

# Journal of Craniovertebral Junction and Spine

Case Report

# Occipitalized os odontoideum: A case report

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#### **Abstract**

We report on a 36-year-old man presenting with a sudden onset of motor weakness and numbness in the upper extremities following a fall from a truck bed. Radiological findings demonstrated an os odontoideum and osseous continuity between the occiput and an ossicle, termed an "occipitalized os odontoideum." The occipitalized ossicle and atlas moved as a functional unit from the body of the axis. He underwent atlantoasxial stabilization with an atlas lateral mass screw and axis pedicle screw. Eighteen months later, he remained free of symptoms and showed solid bone fusion. Atlantoaxial stabilization resulted in an excellent clinical outcome for this condition. Our report provides useful knowledge regarding treatment of extremely rare osseous anomalies in the craniovertebral junction.

Key words: Atlantoaxial instability, os odontoideum, occipitalized atlas, spinal cord injury

#### INTRODUCTION

An os odontoideum is a rare condition in the craniovertebral junction (CVJ) which can cause mild progressive myelopathy or sudden spinal cord injury even after minor trauma. [1] Here, we report our experience with a patient who had a spinal cord injury due to atlantoaxial instability secondary to os odontoideum associated with an osseous continuity between the occiput and an ossicle, which was termed an "occipitalized os odontoideum." To our knowledge, this is the first report describing an occipitalized os odontoideum.

#### **CASE REPORT**

A 36-year-old man presented with sudden motor weakness and numbness in the upper extremities following a fall from a

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truck bed. Neurological examination revealed motor weakness (grade 4/5) in his left triceps and intrinsic muscles. He had decreased sensation in both hands. He had intact cranial nerves and hyperreflexia in the left upper extremity with a positive Hoffmann sign. Knee and ankle reflexes were also abnormally increased bilaterally, with positive Babinski signs. A spastic gait and clumsiness of his hands were also noted. He denied any bowel or bladder difficulties.

Plain lateral radiographs showed an os odontoideum, and atlantoaxial instability was demonstrated during flexion-extension [Figure 1] Reconstruction computed tomography (CT) images demonstrated osseous continuity between the occiput and an ossicle, termed as "occipitalized os odontoideum." The occipitalized ossicle and atlas moved as a functional unit from the body of the axis [Figure 2]. Magnetic resonance imaging (MRI) demonstrated intramedullary high signal intensity changes at the posterior arch of the atlas [Figure 3a]. High signal-intensity changes without spinal cord compression suggest the presence of focal spinal cord contusion due to instability between the cranial unit and the body of the axis.

The patient underwent uncomplicated atlantoaxial stabilization with an atlas lateral mass screw and axis pedicle screw. The bone graft harvested from the iliac crest was interposed between the posterior arc of atlas and the lamina of the axis. His post-operative

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course was uneventful. His symptoms, including motor weakness and sensory disturbance, improved shortly after surgery. At the 8-month follow-up examination, he had no symptoms, and dynamic lateral radiographs showed stabilization between the atlas and axis. MRI revealed that the intramedullary high-intensity lesion had disappeared [Figure 3b]. At month 18, he remained free of symptoms and showed solid bone fusion [Figure 4].

#### DISCUSSION

An os odontoideum is defined as the dissociation between the body of the axis and the dens, such that a disconnected ossicle takes the place of an intact odontoid process.<sup>[2]</sup> Surgical treatments, such as posterior atlantoaxial fixation with an atlas lateral mass screw and axis pedicle screw, have been reported as the mainstay of treatment for the patients who have os odontoideum and show neurological symptoms. They have also been reported, on occasion, to be a preventive treatment for spinal cord injury in patients without neurological symptoms.<sup>[1,3,4]</sup> Although the choice of surgical procedures for

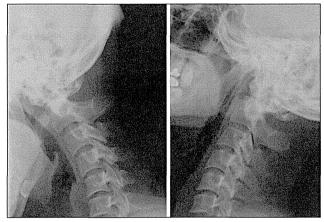


Figure 1: Lateral radiographs of cervical spine showing an osodontoideum and atlantoaxial instability during flexion-extension

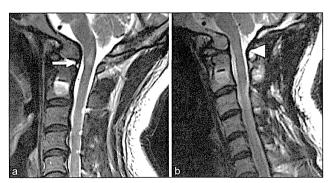


Figure 3:(a) Pre-operative sagittal T2-weighted magnetic resonance image demonstrating intramedullary high signal-intensity changes at the posterior arc of the atlas. High signal-intensity changes between the cranial unit and the body of the axis without spinal cord compression (arrow) suggest the presence of instability at the level (b) Post-operative sagittal T2-weighted magnetic resonance image showing that the intramedullary high signal-intensity changes at the posterior arc of the atlas had disappeared (arrowhead)

patients with atlantoaxial instability depends on the pathology of the instability, the patients' individual anatomical features, and their comorbidity, pre-operative imaging in the craniovertebral junction may often show concomitant diseases or conditions, such as osseous anomalies, [5,6] vertebral artery anomalies, [7] and congenital disease. [8] Knowledge of the treatments for these coexisting states can help the surgeon to prevent intra-or post-operative complications. We reported a case of an occipitalized os odontoideum, which was an extremely rare osseous anomaly in CVJ, causing spinal cord injury following an accidental fall. Atlantoaxial fixation was performed, which resulted in an excellent clinical outcome for this condition. We believe that our experience provide a basis for the selection of surgical procedure in this rare condition in CVJ.

The surgical stabilization of the os odontoideum with atlantoaxial fusion involving transarticular screw fixation or atlas lateral mass and axis pedicle fixation with polyaxial screws and rods have been described as mainstay surgical treatments. However, in this special condition, the main fear was that atlantoaxial fixation with atlas lateral mass screws and axis pedicle screws alone might be insufficient to fix the cranial unit consisting of the occipitalized ossicle and atlas with the axis. Pre-operative imaging revealed instability between the unit and the body of the axis, not atlantodental or occipitoatlas instability. Hence, we chose to perform atlantoaxial stabilization and not occipitocervical fixation. Two previous reports regarding surgical



Figure 2: Sagittal reconstruction computed tomography showing an osseous continuity between occiput and an ossicle

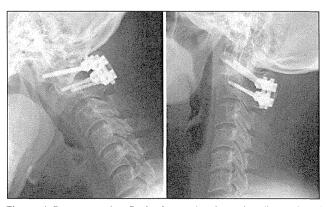


Figure 4: Post-operative flexion/extension lateral radiographs of cervical spine showing rigid fixation between atlas and axis

treatment for atlantoaxial instability with an occipitalized atlas, resembling the current case with the presence of a fused cranial unit, have been published. Jain et al. [9] performed occipitoaxial posterior fusion for 46 patients having congenital atlantoaxial dislocation with an occipitalized atlas. After that, Goel et al.[10] reported eight patients with an occipitalized atlas who had a mobile and reducible atlantoaxial dislocation and underwent lateral mass plate and screw fixation instead of fixation of the much longer plates or rods necessary for occipitocervical fixation. The method could be used even in the situation where the facet of the atlas is occipitalized. This latter report supports our consideration that the shortest fixation for the extent of instability is the ideal intervention. Use of atlantoaxial fixation, instead of occipitoaxial fixation, has the advantage that it can provide stabilization in the anteroposterior direction, while still preserving the flexion-extension motion between the cranial unit and the body of the axis. Although occipitoaxial fixation for this condition seems to be a reasonable strategy for surgical treatment, the longer fixation can spoil the flexion-extension motion and increase the mobility and loading at the inferior adjacent segment.

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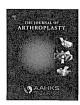
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### Total Hip Arthroplasty After Rotational Acetabular Osteotomy

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#### ABSTRACT

In this study, we aimed to determine whether the outcomes of total hip arthroplasty (THA) after rotational acetabular osteotomy (RAO) are equal to those of primary THA, and to elucidate the characteristics of THA after RAO. The clinical and radiographic findings of THA after RAO (44 hips), with minimum 24 months of follow-up, were compared with a matched control group of 58 hips without prior RAO. We found that the outcomes in terms of functional scores and complication rates did not differ between THA after RAO and THA without previous pelvic osteotomy, indicating that the results of THA after RAO are equivalent to those of primary THA. Although THA after RAO requires technical considerations, similar clinical outcomes to primary THA can be expected.

