoracic spine is usually progressive and responds poorly to conservative therapy, leaving surgery as the only effective treatment option. Despite advances in surgical techniques employed for thoracic OPLL, favorable results are not always achieved.[6] In patients with thoracic myelopathy resulting from OPLL in the thoracic spine, anterior decompression via removal or floating of the ossified PLL (or "plaque") is the most effective method of relieving pressure on the spinal cord.[1,3,5,11–13] However, anterior spinal cord decompression is technically demanding and has been reported to cause postoperative neurological degradation.[1,8]

We developed a novel technique to remove or float the ossified PLL in the thoracic spine via a posterolateral ap-

ABBREVIATIONS JOA = Japanese Orthopaedic Association; OPLL = ossification of the PLL; PLL = posterior longitudinal ligament.

SUBMITTED September 19, 2014. **ACCEPTED** January 13, 2015.
INCLUDE WHEN CITING Published online July 3, 2015; DOI: 10.3171/2015.1.SPINE14960.
DISCLOSURE This work was supported by a Health Labour Sciences Research Grant. No benefits in any form have been or will be received from any commercial party related directly or indirectly to the subject of this article.

proach.[4] This procedure allows the surgeon to perform anterior decompression for the treatment of thoracic OPLL more safely and effectively than conventional procedures. This approach is especially useful for cases of beak-type OPLL, which is known to be the most complicated surgically.[7,10] In our clinical cases in which the ossified PLL was floated during the procedure, we observed gradual spinal cord decompression caused by migration of the floated plaques after surgery (Figs. 1 and 2). The purpose of this study was to address this phenomenon and evaluate the clinical outcomes after anterior decompression via our procedure.

Methods

Patient Selection

Between 2011 and 2013, a total of 12 patients with thoracic myelopathy due to OPLL were surgically treated at our institute. Six patients with beak-type OPLL in the thoracic spine underwent anterior decompression and instrumented fusion via a posterolateral approach. The other 6 patients, who exhibited other types of OPLL, underwent posterior decompression and instrumented fusion. Among the 6 patients who underwent anterior decompression, removal of the ossified PLL was performed in the 3 patients who were treated first and floating was performed in the later 3 patients. In this study, the clinical and radiographic outcomes were primarily examined in patients who were treated with the plaque-floating technique.

Clinical and Radiographic Parameters

The surgical outcomes were assessed according to the Japanese Orthopaedic Association (JOA) score for thoracic myelopathy (total of 11 points), which was derived from the JOA scoring system for cervical myelopathy after eliminating the motor and sensory scores for the upper extremities,[9] and the Hirabayashi recovery rate.[2] JOA scores were recorded both before surgery and at their maximum point after surgery. The Hirabayashi recovery rate (%) was calculated using the following formula: (postoperative JOA score − preoperative JOA score)/(11 − preoperative JOA score) × 100.

The OPLL occupying ratio in the canal diameter and the migration distances of the floated plaque at 1, 3, 6, and 12 months after surgery were measured using multiplanar reconstruction CT (Fig. 3). The anterior decompression ratio (%) was defined and calculated using the following formula: (migration distance of the floated plaque/maximum sagittal length of the plaque) × 100 (%).

Surgical Procedure

With the patient in a prone position, we performed a total resection of the posterior vertebral elements at the anterior decompression levels. This maneuver included not only laminectomy but also removal of the transverse processes and pedicles, thus allowed the creation of space bilaterally at the sides of the dural sac for the subsequent anterior decompression. The thoracic nerves at the anterior decompression levels were ligated bilaterally and lifted to improve the viewing of the ossified ligament and the anterolateral aspect of the dural sac. An anterior decompression was then performed posteriorly. Using these surgical maneuvers, the ossification was floated without difficulty (Fig. 4). Removal of beak-type ossification was also achieved using this technique in the earlier 3 patients with exfoliation of adhesions between ossified ligament and the ventral aspect of the dural sac after floating of plaque.[4] In every step of the anterior decompression, the space created at the bilateral sides of the dural sac and the view created by lifting up of the ligated nerve roots allowed us to see the ossified PLL and the anterolateral aspect of the dural sac directly and easily and use a bur safely (Fig. 4). We placed posterior pedicle screw instrumentation and performed posterolateral fusion using local bone

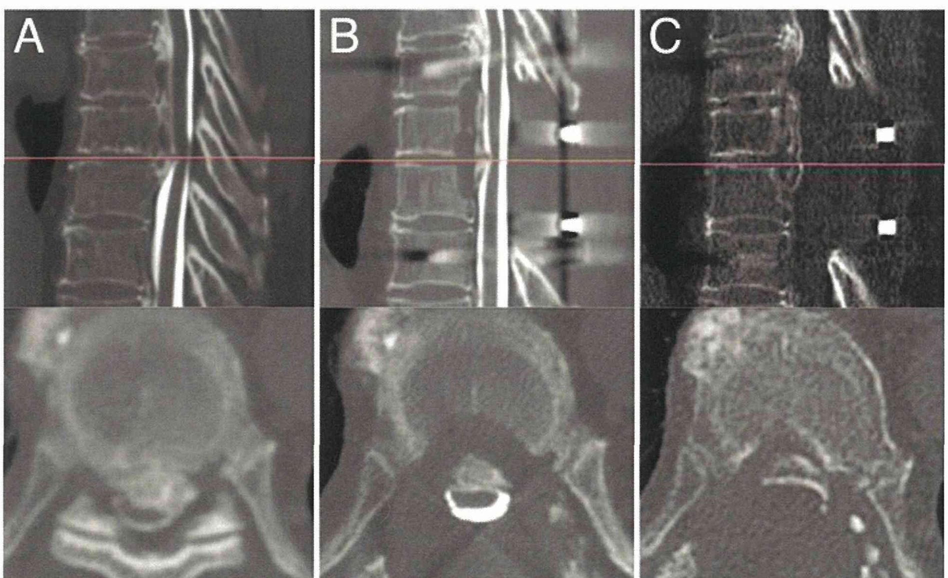

FIG. 1. Case 4. OPLL at T8–9 treated by means of the plaque-floating technique. **A and B:** Preoperative (A) and 1-month postoperative (B) CT myelograms. **C:** Postoperative plain CT image obtained 6 months after surgery. Figure is available in color online only.