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Rotational acetabular osteotomy (RAO) is a type of periacetabular osteotomy used to treat symptomatic dysplasia of the acetabulum [1]. This procedure involves restoration of the femoral head coverage, resulting in pain relief and delays or prevention of the onset of arthritis. In Japan, there are reportedly a higher proportion of patients with dysplastic hips than in other countries [2], and many of these patients have undergone RAO. While some studies have reported good results of RAO [3–6], some patients require subsequent total hip arthroplasty (THA) because of pain secondary to progression of arthritis.

Several reports are available on THA after periacetabular osteotomy [7–11]. Most authors reported that THA after periacetabular osteotomy requires technical consideration and careful radiographic evaluation because the acetabulum may undergo morphologic changes. In terms of clinical results, one study reported that Bernese periacetabular osteotomy does not compromise the outcome of THA [11], whereas another study reported that the outcomes of THA after triple innominate osteotomy were not equivalent to those of primary THA [8]. However, it should be noted that these studies all had small sample

sizes or were not comparative studies. To date, only one published case report of THA after RAO is available [12], and the effects of a previous RAO on subsequent THA are still unknown.

In this study, we aimed to determine whether the outcomes of THA after RAO are equal to those of primary THA, and to elucidate the characteristics of THA after RAO by comparing the clinical and radiographic findings of patients who underwent THA after RAO with matched controls who underwent THA without prior RAO.

#### **Materials and Methods**

This investigation was a retrospective chart and radiographic review comparing two groups of patients. We obtained institutional ethics board approval for the study, which was performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki. All patients provided informed consent to participate in the study. Between 1999 and 2011, we performed THA on 45 hips in 43 patients who had previously undergone RAO. One patient was lost to follow-up, resulting in the study group comprising of 44 hips in 42 patients. For comparative purposes, 58 age- and gender-matched hips in 58 patients who had undergone THA for osteoarthritis secondary to hip dysplasia during same period were identified and included as the control group. None of the patients in the control group had undergone any prior pelvic osteotomy.

The preoperative data analyzed included age at THA, gender, interval from RAO to THA (years), body mass index (BMI), the Crowe classification [13] of hip joints, pre-THA contralateral hip joint status, and previous femoral osteotomy. Post-operative data comprised the follow-up duration after THA.

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Surgical Procedure

All THAs were performed in the lateral decubitus position and through a posterior approach. The incision used differed from that used in the preceding RAO, which had been performed through a combined anterior and posterior approach with a single incision, as described by Ninomiya et al [1]. We used the combined approach described by Lusskin et al [14] for 35/44 hips in the study group and 42/58 hips in the control group. We did not perform trochanteric osteotomy in any joints. We attempted to place the acetabular cup with an abduction angle of between 30° and 50° [15]. After the acetabular preparation, the center of reaming was decided, and a gouge was used to remove the subchondral bone to measure the distance to the medial wall. Initial medialization of the acetabular reaming was performed using the smallest reamer, after which the diameter of the reamer was gradually increased. When there was uniform contact between the reamer and acetabular bone, a cup of that size was selected. All patients received a cementless acetabular component with 4 fins and additional screw fixation if required. After the final femoral reaming and rasping, trial reduction was performed. If a bony impingement occurred, any osteophytes of the acetabulum were removed using a chisel or bone rongeur forceps luer. Upon resolving the bony impingement, the final implantation of the femoral component was performed. All femoral components used were also cementless devices. The Mallory-Head acetabular and Bimetric stem systems (Biomet, Warsaw, IN, USA) were used on 32 hips in the study group and 38 hips in the control group, whereas the Q5LP acetabular and K-MAX stem systems (Kyocera Medical Corp, Osaka, Japan) were used in 12 and 20 hips in the study and control groups, respectively.

Computed tomography (CT) scans were obtained in all patients in the study group in order to determine the three-dimensional shape of the acetabulums.

#### Operative Data and Clinical Evaluation

Operative data, including the operative time, intraoperative estimated blood loss, removal of osteophytes, and the size of acetabular cups used, were obtained using clinical records.

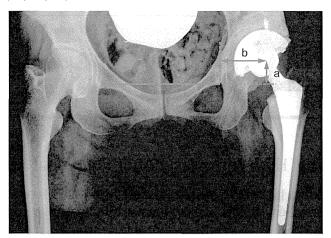
Hip joint function was evaluated according to the Merle d'Aubigné-Postel score [16] preoperatively and at the final follow-up. Reoperation and complications, including infection, venous thromboembolism, dislocation, nerve palsy, and wound healing problems, were recorded.

#### Radiographic Evaluation

Radiographic evaluations were performed using anteroposterior radiographs taken before and immediately after THA, and at the final follow-up. The acetabular cup position was evaluated on the radiographs obtained immediately post-surgery. We measured the abduction angle of the acetabular cup and the hip joint center position. The hip joint center position was defined as the vertical and horizontal distances from the teardrop, as described by Fukui et al [17] (Fig. 1). The magnification of each radiograph was calibrated from the known and measured diameters of the prosthetic femoral head. Loosening of the acetabular cup and heterotopic bone formation were evaluated on the radiographs obtained immediately post-THA and at the final follow-up. The acetabular cup was considered to be loosening if there was more than 3 mm of migration or a change of at least 4° in the abduction angle [18]. We used the classification system developed by Brooker et al [19] to qualitatively evaluate heterotopic bone formation.

#### Statistical Analysis

Statistical analysis of the differences between the study and control groups was conducted using JMP Pro 10.0 (SAS Institute, Cary, NC, USA). The independent-sample t test was used for continuous variables,



**Fig. 1.** Measurement of hip joint center position. The hip joint center position was defined as the vertical distance (a) and horizontal distance (b) from the teardrop.

whereas the chi-square test or Fisher's exact test was used for dichotomous values according to the validity conditions. All statistical tests were two-tailed, and a significance level of 0.05 was used.

#### Results

#### Demographics

The demographic and clinical baseline data of the patients are shown in Table 1. The mean age at the time of THA, gender, BMI, previous femoral osteotomy, and follow-up duration did not differ significantly between the two groups. Furthermore, the ratio of the Crowe classification of the preoperative hip joints and the contralateral hip joint status were also not significantly different between the groups. The average time interval between RAO and THA was 21 years (range, 7–37 years).

#### Operative Data and Clinical Evaluation

The operative and clinical data are shown in Table 2. The operative time in the study group was significantly longer than in the control

**Table 1** Baseline Characteristics of the Study Patients (n = 100).

	Study Group	Control Group	P
Number of hips	44	58	
Number of patients	42	58	
Gender (M/F)	2/40	2/56	
Age at RAO (years)	$34 \pm 12.4 (11-53)$	N/A	
Age at THA (years)	$55.6 \pm 7.8 (36-72)$	$56.2 \pm 5.1 (46-67)$	0.64
Interval from RAO to	$21 \pm 7.3 (7-37)$	N/A	
THA (years)			
BMI (kg/m²)	$22.8 \pm 3.4$	$22.3 \pm 2.7$	0.65
	(17.3-32.0)	(17.5-27.6)	
Crowe classification			0.87
I	29 (66.0%)	39 (67.3%)	
II	10 (22.7%)	14 (24.1%)	
III	3 (6.8%)	4 (6.9%)	
IV	2 (4.5%)	1 (1.7%)	
Contralateral joint			0.77
Normal	14 (31.8%)	21 (38.1%)	
OA	20 (45.5%)	27 (46.6%)	
THA	10 (22.7%)	10 (17.2%)	
Previous femoral osteotomy	2 (4.5%)	4 (6.9%)	1.00
Follow-up (months)	55.8 ± 36.2 (24–107)	62.9 ± 28.4 (24–95)	0.06

Data are presented as mean  $\pm$  standard deviation (range) or number (%). Abbreviations: M, male; F, female; RAO, rotational acetabular osteotomy; THA, total hip arthroplasty; N/A, not applicable; BMI, body mass index; OA, osteoarthritis.

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**Table 2**Operative Data and Clinical Evaluations.

*			
	Study Group $(n = 44)$	Control Group (n = 58)	P
Operative time (min)	177 ± 41 (115-227)	161 ± 36 (91-206)	0.03ª
Blood loss (g)	567 ± 232 (140-1445)	$524 \pm 254 (50 - 1710)$	0.15
Osteophyte removal	40 (90.9%)	31 (53.4%)	<0.001 <sup>a</sup>
Combined approach	35 (79.5%)	42 (72.4%)	0.33
Cup size (mm)	$50 \pm 3.2 (46-58)$	$48 \pm 2.1 (46-54)$	$< 0.001^a$
MA score (preoperative)			
Total	$8.3 \pm 1.7 (5-12)$	$8.4 \pm 2.1 (4-13)$	0.74
Pain	$2.3 \pm 0.7 (1-4)$	$2.2 \pm 0.8 (1-4)$	0.26
Mobility	$3.2 \pm 1.2 (1-6)$	$3.6 \pm 1.3 (1-6)$	$0.04^{a}$
Walking	$2.8 \pm 0.8 (1-5)$	$2.7 \pm 0.8 (1-5)$	0.31
MA score (last follow-up	)		
Total	$15.2 \pm 1.7 (11-18)$	$15.7 \pm 1.8  (11-18)$	0.12
Pain	$5.3 \pm 0.6 (4-6)$	$5.5 \pm 0.6 (4-6)$	0.05
Mobility	$5.1 \pm 0.9 (3-6)$	$5.4 \pm 0.8 (3-6)$	0.12
Walking	$4.8 \pm 0.8 (3-6)$	$4.8 \pm 1 (2-6)$	0.78
Complications			
Infection	0	1	1.00
VTE	0	2	1.00
Dislocation	0	0	1.00
Nerve palsy	0	0	1.00
Wound healing problems	0	0	1.00
Reoperation	0	1	1.00

Data are presented as mean  $\pm$  standard deviation (range) or number (%). Abbreviations: MA, Merle d'Aubigné-Postel; VTE, venous thromboembolism.

group (P = 0.029); however, there were no differences in the estimated blood loss. In 40/44 hips (90.9%) in the study group, removal of osteophytes of the acetabular anterior wall was performed because of bony impingement, whereas this procedure was performed in only 31/58 hips (53.4%) in the control group.

The mean diameter of the acetabular cup used was 50 mm (range, 46–58 mm) in the study group, and 48 mm (range, 46–54 mm) in the control group (P<0.001). There were no differences in the preoperative total Merle d'Aubigné-Postel score between the groups; however, the mobility score in the study group was significantly lower than in the control group (P = 0.043). At the last follow-up, the total Merle d'Aubigné-Postel, pain, mobility, and walk scores in the study and control groups were significantly improved compared with the preoperative scores. However, no significant differences were observed between the groups in terms of the improvements in the clinical results from before THA and the last follow-up. Reoperation was not needed for any patient. Postoperative complications included one case of infection and 2 cases of venous thromboembolism in the control group, whereas there were no cases of dislocation, wound healing problems, or nerve palsy.

#### Radiographic Evaluation

The radiographic data are shown in Table 3. The mean acetabular cup abduction angles were  $40.7^{\circ}$  (range,  $30^{\circ}-52^{\circ}$ ) and  $43.5^{\circ}$  (range,  $22^{\circ}-66^{\circ}$ ) in the study and control groups, respectively. Outliers of acetabular cup abduction angle were one hip >50° in the study group, and  $10 \text{ hips} > 50^{\circ}$  in the control group (P = 0.021).

The mean vertical distances of the hip joint center position after THA were 25.7 mm (range, 11–40 mm) and 23.7 mm (range, 13–41 mm) in the study and control groups, respectively. The mean horizontal distances were 31.2 mm (range, 21–42 mm) and 28.1 mm (range, 19–37 mm) in the study and control groups, respectively. While there was no significant difference in the vertical distance, the horizontal distance in the study group was found to be significantly larger than in the control group (P=0.002), suggesting that the acetabular cup of THA after RAO was placed laterally. Moreover, there was no loosening of the acetabular and femoral component in either group. Heterotopic

**Table 3**Radiographic Evaluations.

	Study Group $(n = 44)$	Control Group $(n = 58)$	P
Loosening	0	0	
Heterotopic ossification			$0.008^{a}$
0	29 (65.9%)	52 (89.7%)	
1	11 (25%)	6 (10.3%)	
2	3 (7.0%)	0 (0%)	
3	1 (2.3%)	0 (0%)	
Cup abduction (°)	$40.7 \pm 5.2 (30-52)$	$43.5 \pm 8.2 (22-66)$	$0.02^{a}$
Hip joint center			
Vertical distance (mm)	$25.7 \pm 6.5 (11-40)$	$23.7 \pm 5.7 (13-41)$	0.09
Horizontal distance (mm)	$31.2 \pm 5.3 (21-42)$	$28.1 \pm 3.8  (19  37)$	$0.002^{a}$

Data are presented as mean  $\pm$  standard deviation (range) or number (%).

bone formations were seen in 15/44 hips (34.1%) (Grade I: 11 hips, Grade II: 3 hips, and grade III: 1 hip) in the study group and in 6/58 hips (10.3%) (Grade I: 6 hips) in the control group (P = 0.008).

#### Discussion

In this study, we demonstrated that the results of THA after RAO were comparable to those of primary THA, and reported on 7 specific characteristics of THA after RAO. We found that the outcomes in terms of the functional scores and complication rates did not differ between THA after RAO and THA without previous pelvic osteotomy, indicating that the results of THA after RAO are equivalent to those of primary THA. The characteristics of THA after RAO (study group) were as follows: the preoperative range of hip motion was poorer, the operative time was longer, the acetabular cups used were larger, removal of osteophytes was needed in more cases, heterotopic bone formations after THA were seen more frequently, the abduction angles of the acetabular cups were smaller, and their position tended to be lateral.

In most patients, removal of large osteophytes was needed after RAO, and we speculate that the presence of osteophytes might be associated with a poorer preoperative range of hip joint motion. In turn, removal of the osteophytes and the poor hip joint motion might be responsible for the prolonged operation time observed in the study group. Moreover, the acetabular cups used in the study group were larger than in the control group, indicating that the acetabulums after RAO may become wider than before RAO.

Interestingly, the abduction angles of the acetabular cups were lower in the study group than those in the control group. The abduction angles of 43/44 (97.7%) acetabular cups in the study group were within the target range, compared to only 48/58 (82.8%) acetabular cups in the control group. All outliers were >50°. These data indicated that the acetabular cups in the control group were occasionally placed too steep, likely because of the presence of acetabular dysplasia [20]. Correction of acetabular dysplasia by RAO may help surgeons place the acetabular cups in an adequate abduction angle. The acetabular cup position in the study group tended to be more lateralized than in the control group. Kaneuji et al [21] reported that the normal hip joint center was  $31.5 \pm 5$  mm lateral from the teardrop. While the acetabular cup position in our study seemed to be largely acceptable, it has been recognized that acetabular cups in the upper and lateral position may lead to poor results during THA [22-24]. The appropriate cup position depends on the position and shape of the acetabulum, and RAO prior to THA may influence the cup position. Thus, this should be evaluated both during the preoperative planning and intraoperatively.

Although anteversion of the acetabular component was not measured in the present study, signs of retroversion of the acetabulum after periacetabular osteotomy have been previously reported [11], and preoperative CT is effective for three-dimensional evaluations of the acetabulum and osteophyte (Fig. 2).

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a P < 0.05.

 $<sup>^{</sup>a}P < 0.05$ .

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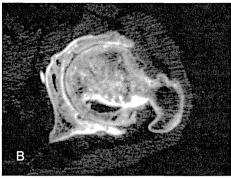




Fig. 2. The preoperative left hip of a 63-year-old woman with osteoarthritis after rotational acetabular osteotomy. (A) Anteroposterior radiograph showing progressive osteoarthritis changes. (B) Axial computed tomography image showing a large osteophyte of the acetabulum needing removal, and the depth of the acetabulum. (C) Three-dimensional computed tomography image showing the three-dimensional shape of the acetabulum.

In the present study, heterotopic bone formations after THA were seen more frequently in the study group (34.1%) than in the control group (10.3%). Similarly, previous studies reported that heterotopic bone formations were seen in 10%–42% of cases of THA after periacetabular osteotomy [10,11]. However, since most cases were classed as stage I or II in this study, heterotopic bone formations did likely not affect the clinical outcomes [25]. Moreover, we were concerned that wound healing problems would occur in the study group, since two separate incisions were performed, however, this did not occur in any case. We often used the combined approach in cases with poor range of hip motion or large osteophytes needing removal, and did not have to use trochanteric osteotomy in any case, suggesting that the approach is useful for exposing the anterior hip joint. Although we did not use any cemented devices or reinforced rings in this study, no component was loose. Thus, standard cementless devices appear to be useful in THA after RAO.

Previous studies have shown that RAO for dysplastic hips results in short-term hip pain relief and intermediate-term prevention of progression of arthritis, and the present study showed that the results of THA after RAO are equivalent to those of primary THA. Accordingly, we believe that RAO followed by THA could have long-term effects and result in long-term maintenance of the hip joint function for young patients with dysplastic hips, and longer follow-up studies are needed to confirm this.