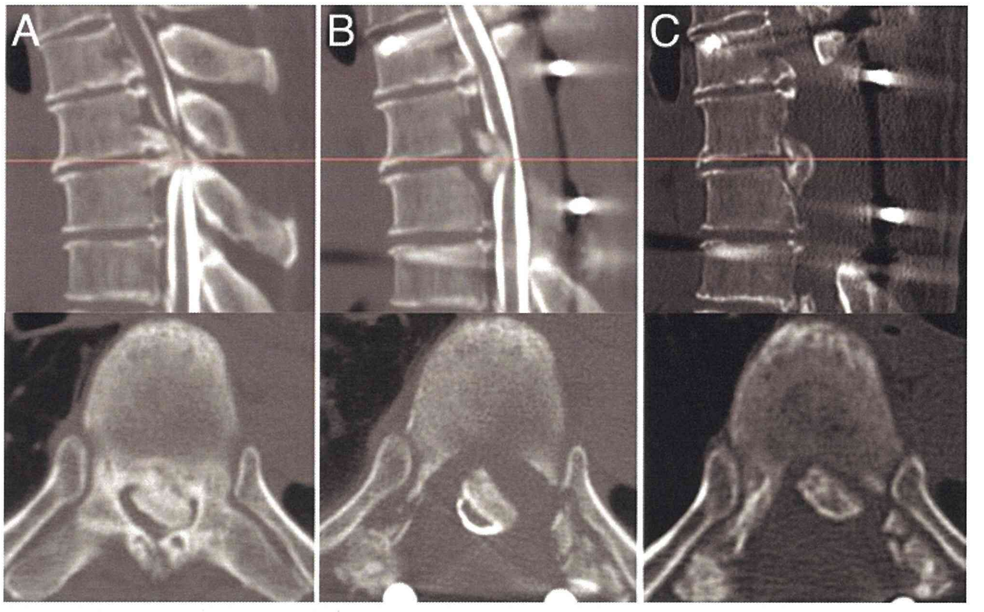

FIG. 2. Case 5. OPLL at T5–6 treated by means of the plaque-floating technique. **A and B:** Preoperative (A) and 1-month postoperative (B) CT myelograms. **C:** Postoperative plain CT image obtained 6 months after surgery. Figure is available in color online only.

chips obtained from the resected laminae and transverse processes. In the 3 patients who were treated with plaque floating (that is, those who were treated later in our experience), kyphosis correction with posterior instrumentation was applied after floating because we observed with ultrasonography that the floated plaque was still causing some spinal cord compression. Our technique allows easier kyphosis correction because the thoracic spine at the decompressed levels becomes more flexible after removal of the transverse processes and pedicles than it is when treated via the conventional posterior approach. The advantages and pitfalls of this surgical technique have been described in detail elsewhere.[4]

Results

Clinical Outcomes

We used this surgical technique to treat 6 patients with beak-type OPLL in the middle thoracic spine. The complete removal of the ossification at the anterior decompression levels was achieved in 3 patients. Adequate plaque floating was achieved in the other 3 patients. Anterior decompression was performed at 2 consecutive vertebral levels in 5 patients (Cases 1, 2, 3, 5, and 6 in Table 1) and at 3 consecutive vertebral levels in the remaining patient (Case 4). In all 6 patients, 2 pairs of thoracic nerve roots were sacrificed. There was no significant girdle pain requiring medication after surgery. A temporary mild neurological deterioration, evident in muscle weakness of the lower extremities and resolving within 1 month after surgery, and a minor dural tear occurred in 2 of the patients who underwent complete removal of the ossified PLL (Cases 2 and 3, respectively). No significant complications such as dural tear or neurological deterioration were encountered in the patients who underwent plaque floating. The recovery rates were 52.4% in the removal group and 60.0% in the floating group (Table 1).

Outcomes of the Radiographic Parameters

The mean OPLL occupying ratio in the floating group was 85.3%. In all 3 floating group patients, a minimum concentric bone resection was observed in the posterior portion of the vertebral body, and the floated plaques gradually migrated into the ventral bone resection areas (Figs. 1 and 2). The mean floated plaque migration distances were 2.4 mm, 4.3 mm, 4.7 mm, and 4.8 mm at 1, 3, 6, and 12 months after surgery. In all 3 patients, bony fusion between the floated plaque and the vertebral body was observed via CT at 12 months after surgery. The mean anterior decompression ratios were 23.5%, 42.2%, 45.5%, and 46.8% at 1, 3, 6, and 12 months after surgery (Table 2).

Discussion

The outcomes of surgical treatment of thoracic OPLL compare unfavorably with those of cervical OPLL.[6] The thoracic spine is naturally kyphotic, and posterior decompression is thus less effective because the backward movement of the spinal cord is restricted. Obviously, anterior spinal cord decompression is ideal for thoracic OPLL if it can be achieved safely and effectively. In all patients in this series, this procedure was used to sufficiently float the plaques of beak-type OPLL without difficulty. A main advantage of this procedure is that it provides more space at the bilateral sides of the dural sac for maneuvering in the anterior decompression relative to the conventional posterior approach. This space in combination with lifting of the ligated thoracic nerves also allows surgeons to directly visualize the OPLL and the anterolateral aspect of the dural sac. In this procedure, surgeons can perform anterior decompression with adequate recognition of the position between the OPLL and the whole dural sac, and this information facilitates the anterior decompression procedure. This advantage is not afforded during anterior decompression via an anterior approach. We therefore

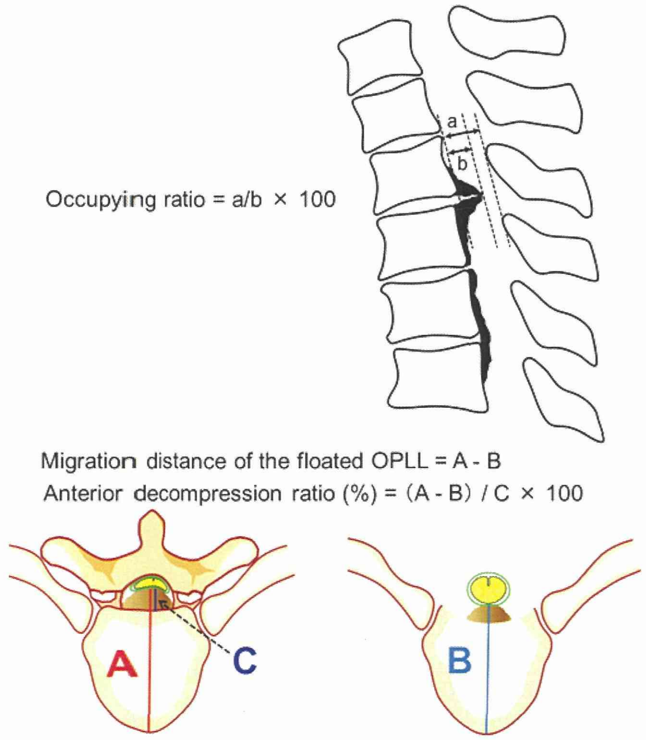

FIG. 3. Radiographic parameters used in this study. **Upper:** Schematic showing the method of calculating the occupying ratio. The canal diameter *(a)* is divided by the maximum sagittal length of the ossified PLL *(b)*. **Lower:** Schematics showing the method of calculating the migration distance of the floated plaque (in millimeters) and the anterior decompression ratio as a percentage ([(A – B)/C] × 100). *A* is the length from the front edge of a vertebral body to the top of an ossified PLL in a presurgical axial CT image; *B*, the distance from the front edge of a vertebral body to the top of an ossified PLL in a postsurgical axial CT image; and *C*, the length of the ossified PLL. *Copyright Satoshi Kato. Published with permission. Figure is available in color online only.*