In previous studies on the topic, the average time interval between a preceding periacetabular osteotomy and THA has been reported to range between 3.3 and 7.5 years [7,8,10,11]. In the present study, the average time interval between RAO and THA was 21 years. Thus, our results suggest that RAO may be a good procedure of joint preservation for dysplasia of the acetabulum. However, further large-scale studies are still needed in the future to investigate which periacetabular osteotomy procedure preserves the joint for the longest duration

This study has several limitations, including its retrospective nature and relatively short follow-up period. Moreover, three-dimensional radiographic evaluation was not performed.

In conclusion, although THA after RAO requires technical considerations, similar clinical outcomes to primary THA can be expected. However, further large-scale, long-term studies using three-dimensional radiographs are needed in the future to confirm our findings.

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# Potential Risk Factors of Persistent Low Back Pain Developing from Mild Low Back Pain in Urban Japanese Workers



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#### **Abstract**

Study Design: Two-year, prospective cohort data from the Japan epidemiological research of occupation-related back pain study in urban settings were used for this analysis.

Objective: To examine the association between aggravated low back pain and psychosocial factors among Japanese workers with mild low back pain.

Summary of Background Data: Although psychosocial factors are strongly indicated as yellow flags of low back pain (LBP) leading to disability, the association between aggravated LBP and psychosocial factors has not been well assessed in Japanese workers.

Methods: At baseline, 5,310 participants responded to a self-administered questionnaire including questions about individual characteristics, ergonomic work demands, and work-related psychosocial factors (response rate: 86.5%), with 3,811 respondents completing the 1-year follow-up questionnaire. The target outcome was aggravation of mild LBP into persistent LBP during the follow-up period. Incidence was calculated for the participants with mild LBP during the past year at baseline. Logistic regression was used to explore risk factors associated with persistent LBP.

Results: Of 1,675 participants who had mild LBP during the preceding year, 43 (2.6%) developed persistent LBP during the follow-up year. Multivariate analyses adjusted for individual factors and an ergonomic factor found statistically significant or almost significant associations of the following psychosocial factors with persistent LBP: interpersonal stress at work [adjusted odds ratio (OR): 1.96 and 95% confidence interval (95%CI): 1.00-3.82], job satisfaction (OR: 2.34, 95%CI: 1.21-4.54), depression (OR: 1.92, 95%CI: 1.00-3.69), somatic symptoms (OR: 2.78, 95%CI: 1.44-5.40), support from supervisors (OR: 2.01, 95%CI: 1.05-3.85), previous sick-leave due to LBP (OR: 1.94, 95%CI: 0.98-3.86) and family history of LBP with disability (OR: 1.98, 95%CI: 1.04-3.78).

Conclusions: Psychosocial factors are important risk factors for persistent LBP in urban Japanese workers. It may be necessary to take psychosocial factors into account, along with physical work demands, to reduce LBP related disability.

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#### Introduction

Low back pain (LBP) is a common musculoskeletal occupational health problem in industrialized countries and was found to be the leading specific cause of years lived with disability [1]. Japan is no exception, and LBP is one of the five most common health complaints of the Japanese general population [2]. Typically, 85-

90% of the cases are classified as 'non-specific' [3,4], and the majority of LBP is mild, so they do not become severely disabled [5,6]. However, in terms of cost and work loss, the small proportion of people who become disabled due to LBP account for the largest occupational health care cost and the greatest number of work days lost around the world [7,8]. Therefore,

clarifying potential risk factors that could aggravate the LBP condition and lead to disability to work would be very important.

Many epidemiological studies of LBP have been conducted worldwide for decades. Psychosocial factors such as low job satisfaction, depression, or the tendency to somatize have been strongly indicated as 'yellow flags' for LBP leading to disability, as have ergonomic factors such as physical work demands [8–11], although the magnitude or intensity of each factor may vary across cultures or work environments [12]. Based on the above evidence, recently in Japan psychosocial factors began to be considered as a major risk for aggravating LBP. However, to our knowledge, the association between aggravation of Japanese workers' back pain and psychosocial factors has not been thoroughly assessed in prospective epidemiological research studies.

Previously, we reported potential risk factors for new onset of back pain disability in Japanese workers enrolled in a prospective cohort study in urban settings [13]. Data regarding various potential risk factors at baseline, as well as LBP-related outcomes, were collected prospectively. The cohort study focused mainly on LBP that caused work disability, a subject of critical importance to employers as well as workers, in terms of occupational health care.

The present study was designed to ascertain whether various psychosocial factors are associated with aggravating mild LBP into persistent LBP in workers with a 1-year history of mild LBP, using data from the previously reported cohort study; the findings of this further data analysis are reported here. This study was part of a series of clinical research projects conducted by the Japan Labor, Health and Welfare Organization related to 13 fields of occupational injuries and illnesses, including musculoskeletal disorders, mental health, and cancer. The research projects were conducted to help resolve occupational health issues and to disseminate the findings.

#### **Materials and Methods**

#### Data source

Data were extracted from a prospective cohort of the "The Japan epidemiological research of Occupation-related Back pain (JOB)" study. Participants were recruited from 16 workplaces in various occupational fields, located in or near Tokyo. The major occupational groups at these workplaces were office workers, nurses, sales/marketing personnel, and manufacturing engineers. Each participating organization was asked to distribute a self-administrated questionnaire to their workers, along with a cover letter from the study administration office. Respondents were asked to return their completed questionnaires by post, including their names and mailing addresses, which were used to send follow-up questionnaires directly from the study administration office. A total of 6,140 baseline questionnaires were distributed during September 2005 and February 2006, and 5,310 completed questionnaires were returned (response rate: 86.5%).

The baseline questionnaire included questions about the severity of the respondent's LBP and various individual and work-related factors. LBP severity was evaluated by the respondents themselves, who were asked to quantify the severity into one of four grades: grade 0, no LBP; grade 1, LBP not interfering with work; grade 2, LBP interfering with work; and grade 3, LBP interfering with work and leading to sick leave. The grades were determined with reference to Von Korff's grading method [14]. LBP was defined as pain localized between the costal margin and the inferior gluteal folds [3], and the area was depicted in the questionnaire. The baseline questionnaire included questions about the following: individual characteristics, including gender, age, obesity, smoking habits, history of LBP, and previous sick

leave due to LBP; ergonomic work demands, such as frequency of bending, twisting or lifting at work; and psychosocial factors, such as depression, interpersonal stress at work, job control, job satisfaction, and somatization. A brief job stress questionnaire (BJSQ) was used to evaluate the major psychosocial factors [15,16]. The BJSQ is a self-administered scale having a total of 57 items, developed by a research working group organized by the Japan Labour, Health and Welfare Organization. Question items for the questionnaire were extracted from standard questionnaires commonly used for evaluating stress related factors, psychological stress response, depression, anxiety, and somatization [17-23]. The questionnaire was assessed using standardized scores, which were classified into 19 work-related stress factors: mental workload (quantitative aspect), mental workload (qualitative aspect), physical workload, interpersonal stress at work, environmental work stress, job control, utilization of skills and expertise, physical fitness, job satisfaction, vigor, irritability, fatigue, anxiety, depression, somatic symptoms, support from supervisors, support from co-workers, support from family or friends, and daily-life satisfaction. For each factor above, standardized scores were developed on a 5-point scale ranging from 1 (lowest) to 5 (highest) based on a sample of more than 10,000 Japanese workers. The questionnaire has demonstrated moderate reliability, high internal consistency, and its criterion validity has been assessed with respect to the Job Content Questionnaire (JCQ) and The National Institute for Occupational Safety and Health (NIOSH) [24].

The follow-up questionnaire was distributed 1 year after the baseline questionnaire was administered. Of the 5,310 participants who completed the baseline questionnaire, 3,811 successfully completed and returned the follow-up questionnaire, resulting in a follow-up rate of 71.8%. The follow-up questionnaire included questions relating to LBP, such as severity of LBP during the past year, length of sick-leave due to LBP, whether medical care was sought, pain duration, and onset pattern. LBP severity was assessed by the respondents themselves, using the same categories as those of the baseline questionnaire.

Ethical approval for the study was provided by the review board of the Japan Labour, Health and Welfare Organization. Informed consent was obtained in writing from all participants.

#### Data analysis

The outcome of interest was occurrence of persistent LBP during the 1-year follow-up period. In this study, persistent LBP was categorized as LBP interfering with work (grade 2 or grade 3), with disability lasting for longer than 3 months. Incidence was calculated for the participants who reported mild LBP (grade 1) during the past year at baseline. Participants were excluded from the analysis if they met any of the following criteria: a job change for reasons other than LBP; LBP due to a traffic accident; or LBP caused by a tumor, including metastasis, infection or fracture.

In addition to the compilation of simple, descriptive statistics, univariate and multivariate logistic regression analyses were used to explore risk factors associated with persistent LBP. Associations found by logistic regression analysis were summarized as odds ratios (ORs) with 95% confidence intervals (CIs). For the assessment of potential risk factors, crude ORs initially were estimated. Next, factors with P-values<0.1 were adjusted for individual factors and an ergonomic factor, in order to explore their potential risk factors. Factors with adjusted ORs that were statistically significant were considered to be potential risk factors. The following factors were used as adjusting factors because they are considered to be representative of individual and ergonomic factors: age, sex, obesity, smoking habits, education, and manual handling of

objects [25–27]. Additionally, the above psychosocial risk factors were grouped by their correlations to explore multicollinearity, and then a statistically significant factor that had the highest adjusted ORs were selected from each group and applied to multivariate regression analysis. Statistical significance was assumed at the 5% level if the 95% CI did not overlap 1. All statistical calculations were carried out using the STATA 9.0 software package.

#### Results

#### Baseline characteristics of study participants

Of the 3,811 participants who responded to the 1-year follow-up questionnaires, 1,675 (excluding 43 who did not answer the question on LBP severity on their follow-up questionnaire) reported mild LBP during the past year at baseline and met the selection criteria. The mean age was 43.1 years (SD 10.1 years) and 1,342 (78.6%) were male. The mean BMI was 23.1 kg/m² (SD 3.4 kg/m²). Of these participants, 1,165 (68.2%) were categorized as non-manual laborers; 147 (8.6%) as manual handlers of < 20-kg objects; 338 (19.8%) as manual handlers of  $\geq$  20-kg objects or as caregivers; and 58 (3.4%) were lacking job description data. In each category, the most common occupations were office work in the non-manual laborer category; manufacturing/engineering in the manual handler of < 20-kg objects or caregiver category.

The baseline characteristics of the 3,811 participants who provided follow-up data appeared to be not much different from those who did not. The mean (SD) ages were 42.9 (10.1) years and 38.0 (10.2) years, respectively, and the majority were male in both groups (80.6% and 82.8%, respectively). Those who completed the study had a mean (SD) BMI of 23.1 (3.3) while the values for dropouts were 22.9 (4.1). In the follow-up group (vs. the drop-out group), 78.6% (vs. 75.5%) were categorized as manually handling < 20-kg objects or not manually handling any objects in their work, 17.8% (vs. 18.9%) manually handled ≥ 20-kg objects or were working as caregivers, and data were lacking for 3.6% (vs. 5.6%). In both groups, the most common occupational fields in the categories of "manual handling of < 20-kg objects or not manually handling any objects", and "manual handling of ≥ 20-kg objects or working as a caregiver" were office worker and nurse, respectively.

#### Incidence of persistent LBP

Of the 1,675 eligible participants, 43 (2.6%) reported persistent LBP within the 1-year follow-up period. Of the 43 participants reporting persistent LBP, 76.7% had pain that persisted for longer than 6 months.

# Association between persistent LBP and potential risk factors

Crude ORs for persistent LBP, their 95% CIs, and P-values are shown in Table S1. The "somatic symptoms" risk factor was associated with an approximately 2.5-fold higher risk of suffering from persistent LBP. Associations of persistent LBP, with about a 2-fold risk increase, were also found with the following 5 psychosocial factors: interpersonal stress at work, job satisfaction, depression, support from supervisors, and daily-life satisfaction factors. An approximately 2-fold risk increase was found for the following 2 factors: previous sick-leave due to LBP and family history of LBP with work disability. Of the ergonomic factors, 7 (manual handling of objects at work, frequent bending, twisting, lifting, or pushing, hours of desk work, and physical workload)

were associated with about a 3- to 4-fold higher risk of developing persistent LBP. These 15 factors were chosen for multivariate logistic regressions, and the results are shown in Table 1. Most of the ergonomic factors were significant with the ORs adjusted for individual factors. Five factors from the BJSQ (interpersonal stress at work, job satisfaction, depression, somatic symptoms, and support from supervisors), as well as previous sick-leave due to LBP and family history of LBP with disability, remained statistically significant or almost significant by adjusted ORs. The magnitudes of adjusted ORs of these factors did not markedly change from our crude OR analyses. Among the 5 factors from the BISO, interpersonal stress at work, job satisfaction, and support from supervisors tended to correlate to each other, and depression and somatic symptoms tended to correlate to each other (Spearman's rho, data not shown). Additional multivariate regression analysis included job satisfaction and somatic symptoms from the BJSQ psychosocial factors and family history of LBP with disability, chosen by the statistical significance of the adjusted OR. As shown in Table 2, all of the factors remained statistically significant or almost significant in the multivariate analysis.

#### Discussion

Potential risk factors for people with LBP that could aggravate the condition and cause too much disability to work were explored in a cohort of urban Japanese workers. The incidence of persistent LBP developing from mild LBP was 2.6%. ORs adjusted for individual factors and an ergonomic factor (manual handling of objects) showed that low job satisfaction, lack of support from supervisors, interpersonal stress at work, depression, somatic symptoms, and a family history of LBP with disability were significant risk factors, and previous sick leave a nearly significant risk factor, for development of persistent from mild LBP. Our results indicate that these psychosocial factors are important in urban Japanese workers who have made the transition from mild to persistent LBP.

In this study, the definition of persistent LBP was disability longer than 3 months, and the index for disability was LBP interfering with work, with or without sick leave. In Western countries, 'absence from work' is often used as an outcome measurement for disability. The number of participants who were absent due to LBP (grade 3) was relatively small. Our previous international epidemiological study showed that taking sick leave due to musculoskeletal disorders, mostly LBP, appears to be less common among Japanese workers than British workers [28]. The lower percentage of absence due to LBP in Japanese workers compared to workers in European countries may be due to a difference in concerns about being absent, such as worries that it might affect employment, salary increases, or evaluations of work performance. In fact, the proportion of Japanese workers with disability irrespective of taking sick leave (sick leave defined as any unplanned absence from work) was approximately the same as the proportion of UK workers with sickness-related absences. Additionally, in another international cross-sectional study, the prevalence of disabling LBP varied markedly across countries, and the Japanese workers showed the lower prevalence than in other countries [29]. Therefore, when assessing Japanese workers, it seems appropriate to define LBP disability as LBP interfering with work, with or without sick leave.

Among the five factors from the BJSQ (low job satisfaction, little support from supervisors, interpersonal stress at work, depression, and somatic symptoms), low job satisfaction, little support from supervisors, and interpersonal stress at work tend to relate to each other, and depression and somatic symptoms tend to relate to each

**Table 1.** Adjusted odds ratios of the baseline factors for persistent low back pain (LBP) with work disability; factors with crude odds ratio P values < 0.1.