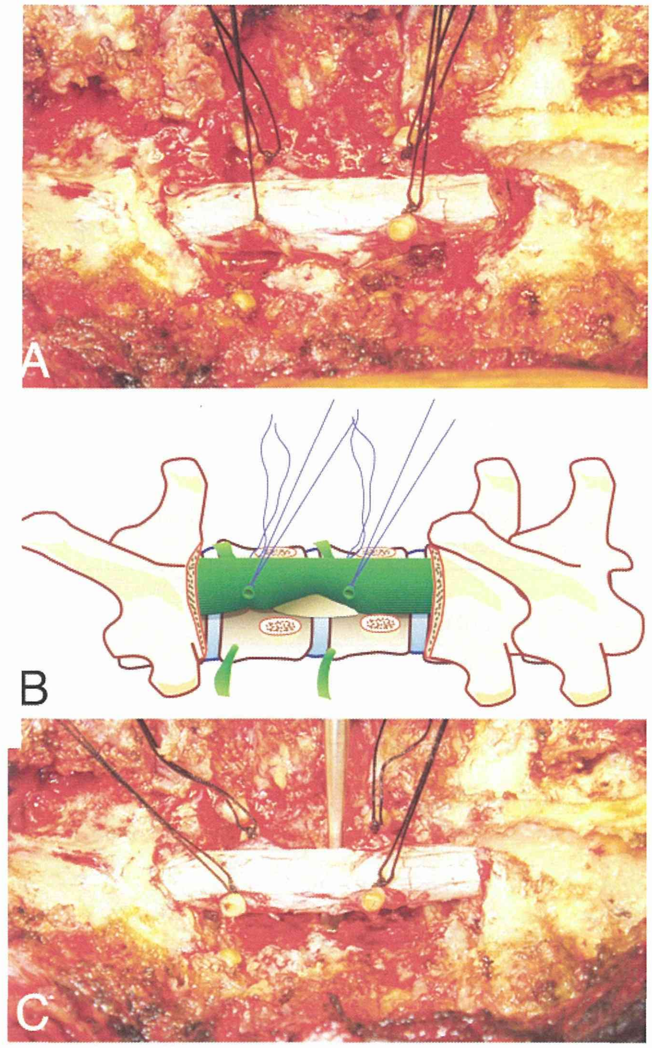

FIG. 4. Case 4. Intraoperative images. **A:** Photograph taken before anterior decompression. **B:** Illustration of intraoperative view before anterior decompression. Copyright Satoshi Kato. Published with permission. **C:** Photograph taken after anterior decompression (floating of ossified PLL). Figure is available in color online only.

consider that our approach is the safest and most feasible surgical procedure for floating the plaque in patients with beak-type OPLL.

On the other hand, the ossified ligament tends to adhere strongly to the ventral aspect of the dural sac, thus increasing the difficulty of direct removal and the risk of spinal cord injury. Considerable improvements in myelopathy were observed during the postoperative course in all patients who underwent anterior decompression via the procedure described in this study. However, 2 of the 3 patients who underwent removal of the floated plaque experienced significant complications, including a dural tear and temporary neurological deterioration. In this study, the final recovery rate of patients who underwent plaque floating was equivalent to that of patients who underwent plaque removal, and the former had no significant intraoperative or postoperative complications. This result demonstrates that plaque floating via this procedure is sufficiently effective for anterior spinal cord decompression and serves as the most appropriate method without significant risks.

For every case in which the ossification was floated, migration of the floated plaque into the ventral bone resection area was observed during the postoperative period. This migration had gradually and obviously advanced at 6 months after surgery. Additionally, the majority of the migration had occurred by 3 months after surgery. This is the first report to describe this migration phenomenon in anterior decompression for thoracic OPLL. The CT myelograms obtained 1 month after surgery revealed spinal cord decompression resulting from adequate floating of the ossified PLL (Figs. 2 and 3). The subsequent plaque migration both demonstrated that floating had been completely achieved and provided additional spinal cord decompression during the postoperative course.

Conclusions

Floating of the ossified PLL via our posterolateral approach-based surgical technique was safe and effective in patients with beak-type OPLL in the thoracic spine. Gradual migration of the floated plaques provided additional spinal cord decompression during the postoperative course.

TABLE 1. Demographic and clinical characteristics of 6 patients

Case No.	Age (yrs)	Sex	Anterior Decompression Levels	Instrumented Fusion Levels	Anterior Decompression Type	OR (%)	Duration of Follow-Up	History	Operative Complication	Preop JOA	Postop JOA	Recovery (%)
1	63	M	T6–7	T4–9	Removal	71	34	RF, HT	None	1	5	57*
2	65	F	T5–6	T3–8	Removal	51	27	DM, HT	Dural tear	8	9.5	50
3	62	M	T5–6	T3–8	Removal	67	23	DM, HT	Neurol deter	5	8	50
4	53	F	T7–9	T5–11	Floating	82	23	DM	None	4.5	8	54
5	51	F	T5–6	T3–8	Floating	93	19	CP, LC	None	1.5	6	69†
6	58	F	T3–4	T1–7	Floating	81	14	DM	None	0.5	6.5	57

CP = cerebral palsy; DM = diabetes mellitus; HT = hypertension; JOA = JOA score; LC = liver cirrhosis; neurol deter = neurological deterioration; OR = occupying ratio; RF = renal failure.

* In Case 1, the total JOA score was calculated as 8 points by eliminating the score of 3 points for bladder function due to the patient's anuric renal failure.

† In Case 5, the total JOA score was calculated as 8 points by eliminating the score of 3 points for lower-extremity motor function due to the patient's cerebral palsy.