Factors		%	OR Adjusted for individual factors <sup>a</sup>		OR Adjusted for individual factors and an ergonomic factor <sup>b</sup>	
in the second of the second dis-	ASS THE CONTROL OF TH		OR	95%CI	OR	95%CI
Previous sick leave due to LBP	No previous sick leave	76.5	1.00		1.00	
	Previous sick leave	23.5	1.92	0.99-3.74	1.94	0.98–3.86
Manual handling of materials at work	Manual handling of < 20-kg objects including desk work	79.5	1.00			
dece is member in indi- principal beautiful montpressioners and	Manual handling of ≥ 20-kg objects or working as a caregiver	20.5	2.70	1.98–8.67		Para (1948) - prima matematic 1. Pistogra - Par J., para distance 1. Pistografia (1971), para distance
Bending <sup>c</sup>	Infrequent	88.7	1.00			
	Frequent	11,3	3.45	1.54–7.72	_	
Twisting <sup>c</sup>	Infrequent	94.6	1.00			
	Frequent	5.4	4.35	1.80–10.52	-	-
Lifting <sup>c</sup>	Infrequent	89.6	1.00			
	Frequent	10.4	2.81	1.18–6.66	20.	
Pushing <sup>c</sup>	Infrequent	95.2	1.00			
THE RESERVE OF THE PARTY OF THE	Frequent	4.8	3.48	1.24–9.76	- 10 A S	
Hours of desk work <sup>d</sup>	< 6 hours per day	53,9	1.00		1.00	
Especial languages and	≥ 6 hours per day	46.1	0.45	0.23-0.88	0.66	0.31-1.40
Physical workload <sup>e</sup>	No stress	61.9	1.00		1.00	
Maria masanah sa	Stress	38.1	2.22	1.16-4.23	1.53	0.70-3.33
Interpersonal stress at work <sup>e</sup>	No stress	78.8	1.00		1.00	
	Stress	21.2	2.04	1.06-3.93	1.96	1.00-3.82
Job satisfaction <sup>e</sup>	Satisfied	77,3	1.00		1.00	
	Not satisfied	22.7	2.48	1.31-4.70	2.34	1.21-4.54
Depression <sup>e</sup>	Not feeling depressed	64.6	1.00		1.00	
elikinde ene 1211 mont	Depressed	35.4	2.09	1.10-3.99	1.92	1.00-3.69
Somatic symptoms <sup>e</sup>	No somatic symptoms	63,4	1.00	200X	1.00	
	Somatic symptoms	36,6	2.99	1.55–5.75	2.78	1.44–5.40
Support from supervisors <sup>e</sup>	Support	74,0	1.00		1.00	
	No support	26.0	1.97	1.04–3.73	2.01	1.05–3.85
Daily-life satisfaction <sup>e</sup>	Satisfied	68.7	1.00		1.00	
Person of exercise St. 1 -	Not satisfied	31,3	1.81	0.97-3.40	1.61	0.84-3.08
Family history of LBP with disability	No LBP with disability	74.6	1.00		1.00	
	LBP with disability	25,4	2.02	1.07-3.81	1.98	1.04–3.78

OR: odds ratio, CI: confidence interval, LBP: low back pain

other. The first three factors (e.g., low job satisfaction) could be considered stressful conditions that directly and negatively affect the individual, and the latter two factors (e.g., depression) as symptoms of both physical and mental stress. Generally, the symptoms of somatization are headaches, neck and shoulder discomfort, dizziness, palpitations or shortness of breath, diarrhea or constipation, and back pain, and these symptoms are triggered by emotional discomfort and psychosocial distress [30]. Individuals

with somatization often complain of pain in various locations, functional disturbance of various organ systems, and are depressed or overwhelmed by these symptoms. Patients falling into such a situation are usually said to suffer from functional somatic syndrome (FSS) [31,32]. Our results could suggest that workers with mild LBP, under frazzled, depressed, or somatizing conditions, accompanied by emotional discomfort and psychosocial distress (e.g., low job satisfaction, little social support from

<sup>&</sup>lt;sup>a</sup>Adjusted for age, gender, obesity, smoking habits, and education.

Adjusted for age, gender, obesity, smoking habits, education, and manual handling of materials at work.

<sup>&</sup>lt;sup>c</sup>Bending, twisting, lifting, and pushing: ≥ half of the day was considered frequent.

dHours of desk work: longer than 6 hours per day was considered to be static posture.

eWork-related stress factors assessed with the brief job stress questionnaire: not feeling stressed, feeling stressed: the 5 original responses were reclassified into "not feeling stressed", where low, slightly low and moderate were combined, and "feeling stressed", where slightly high and high were combined. doi:10.1371/journal.pone.0093924.t001

Table 2. Multivariate-adjusted odds ratios for the persistent low back pain (LBP).

Factors		Adjusted OR <sup>a</sup>	95%CI	P value
Job satisfaction	Satisfied	1.00		THE RESERVE OF THE SECOND
VICE LEADING TO THE HEAT OF THE SET OF THE S	Not satisfied	2.03	1.01-4.07	0.046
Somatic symptoms	No somatic symptoms	1.00		
	Somatic symptoms	2.46	1.25-4.83	0.009
Family history of LBP with disability	No LBP with disability	1.00		
	LBP with disability	2.00	1.03-3.88	0.042

OR: odds ratio, CI: confidence interval, LBP: low back pain.

<sup>a</sup>Adjusted for individual factors (age, gender, obesity, smoking habits, and education) and an ergonomic factor (manual handling). doi:10.1371/journal.pone.0093924.t002

supervisors, and interpersonal stress at work), did not manifest disabling back pain as a symptom of FSS at baseline, but the pain became disabling during the following year.

A family history of persistent LBP was also suggested as a psychosocial risk factor in this analysis. Second-hand experience of LBP among people with whom a worker is in very close contact (families, friends, or partners) may make it easier to imagine how mild LBP transforms to persistent LBP. Previous research has revealed that some people can share another person's physical pain experience, in both emotional and sensory components, by just observing the other person's pain [33,34]. Family members, therefore, may provide reinforcement for sick behavior [35], even though these family members do not have had any disorders, such as back pain [36–39].

Psychosocial intervention has been reported to improve overall well-being, as well as reducing distress and physical complaints, in patients with LBP in Western countries [40]. This intervention is based on the hypothesis that psychosocial factors are associated with the transition to persistent LBP, and should be examined in future research studies in Japan.

Limitations of the current study should be mentioned. One is the fact that the majority of the subjects were males, and that a broad range of Japanese occupations was not represented. The study cohort was not a representative sample of the entire Japanese workers in urban areas; therefore, the generalizability of the findings may be limited. Secondly, although cognitive and emotional aspects of back pain are known to influence disability aggravation, some important psychosocial factors, such as the attitudes of health care providers, and catastrophizing and fear-avoidance beliefs, were not included in this analysis. This was because appropriate questionnaires were not available in the Japanese language. Future studies should include additional self-reported outcome measures, such as results of the Fear-Avoidance Belief Questionnaire (FABQ) [41,42] or the Tampa Scale of Kinesiophobia (TSK) [43,44], to assess the impact of these factors

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in Japanese workers. The Japanese versions of these questionnaires are now being developed.

Psychosocial factors are one of the most important risk factors for making the transition to persistent LBP from mild LBP in urban Japanese workers. In the future, preventive strategies for reducing persistent LBP in the workplace should deal not only with physical work demands, which is already well-understood, but potentially should incorporate psychosocial management techniques as well.

#### **Supporting Information**

Table \$1 Crude odds ratios of the baseline factors for persistent low back pain (LBP) with work disability. OR: odds ratio, CI: confidence interval, BMI: body mass index, LBP: low back pain. a Obesity: BMI of ≥ 25 is defined as obesity in Japan. <sup>b</sup> Smoking habits: Brinkmann index of ≥ 400 was defined as heavy smoker, calculated from the total number of cigarettes smoked per day multiplied by duration of smoking in years [45]. Working hours: ≥ 60 hours per week was assumed to be uncontrolled overtime. d Bending, twisting, lifting, and pushing: ≥ half of the day was considered frequent. e Hours of desk work: longer than 6 hours per day was considered as static posture. Work-related stress factors assessed with the brief job stress questionnaire: not feeling stressed, feeling stressed: the 5 original responses were reclassified into "not feeling stressed", where low, slightly low and moderate were combined, and "feeling stressed", where slightly high and high were combined. g Monotonous task: feelings of monotony or boredom at work. (DOC)

#### **Author Contributions**

Conceived and designed the experiments: K. Matsudaira HK K. Miyoshi. Performed the experiments: K. Matsudaira HK K. Miyoshi. Analyzed the data: K. Matsudaira TI KI. Wrote the paper: K. Matsudaira TI KI.

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#### ORIGINAL ARTICLE

# Psychometric properties of the Japanese version of the Fear-Avoidance Beliefs Questionnaire (FABQ)

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#### **Abstract**

Background The Fear-Avoidance Beliefs Questionnaire (FABQ) is useful for measuring fear-avoidance beliefs in patients with low back pain (LBP); however, no psychometrically validated Japanese version is available. The objective of this study was to evaluate reliability and validity of the Japanese version of the FABQ for use with Japanese workers with LBP.

Methods This was conducted as a web-based survey. Both confirmatory and exploratory factor analysis were performed to examine domain structure of the Japanese version of the FABQ. For reliability, internal consistency was assessed with Cronbach's alpha coefficient. For concurrent validity, correlation coefficients between the FABQ and the Pain Catastrophizing Scale (PCS) were calculated. For known-group validity, the relationship between FABQ score and clinical variables such as pain and depression was examined.

Results Analyses were based on responses of 1,786 adult Japanese workers with LBP. Factor analysis using the principal factor method with promax rotation revealed two factors, work and physical activity, in accordance with the domain structure of the original version of the scale. For reliability, acceptable internal consistency was demonstrated with Cronbach's alpha coefficient of 0.882 and 0.783 for each subscale. For concurrent validity, significantly moderate correlations were demonstrated between

FABQ subscales and PCS subscales (r = 0.30–0.39). For known-group validity, as hypothesized, significantly higher FABQ subscale scores were observed in workers who had stronger pain, who experienced routine work disability with sick leave, who experienced recurrence of LBP, and who had depressed mood.

Conclusions This analysis showed that the Japanese version of the FABQ is psychometrically reliable and valid to detect fear-avoidance beliefs in Japanese workers with LBP.

#### Introduction

Many patients with low back pain (LBP) experience fear of future pain. Patients with LBP who experience strong pain may avoid certain movements or physical activities because of exaggerated fears that pain will result in more functional restriction [1]. The repeating cycle of pain-related fear and avoidance behaviors can continue. Avoidance of physical activities based on fear-avoidance beliefs leads to further avoidance [2]. Little evidence has shown that avoidance behavior reduces chronic LBP either on a short- or long-term basis. Rather, fear-avoidance beliefs have been shown to play a contributing role in the development of long-term disability [3-5]. The Global Burden of Disease (GBD) studies done in 1990 and 2000 demonstrate that LBP is one of the leading specific causes of years living with disability (years of life lived in less than ideal health) [6]. A low level of fear avoidance is the most useful item for predicting earlier recovery [7, 8]. Avoidance of pain-inducing activities can result in reduced muscle strength and flexibility, which may partly contribute to a delay in recovery. Thus, chronic pain and disability may be perpetuated by fear-avoidance beliefs and behaviors.

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The Fear Avoidance Belief Questionnaire (FABQ), introduced by Waddell et al. [9], is a useful measure for assessing fear-avoidance beliefs in patients with LBP. The Multinational Musculoskeletal Inception Cohort Study Statement proposed that fear avoidance should be included in the core set of factors to be measured for prospective cohorts of patients with LBP, and the FABQ was recommended as the appropriate measure to detect fear-avoidance beliefs [10]. This 16-item self-report questionnaire can assess patients' beliefs about how physical activity and work affect their present LBP. Factor analysis in the development of the original version revealed two subscales: work (seven items) and physical activity (four items). The other five items are not included in the score calculation. Patients rate their agreement with each statement on a 7-point Likert scale (0 for completely disagree, 3 for unsure, 6 for completely agree). A subscale score is calculated by the simple sum of attribute item scores. Scores range 0-42 for the work subscale and 0-24 for the physical activity subscale. A higher score indicates stronger fear-avoidance beliefs or behaviors.

The FABQ was originally developed in English, and its reliability and validity have been demonstrated in British patients with LBP and/or sciatica [9]. It has been successfully translated into several languages and is widely used for evaluation in clinical studies [11–16]. However, a psychometrically validated Japanese version of the FABQ is not available. Therefore, we translated the original English version into Japanese and validated it linguistically, aiming to introduce the FABQ in Japan [17]. In this study, we performed a psychometric assessment of the Japanese version of the FABQ to evaluate its reliability and validity for use in Japanese patients with LBP.

#### Materials and methods

#### Validation sample

The data subset used in this psychometric testing was derived from an Internet survey used to collect information on Japanese people, including prevalence of LBP, work-related ergonomic characteristics, and attitudes/beliefs about LBP [18]. An Internet research company registered 1.8 million individuals who were 20–79 years old as monitors. These monitors were stratified by sex and age, and 1,063,083 monitors were randomly selected in accordance with Japanese demographic composition and invited to participate in research on LBP by an e-mail containing a link to the survey. Double registration as a monitor was prevented by checking e-mail addresses and by blocking access to the questionnaire once the responder completed the survey. Among the selected monitors,

77,709 individuals completed the survey, which resulted in a response rate of 7.31 %. Individuals whose reported age was <20 or >79 were excluded when calculating LBP prevalence, resulting in 65,496 participants. Of these, 3,220 Japanese workers who reported that they had experienced LBP within the previous 4 weeks were contacted by e-mail and invited to complete an online questionnaire. Under the assumption of a relatively low response rate (around 30 %), our goal was to obtain at least 1,000 completed questionnaires, and the survey was closed on the day the number of respondents exceeded 1,000.

Based on the consensus approach to back pain definition proposed by Dionne et al., LBP was defined as pain localized between the costal margin and the inferior gluteal folds that lasted for more than a day at any time during the past 4 weeks [19]. Pain associated only with menstrual periods, pregnancy, or during a course of a feverish illness was not included. A definition of LBP and a diagram of the affected area were provided within the questionnaire.

The questionnaire included the Japanese version of the Pain Catastrophizing Scale (PCS) [20, 21], a 13-item scale used to measure negative attitudes toward pain, involving rumination, helplessness, and magnification. The reliability and validity of the Japanese version were previously confirmed [21]. Total PCS scores range from 0 (no catastrophizing) to 52 (severe catastrophizing). The Mental Health (MH) domain of the Short-Form Health Survey of 36 questions (SF-36) and an 11-point numerical rating scale (NRS) were also included to assess MH and pain, respectively. The survey was approved by the medical/ethics review board of the Japan Labour Health and Welfare Organization. Personal identifiable information, including name, phone number, and permanent address, were not collected. Due to the nature of this study (web-based survey), no written informed consent was obtained; however, receiving an answered questionnaire was considered evidence of consent.

#### Data analysis

Demographic characteristics of the validation sample were summarized with simple descriptive analysis. In the item analysis, the percentage of missing responses was examined for each item. We also examined whether each item's response distribution was strongly skewed, that is, whether it had a floor or ceiling effect of  $\geq 60$  %. For construct validity, confirmatory factor analysis (CFA) using the principal factor method with a promax rotation was performed on the original two-factor model. Both Goodness of Fit Index (GFI) and Adjusted GFI (AGFI) of  $\geq 0.9$  are considered a reflection of good fit. Exploratory factor analysis (EFA) was also used as necessary. For convergent



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and discriminant validity, multitrait analysis was used. For each item, if the correlation coefficient between the score of the individual item and the subscale score to which that item was attributed (subscale score except for that item) is not extremely low, convergent validity is judged as acceptable. Also, for each item, if the correlation coefficient between the score of the individual item and the subscale score to which that item is attributed is greater than the correlation coefficients between the score of that item and the other subscale score to which that item is not attributed, then discriminant validity is judged as acceptable. With regard to internal consistency, the homogeneity of the items in each subscale was evaluated using Cronbach's alpha statistic. Cronbach's alpha coefficient of 0.7 or higher for both subscales is needed to claim that the FABQ is internally consistent [22]. Concurrent validity was evaluated using Spearman's rank correlation coefficient with the PCS. According to the criterion of correlation strength in the psychometric validation proposed by Cohen, the correlation coefficient was judged as follows: 0.1, weak correlation; 0.3, medium correlation; and 0.5, strong correlation [23]. For the known-group validity, relationships between selected variables and subscale scores were examined using the t test or analysis of variance (ANOVA), depending on the number of categories in a selected variable. If a statistically significant difference was found with ANOVA, then Tukey-Kramer multiple comparison was used to identify specific differences between pairs of groups. We hypothesized that workers who met the following attributes would show significantly higher FABQ scores: (1) workers with greater pain, (2) workers who experienced work disability with sick leave, (3) workers with more episodes of LBP, and (4) workers with depressed mood. In terms of pain, workers were categorized by the degree of pain as assessed using an NRS (0 = no pain to 10 = worst pain imaginable). The group in the first tertile was categorized as having slight pain, the second tertile as having moderate pain, and the third tertile as having severe pain. With regards to sick leave, if workers had to miss work due to LBP at least 1 day during 4 weeks, it is considered sick leave. In terms of the number of LBP episodes, if workers experience LBP after at least 1 month of being pain free, it is considered recurrence [24]. Depressed mood was assessed using the SF-36 Mental Health domain [25, 26]. A score of <52 was considered as depressed mood (range 0-100, low scores indicate more psychological distress) [27]. All statistical tests were two-tailed, and the level of significance was set at 0.05. Statistical calculations were performed using SAS version 9.2 (SAS Institute, Cary, NC, USA).