TABLE 2. Radiographic data for patients treated with the plaque-floating technique

	Time Since Op in Mos			
Variable & Case No.	1	3	6	12
Migration distance of floated OPLL (mm)				
4	2.9	5.0	5.3	5.4
5	1.5	4.0	4.3	4.4
6	2.8	4.0	4.4	4.6
Anterior decompression rate (%)				
4	31.9	54.9	58.2	59.3
5	13.6	36.0	39.1	40.0
6	25.0	35.7	39.3	41.1
Percentage recovery rate in JOA score (%)				
4	31	46	54	54
5	23	54	69	69
6	43	52	57	57

References

1. Fujimura Y, Nishi Y, Nakamura M, Toyama Y, Suzuki N: Long-term follow-up study of anterior decompression and fusion for thoracic myelopathy resulting from ossification of the posterior longitudinal ligament. **Spine (Phila Pa 1976) 22:**305–311, 1997
2. Hirabayashi K, Miyakawa J, Satomi K, Maruyama T, Wakano K: Operative results and postoperative progression of ossification among patients with ossification of cervical posterior longitudinal ligament. **Spine (Phila Pa 1976) 6:**354–364, 1981
3. Ido K, Shimizu K, Nakayama Y, Yamamuro T, Shikata J, Matsushita M, et al: Anterior decompression and fusion for ossification of posterior longitudinal ligament in the thoracic spine. **J Spinal Disord 8:**317–323, 1995
4. Kato S, Murakami H, Demura S, Yoshioka K, Hayashi H, Tsuchiya H: Novel surgical technique for ossification of posterior longitudinal ligament in the thoracic spine. **J Neurosurg Spine 17:**525–529, 2012
5. Kurosa Y, Yamaura I, Nakai O, Shinomiya K: Selecting a surgical method for thoracic myelopathy caused by ossification of the posterior longitudinal ligament. **Spine (Phila Pa 1976) 21:**1458–1466, 1996
6. Matsumoto M, Chiba K, Toyama Y, Takeshita K, Seichi A, Nakamura K, et al: Surgical results and related factors for ossification of posterior longitudinal ligament of the thoracic spine: a multi-institutional retrospective study. **Spine (Phila Pa 1976) 33:**1034–1041, 2008
7. Matsuyama Y, Yoshihara H, Tsuji T, Sakai Y, Yukawa Y, Nakamura H, et al: Surgical outcome of ossification of the posterior longitudinal ligament (OPLL) of the thoracic spine: implication of the type of ossification and surgical options. **J Spinal Disord Tech 18:**492–498, 2005
8. Min JH, Jang JS, Lee SH: Clinical results of ossification of the posterior longitudinal ligament (OPLL) of the thoracic spine treated by anterior decompression. **J Spinal Disord Tech 21:**116–119, 2008
9. Takahata M, Ito M, Abumi K, Kotani Y, Sudo H, Minami A: Clinical results and complications of circumferential spinal cord decompression through a single posterior approach for thoracic myelopathy caused by ossification of posterior longitudinal ligament. **Spine (Phila Pa 1976) 33:**1199–1208, 2008
10. Tokuhashi Y, Matsuzaki H, Oda H, Uei H: Effectiveness of posterior decompression for patients with ossification of the posterior longitudinal ligament in the thoracic spine: usefulness of the ossification-kyphosis angle on MRI. **Spine (Phila Pa 1976) 31:**E26–E30, 2006
11. Tomita K, Kawahara N, Baba H, Kikuchi Y, Nishimura H: Circumspinal decompression for thoracic myelopathy due to combined ossification of the posterior longitudinal ligament and ligamentum flavum. **Spine (Phila Pa 1976) 15:**1114–1120, 1990
12. Tsuzuki N, Hirabayashi S, Abe R, Saiki K: Staged spinal cord decompression through posterior approach for thoracic myelopathy caused by ossification of posterior longitudinal ligament. **Spine (Phila Pa 1976) 14:**1623–1630, 2001
13. Yonenobu K, Korkusuz F, Hosono N, Ebara S, Ono K: Lateral rhachotomy for thoracic spinal lesions. **Spine (Phila Pa 1976) 15:**1121–1125, 1990

Author Contributions

Conception and design: Kato, Murakami. Acquisition of data: Kato, Hayashi, Yokogawa, Fang. Analysis and interpretation of data: Kato, Demura, Yoshioka, Hayashi, Yokogawa. Drafting the article: Kato. Critically revising the article: Murakami, Demura, Yoshioka, Fang, Tsuchiya. Administrative/technical/material support: Murakami, Tsuchiya. Study supervision: Murakami, Tsuchiya.

Correspondence

Satoshi Kato, Department of Orthopaedic Surgery, Kanazawa University School of Medicine, 13-1 Takara-machi, Kanazawa, 920-8641, Japan. email: skato323@gmail.com.

Asian Spine Journal
Clinical Study
Asian Spine J 2015;9(3):321-326 • http://dx.doi.org/10.4184/asj.2015.9.3.321

Gait Analysis in Cervical Spondylotic Myelopathy

Hirosuke Nishimura, Kenji Endo, Hidekazu Suzuki, Hidetoshi Tanaka, Takaaki Shishido, Kengo Yamamoto

Department of Orthopedic Surgery, Tokyo Medical University, Tokyo, Japan

Study Design: Gait analysis of patients with cervical spondylotic myelopathy (CSM) by using a sheet-type gait analysis system.
Purpose: The aim of this study was to compare the gait patterns of patients with CSM, evaluated by the Nurick grades, and to determine the threshold values of gait parameters predicting the occurrence of a fall by using a gait recorder.
Overview of Literature: Gait disorder due to CSM may progress to severe paraplegia, following even a minor trauma such as a fall. The indications for the surgery of CSM without severe paralysis remain controversial. The quantitative gait analysis and the decision for decompressive surgery in patients with CSM are important in order to prevent severe paraplegia from a fall.
Methods: One hundred thirty-two subjects (normal, 34; CSM, 98) underwent gait analysis by using a sensor sheet. Measurements of gait cycle parameters included the step and stride length, step width, foot angle, swing phase, and stance phase. CSM was assessed by Nurick grade.
Results: Although the clinical symptoms were lacking, Nurick grade 1 had significant abnormalities in the parameters of velocity, step length, and step angle ($p<0.05$). Regarding the Nurick grade and walking phase, the length of the stance phase was increased to more than 70% of the entire walking cycle in Nurick grade 4.
Conclusions: Gait analysis was an objective tool for evaluating the gait stability. Our results suggested that when the percentage of the stance phase in the gait cycle increases to above 70%, the CSM patients have an increased fall risk.

Keywords: Gait; Movement disorders; Physical examination; Spinal cord diseases

Introduction

The total population of Japan is predicted to decrease by 2050, and a substantial increase is expected in the population of age 85 years and above [1]. With the rapid increase in the elderly population, the number of patients with locomotive syndrome has increased in Japan [2,3]. Locomotive syndrome increases the societal burdens with respect to mortality, quality of life, and economic costs. Cervical cord compression was present in 5% to 7% of asymptomatic subjects and was associated with physical performances in elderly people. It is suggested that locomotive syndrome involves the early stages of cervical spondylotic myelopathy (CSM). Especially in the elderly people, the gait impairment can be due to CSM which could aggravate the symptoms of cervical cord injury from a relatively minor trauma, such as a fall from a height, or even progress to severe quadriparesis. However, to the best of our knowledge, there are no objective criteria for the evaluation of gait analysis. The number of studies for gait analysis in CSM patients is small, and the indications for the surgery of CSM without severe paralysis still remain controversial [4-9]. Quantitative gait analysis and the decision for decompressive surgery in the patients with CSM are

Received Nov 10, 2014; Revised Nov 28, 2014; Accepted Nov 28, 2014
Corresponding author: Hirosuke Nishimura
Department of Orthopedic Surgery, Tokyo Medical University,
6-7-1 Nishishinjuku, Shinjuku-ku, Tokyo 160-0023, Japan
Tel: +81-3-3342-6111, Fax: +81-3-3342-5295, E-mail: hirosuke819@hotmail.com

Asian Spine Journal • pISSN 1976-1902 eISSN 1976-7846 • www.asianspinejournal.org

Table 1. Subjects

Nurick grade	Number	Age (yr)	Height (cm)	Weigt (kg)	Sex (male:female)
0	34	52.7±16.5	165±8.4	59.2±10.5	18:16
1	32	67.0±16.9	16±16.9	64.8±9.5	32:0
2	14	52.7±15.9	165±8.19	59.6±10.5	14:0
3	22	58.2±10.8	167±7.82	65.6±8.91	22:0
4	30	67.0±16.9	159±7.54	61.9±10.6	12:18

Values are presented as mean±standard deviation.

important, in order to prevent severe paraplegia resulting from a fall. The objective of our study is to compare the gait patterns in patients with CSM, which is evaluated by the Nurick grades, and to determine the threshold values of gait parameters predicting the occurrence of a fall by using a gait recorder.

Materials and Methods

The participants had a clinical diagnosis of CSM confirmed by magnetic resonance imaging findings. CSM was defined as a constellation of symptoms and signs supported by appropriate radiological findings, including symptoms (numb clumsy hands, impairment of gait, bilateral arm paresthesia) and signs (corticospinal distribution motor deficits, atrophy of hand intrinsic muscles, hyperflexia, positive Hoffman sign, upgoing plantar responses, lower limb spasticity, broad-based unstable gait). One hundred thirty-two subjects (normal, 34; CSM, 98) underwent gait analysis by using a sheet-type gait analysis system. All subjects provided written informed consent after receiving explanation of the experimental protocol, and this study was approved by the Institutional Review Board of our institution. The patient characteristics are summarized in Table 1. Patients with the following conditions were excluded from this study: hip, knee, and ankle joint disease; other spinal disease; and scoliosis of more than 10°. Participants with clear clinical and radiological evidence of CSM were included in this study.

The walking test was performed by using a 2.4 m long thin-type sensor sheet (Sheet Type Gait Analyzer Walk Way MW-1000, Anima, Tokyo, Japan).

The patients were examined 3 times, and the average of those results were taken. Measurements of gait cycle parameters included the step angle, step length, step width, and gait velocity. Outward rotation of the feet was

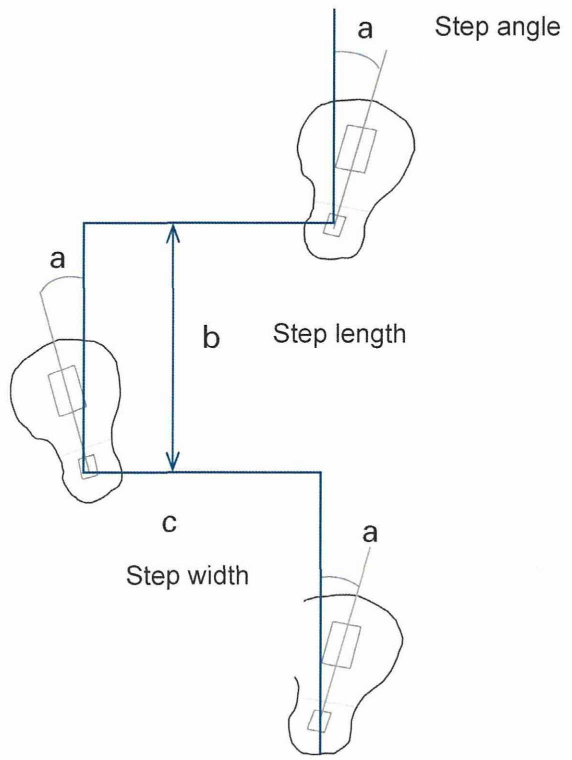

Fig. 1. Definitions of gait parameters.

measured as a positive angle (Fig. 1). The gait cycle consists of a swing phase and a stance phase. Generally, the stance phase and swing phase account for 60% and 40%, respectively (Fig. 2) [10]. The clinical gait disturbance in patients with CSM was evaluated according to the Nurick grade (Table 2) [11].

A medical statistical consultant performed the statistical analyses using the JMP software package, ver. 8.0 (SAS Institute Inc., Cary, NC, USA). The Tukey-Kramer HSD test was used to analyze the differences in the gait parameters among Nurick grades. The correlations among the variables of gait parameters were examined using the Spearman's rank correlation coefficient. The gait param-

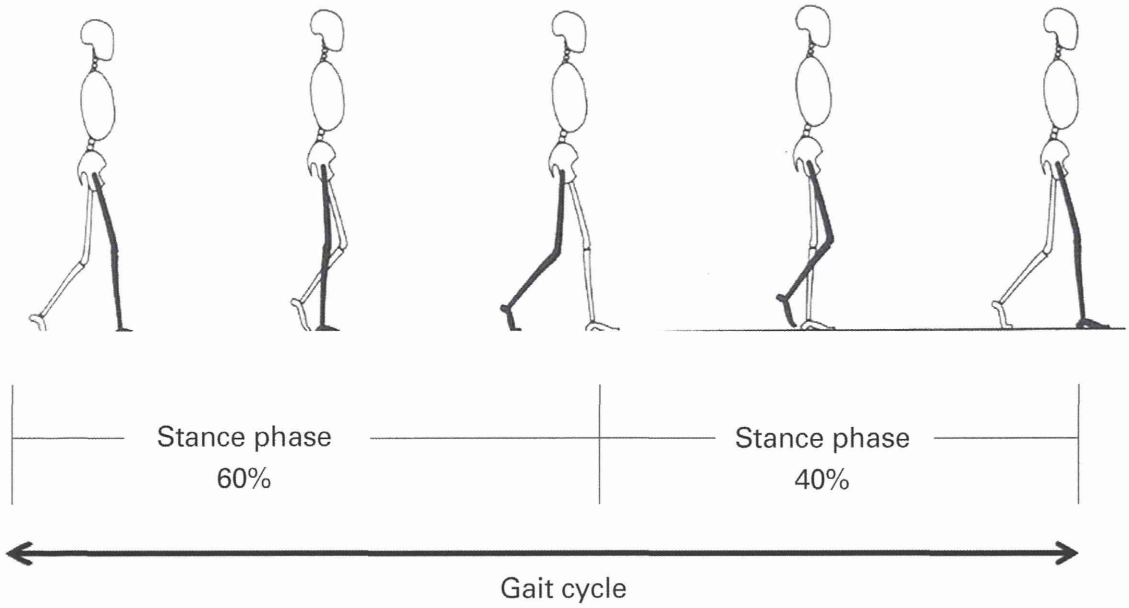

Fig. 2. Gait cycle.

Table 2. Nurick grades

Grade	Description
0	No evidence of cord involvement
1	Signs of cord involvement normal gait
2	Mild gait involvement able to be employed
3	Gait abnormality prevents employment
4	Able to ambulate only with assistance
5	Chair bound or bedridden → Exclude

Modified from Nurick [11].

eters and risk factors in the patients with Nurick grade 4 were evaluated by logistic regression analysis. A p-value of less than 0.05 was considered to indicate a statistically significant difference.

Results

Gait analysis without any subjective gait disturbance in patents with CSM.

In order to assess the subclinical findings, we compared Nurick grade 0 and 1. Although clinical symptoms were lacking, grade 1 had significant abnormalities in the parameters of velocity, step length, and step angle ($p<0.05$) (Fig. 3). The gait velocity and step length decreased as symptoms progressed. In contrast, step angle and step width increased (Fig. 3).

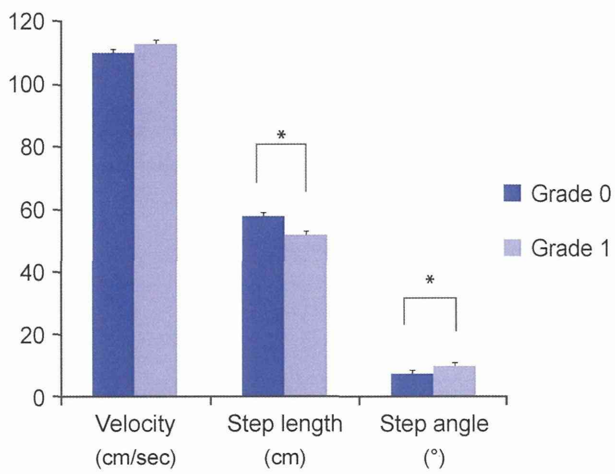

Fig. 3. Nurick grade 0 vs. grade 1. *$p<0.05$

1. Gait analysis and CSM severity

For the Nurick grades 1, 2, 3, and 4, the gait parameters were as follows, respectively (Figs. 4, 5): gait velocity (cm/sec) 107.7±2.5, 94.5±2.7, 92.2±4.0, 76.3±4.1; step length (cm) 59.0±1.2, 52.9±1.3, 48.4±1.7, 44.3±1.7, 32.0±2.9; step width (cm) 7.73±2.0, 9.1±2.2, 9.8±2.1, 11.3±2.1, 12.5±3.0; step angle (°) 7.57±2.1, 10.5±1.6, 11.9±2.1, 14.5±1.6, 23.5±2.2; stance phase (%) 64±4.4, 67±4.4, 67±3.4, 73±4.4. When the grade of Nurick classification became worse, the velocity and step length decreased, and the step width and step angle increased (Fig. 4).

Regarding the Nurick grade and walking phase, the length of the stance phase increased in more than 70% of the entire walking cycle in the patients with Nurick grade 4. There were no changes in the swing phase (Fig. 5).

2. The evaluation of fall risk

We established that grade 4 patients, who were able to ambulate only with assistance, were at the great risk for a fall.

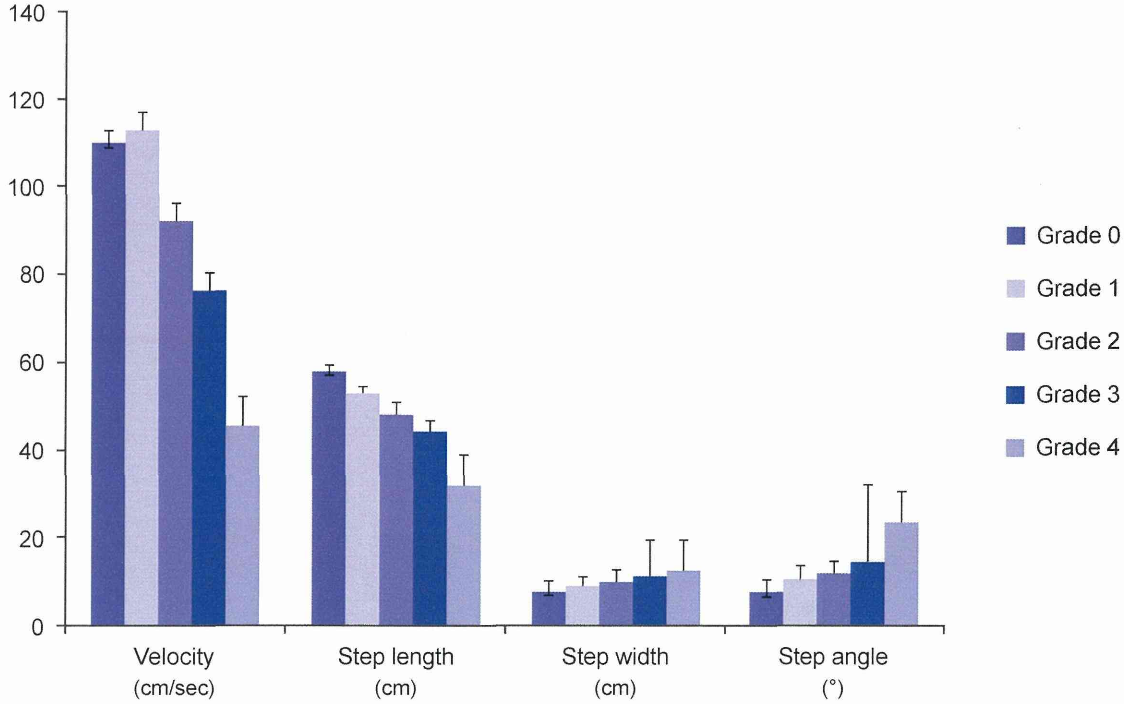

Fig. 4. Nurick grade and gait parameters.

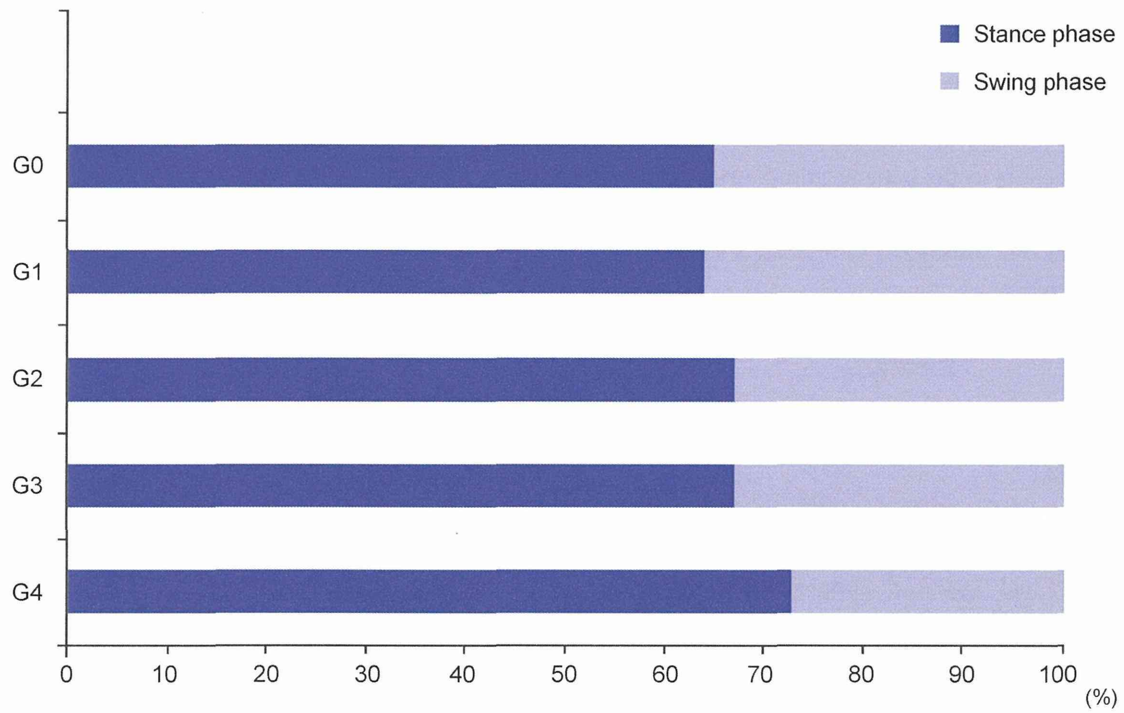

Fig. 5. Nurick grade and walking phase.

Table 3. Risk factors in Nurick grade 4 patients

	Odds ratio	p-value
Velocity	2.8	0.001
Step length	2.1	<0.001
Step width	1.2	0.02
Step angle	1.3	<0.001
Stance phase	4.1	<0.001
Swing phase	0.04	0.006
Gait cycle	1.2	<0.001

Logistic regression analysis showed that the stance phase and swing phase were risk factors for falls for Nurick grade 4 patients (odds ratio=4.1; $p<0.002$).

Logistic regression analysis revealed that the stance phase was a risk factor for falls in the patients with Nurick grade 4 (odds ratio=4.1; $p<0.002$) (Table 3). Our results suggested that the threshold value of the stance phase for increasing risk of falls was likely to be about 70% of the gait cycle.

Discussion

The Japanese Orthopedic Association (JOA) has proposed the term "locomotive syndrome". This syndrome is caused by weakening of the musculoskeletal organs such as bones, joints, and muscles [2,3]. It is suggested that locomotive syndrome involves the early stages of CSM. Culhane et al. [12] demonstrated that the falls are a major source of morbidity and mortality in the elderly population. Falls occurring in the patients with CSM are particularly likely to result in a permanent, severe quadriparesis. It was reported that the injury in CSM, associated with ossification of the posterior longitudinal ligament, is a poor prognostic factor; and in 15/156 patients (9.6%), CSM became worse by even a minor external injury [13].

1. Gait analysis

Gait impairment is a primary symptom of CSM. Previous studies have identified the reduction in knee flexion, in the early stages of the disease; and in more severe stages of the disease, decreased ankle plantar flexion at the terminal stance and reduced knee flexion during loading response were found [13]. However, little is known about the specific kinetic and kinematic gait parameters.

2. Changes in gait parameters without subjective symptoms

It has been stated that CSM patients have an unstable gait due to spasticity of the gastrocnemius at the time of gait grounding [14]. However, the gait pattern of patients with early stage CSM is unclear. Our results suggested that in CSM patients without subjective symptoms, the gait analysis showed significant changes in the step length, step width, and step angle, as compared with healthy persons. Gait analysis could be reflecting the subclinical conditions.

3. The differences among gait parameters according to Nurick grade

When the grade of Nurick classification worsened, the velocity and step length were decreased, and the step width and step angle were increased, so as to maintain a stable walk. The differences among those parameters were remarkable, especially between the grades 3 and 4 (Fig. 4). Regarding the risk factors among gait parameters in the patients with Nurick grade 4, the stance phase was remarkable in the odds ratio. This feature may be an expression of the myelopathy and may serve as a protective mechanism from falls.

4. The evaluation of fall risk and treatment

We established that the patients with grade 4, who are able to ambulate only with assistance, are at great risk for a fall. Our results suggested that the threshold value of the stance phase for preventing falls might be about 70%. A recent study has recommended that regarding the timing of operations for CSM, patients with Nurick grade 2 CSM were most likely to improve from surgery [15]. The gait analysis using a sheet-type gait analysis system was a useful tool for catching a gait abnormality in the initial stage of CSM without gait disturbance. The compensation mechanism for unstable gait in the patients with CSM was reduction of the velocity and prolongation of the stance phase by a greater step angle and a smaller step length. A previous paper mentioned that at a self-selected speed, the CSM patients walked slowly, with shorter stride lengths and longer double support durations [4]. When the protective mechanism was beyond control, a fall could occur with a smaller step angle and step width. The extension of stance phase more than 70% in the total cycle period may be a threshold of falls. This study has some limitations. There are some differences between the males and females, in the factors including height and weight. However, these may have little influence on the gait cycle, and more complex measurements (e.q., three dementional analysis) were not performed.

Another limitation of this study was that the investigation of the direct link between gait disorder and the resulting occurrence of actual falls in CSM patients could not be carried out for the obvious ethical reasons. For future research, when gait disorder worsens by the aggravation of spastic paralysis, we should consider the breakdown of these protective mechanisms and facilitate the prevention of falls by implementing safety protocols.

Conclusions

Gait analysis was an objective tool for evaluating the gait stability, even when the CSM patients had no subjective gait disturbance. The compensation mechanism for unstable gait in the patients with CSM was the reduction of velocity and prolongation of the stance phase by a greater step angle and smaller step length. When the stance phase in the gait cycle increases to above 70% of the total cycle period, the CSM patients may have an increased fall risk.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

Acknowledgments

We are indebted to the Department of International Medical Communications of Tokyo Medical University for the editorial review of the English manuscript. The authors would like to thank Ms. Yuri Amamizu of the Department of Orthopedic Surgery for the initial English draft.

References

1. National Institute of Population and Social Security Research. Population projection for Japan [Internet]. Tokyo: National Institute of Population and Social Security Research; 2012 [cited 2015 Apr 21]. Available from: http://www.ipss.go.jp/syoushika/tohkei/newest04/sh2401smm.html .
2. Nakamura K. A "super-aged" society and the "locomotive syndrome". J Orthop Sci 2008;13:1-2.
3. Nakamura K. The concept and treatment of locomotive syndrome: its acceptance and spread in Japan. J Orthop Sci 2011;16:489-91.
4. Malone A, Meldrum D, Bolger C. Gait impairment in cervical spondylotic myelopathy: comparison with age- and gender-matched healthy controls. Eur Spine J 2012;21:2456-66.
5. Singh A, Crockard HA. Quantitative assessment of cervical spondylotic myelopathy by a simple walking test. Lancet 1999;354:370-3.
6. Lee JH, Lee SH, Seo IS. The characteristics of gait disturbance and its relationship with posterior tibial somatosensory evoked potentials in patients with cervical myelopathy. Spine (Phila Pa 1976) 2011;36:E524-30.
7. Suzuki E, Nakamura H, Konishi S, Yamano Y. Analysis of the spastic gait caused by cervical compression myelopathy. J Spinal Disord Tech 2002;15:519-22.
8. Moorthy RK, Bhattacharji S, Thayumanasamy G, Rajshekhar V. Quantitative changes in gait parameters after central corpectomy for cervical spondylotic myelopathy. J Neurosurg Spine 2005;2:418-24.
9. McDermott A, Bolger C, Keating L, McEvoy L, Meldrum D. Reliability of three-dimensional gait analysis in cervical spondylotic myelopathy. Gait Posture 2010;32:552-8.
10. Murray MP, Drought AB, Kory RC. Walking patterns of normal men. J Bone Joint Surg Am 1964;46:335-60.
11. Nurick S. The pathogenesis of the spinal cord disorder associated with cervical spondylosis. Brain 1972;95: 87-100.
12. Culhane KM, O'Connor M, Lyons D, Lyons GM. Accelerometers in rehabilitation medicine for older adults. Age Ageing 2005;34:556-60.
13. Matsunaga S, Nakamura K, Seichi A, et al. Radiographic predictors for the development of myelopathy in patients with ossification of the posterior longitudinal ligament: a multicenter cohort study. Spine (Phila Pa 1976) 2008;33:2648-50.
14. Maezawa Y, Uchida K, Baba H. Gait analysis of spastic walking in patients with cervical compressive myelopathy. J Orthop Sci 2001;6:378-84.
15. Pumberger M, Froemel D, Aichmair A, et al. Clinical predictors of surgical outcome in cervical spondylotic myelopathy: an analysis of 248 patients. Bone Joint J 2013;95:966-71.

Research article

Cervical ossification of the posterior longitudinal ligament: Biomechanical analysis of the influence of static and dynamic factors

Norihiro Nishida, Tsukasa Kanchiku, Yoshihiko Kato, Yasuaki Imajo, Yuichiro Yoshida, Syunichi Kawano, Toshihiko Taguchi

Department of Orthopedic Surgery, Yamaguchi University Graduate School of Medicine, Ube, Yamaguchi, Japan

Objective: Cervical myelopathy due to ossification of the posterior longitudinal ligament (OPLL) is induced by static factors, dynamic factors, or a combination of both. We used a three-dimensional finite element method (3D-FEM) to analyze the stress distributions in the cervical spinal cord under static compression, dynamic compression, or a combination of both in the context of OPLL.

Methods: Experimental conditions were established for the 3D-FEM spinal cord, lamina, and hill-shaped OPLL. To simulate static compression of the spinal cord, anterior compression at 10, 20, and 30% of the
1 anterior–posterior diameter of the spinal cord was applied by the OPLL. To simulate dynamic compression, the OPLL was rotated 5°, 10°, and 15° in the flexion direction. To simulate combined static and dynamic compression under 10 and 20% anterior static compression, the OPLL was rotated 5°, 10°, and 15° in the flexion direction.

Results: The stress distribution in the spinal cord increased following static and dynamic compression by cervical OPLL. However, the stress distribution did not increase throughout the entire spinal cord. For combined static and dynamic compression, the stress distribution increased as the static compression increased, even for a mild range of motion (ROM).

Conclusion: Symptoms may appear under static or dynamic compression only. However, under static compression, the stress distribution increases with the ROM of the responsible level and this makes it very likely that symptoms will worsen. We conclude that cervical OPLL myelopathy is induced by static factors, dynamic factors, and a combination of both.

Keywords: Cervical myelopathy, Dynamic factor, Finite element method, Static factor, Ossification of the posterior longitudinal ligament

Introduction

Cervical ossification of the posterior longitudinal ligament (C-OPLL) is recognized as a common clinical entity that causes complicated myelopathy of the cervical spinal cord. It is believed that myelopathy develops due to compression of the spine by C-OPLL. However, some patients with little ossification exhibit myelopathy, whereas others with marked ossification do not. Thus, static factors alone cannot account for myelopathy.[1] Dynamic factors such as mobility of the spinal column are also important in the development of cervical myelopathy.[1–4] However, the contribution of dynamic factors to the development of myelopathy in C-OPLL patients has not been fully determined.[5] C-OPLL myelopathy is therefore likely to be induced by static factors, dynamic factors, or a combination of both.[6,7]

In the present study, we used a three-dimensional finite element method (3D-FEM) to analyze the stress distributions of cervical spinal cord under static compression, dynamic compression, and a combination of both in the context of OPLL.

Material and methods

The ABAQUS 6.11 (Valley Street, Providence, RI,
USA) finite element package was used for FEM simu- 2
lation. The 3D-FEM spinal cord model used in this

Correspondence to: Norihiro Nishida, Department of Orthopedic Surgery, Yamaguchi University Graduate School of Medicine, 1-1-1 Minamikogushi, Ube, Yamaguchi 755-8505, Japan. Email: nishida3@yamaguchi-u.ac.jp

DOI 10.1179/2045772314Y.0000000221