**Table 1** Clinical characteristics of the patient sample used for psychometric validation of the Japanese Fear-Avoidance Beliefs Questionnaire (N = 1,786)

Characteristics	Statistics	(%)
Sex		
Male	900	(50.4)
Age, year		
20–39	603	(33.8)
40–59	621	(34.8)
≥60	562	(31.5)
Educational background		
College/technical college/high school/ junior high	951	(53.2)
University or higher	825	(46.2)
Not applicable	10	(0.6)
Pain (NRS), mean $\pm$ SD	$2.9 \pm 2.3$	
Presence of disability		
No	544	(30.5)
Yes with no sick leave	801	(44.8)
Yes with sick leave	441	(24.7)
Job category		
White collar	687	(38.5)
Blue collar	273	(15.3)
Other	826	(46.2)
FABQ score, mean $\pm$ SD		
Work subscale	$16.3 \pm 9.8$	
Physical activity subscale	$14.9 \pm 4.7$	
PCS score, mean $\pm$ SD	$24.6 \pm 10.9$	)
MH subscale score in SF-36, mean $\pm$ SD	$55.1 \pm 20.9$	)

Unless otherwise specified, n (%) is shown

NRS numerical rating scale (0–10, higher score indicates greater pain), FABQ Fear-Avoidance Belief Questionnaire (0–42 for work subscale and 0–24 for physical activity subscale. A higher score indicates stronger fear-avoidance beliefs or behaviors), PCS Pain Catastrophizing Scale (0–52, a higher score indicates severe catastrophizing), MH Mental Health (0–100, a lower score indicates more psychological distress), SF-36 Short-Form Health Survey with 36 questions, SD standard deviation

#### Results

#### Patient background

Overall, 1,786 workers were analyzed, and their characteristics are shown in Table 1. Mean age was 48.7 years, and 50.4 % were men. FABQ scores [mean  $\pm$  standard deviation (SD)] were 16.3  $\pm$  9.8 for the work subscale and 14.9  $\pm$  4.7 for the physical activity subscale. Scores in the PCS and MH of the SF-36 were 24.6  $\pm$  10.9 and 55.1  $\pm$  20.9, respectively.



#### Item analysis

There was no missing response in any item. Neither floor nor ceiling responses were observed in the distribution of responses, although skewed distribution was found in items 8, 15, and 16 (responses for completely disagree were 47.9, 48.6, and 55.2 %, respectively).

#### Factor analysis

CFA was performed on the original two-factor model, and GFI and AGFI were 0.84 and 0.76, respectively, indicating that there was no evidence of good fit. Thus, further assessment was performed using EFA with promax rotation. As a result, the eigenvalue was >1 with the two-factor model (i.e., 1.36), and all items were clearly regressed to the same factors as the original version, with factor-loading values >0.4 (Table 2). In addition, the result of multitrait analysis demonstrated satisfactory convergent and discriminant validity (Table 3). For these reasons, we consequently adopted the two-factor model. Also, no floor/ceiling effects were observed in the subscales (9.7 and 1.0 % for the work subscale; 1.5 and 5.2 % for the physical activity subscale).

#### Reliability

Cronbach's alpha coefficient was 0.882 for the work subscale and 0.783 for the physical activity subscale, indicating sufficient internal consistency.

#### Concurrent validity

Correlation coefficients between FABQ subscales and the PCS were calculated to examine concurrent validity. The work subscale moderately correlated with the PCS total score, the helplessness subscale, and the magnification subscale ( $r=0.38,\ 0.39,\$ and  $0.34,\$ respectively; P<0.0001 for all). The physical activity subscale moderately correlated with the PCS total score, the rumination subscale, and the magnification subscale ( $r=0.36,\ 0.36,\$ and  $0.30,\$ respectively; P<0.0001 for all).

#### Known-group validity

The relationship with variables that may affect the FABQ score was examined. As hypothesized, significantly higher FABQ subscale scores were observed in workers with stronger pain, workers who experienced work disability with sick leave, workers who experienced recurrence of LBP, and workers with depressed mood (Fig. 1).

Table 2 Rotated factor pattern (standardized regression coefficient)

Item	Factor 1	Factor 2
Factor 1: fear-avoidance beliefs about work		
6 My pain was caused by my work or by an accident at work	0.611	-0.005
7 My work aggravated my pain	0.770	0.023
9 My work is too heavy for me	0.760	-0.041
10 My work makes or would make my pain worse	0.904	-0.002
11 My work might harm my back	0.884	0.019
12 I should not do my normal work with my present pain	0.669	0.087
15 I do not think that I will be back to my normal work within 3 months	0.444	0.014
Factor 2: fear-avoidance beliefs about physical	l activity	
2 Physical activity makes my pain worse	0.017	0.799
3 Physical activity might harm my back	0.013	0.843
4 I should not do physical activities which (might) make my pain worse	-0.009	0.633
5 I cannot do physical activities which (might) make my pain worse	0.028	0.493

Table 3 Convergent and discriminant validity: correlation coefficients for question items and domain score (Spearman's correlation)

Item	Factor 1	Factor 2
Factor 1: fear-avoidance beliefs about work		
6 My pain was caused by my work or by an accident at work	0.565	0.217
7 My work aggravated my pain	0.720	0.306
9 My work is too heavy for me	0.711	0.230
10 My work makes or would make my pain worse	0.786	0.328
11 My work might harm my back	0.771	0.338
12 I should not do my normal work with my present pain	0.659	0.336
$15\ I$ do not think that I will be back to my normal work within 3 months	0.417	0.166
Factor 2: fear-avoidance beliefs about physical act	ivity	
2 Physical activity makes my pain worse	0.326	0.577
3 Physical activity might harm my back	0.335	0.625
4 I should not do physical activities which (might) make my pain worse	0.228	0.563
5 I cannot do physical activities which (might) make my pain worse	0.233	0.416

Subscale scores were computed excluding the scores for items within a factor



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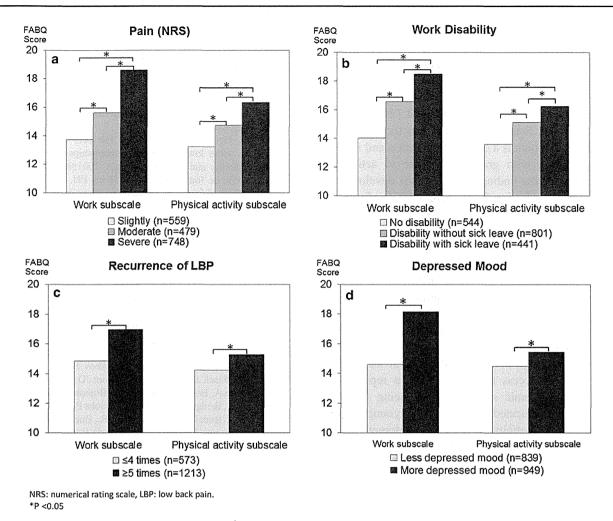


Fig. 1 Known-group validity: Fear-Avoidance Beliefs Questionnaire scores and associated variables: P values were calculated by Tukey-Kramer method for pain and routine work disability, and the t test was used to evaluate recurrence of low back pain (LBP) and depression

#### Discussion

Before the analysis performed in this study, we proposed a linguistically validated Japanese version of the FABO [17], which was assured by following a standardized manner for developing a translated questionnaire [28]. In the study reported here, we assessed its psychometric properties. In the factor analysis conducted in a confirmatory manner, goodness of fit indicators did not satisfy the preset level. However, EFA revealed a two-factor solution consistent with the original questionnaire. In general, it is preferable that a translated version of a specific questionnaire maintains the same domain structure as the original version to enable comparison of data derived from different translated versions. Taking this into account, a two-factor model of the FABQ (work and physical activity subscales) was finally adopted in the Japanese version. Our decision in selecting the two-factor model was supported by the fact that reliability of the measure was demonstrated. As an index to assess reliability, a sufficient internal consistency with Cronbach's alpha statistic of 0.882 and 0.783, respectively, for the subscales was demonstrated. Internal consistency in the Japanese version was considered sufficient to be in agreement with the original version (Cronbach's alpha of 0.88 and 0.77) [9].

For known-group validity, as hypothesized, relevance was observed between FABQ score and variables that might affect the scores, including the degree of pain, work disability with sick leave, recurrence of LBP, and depressed mood. Similarly, in the development of the original version, significant correlations of FABQ were observed for pain severity (r = 0.23 for the work subscale), work loss in the past year (r = 0.55 for the work subscale, 0.23 for the physical activity subscale), and depressive symptoms (r = 0.41 and 0.34, respectively) [9].

Avoidance behavior led by fear and avoidance beliefs in patients with LBP contributes to the development of chronic disability. In fact, fear-avoidance behavior was



shown to be an important risk factor for chronicity. Thus, encouraging patients to change their beliefs and behaviors has become more important in managing LBP, especially in the early phase. In recent guidelines for managing nonspecific, acute LBP, continuing normal daily activity is recommended and bed rest is discouraged [29]. To help reduce pain-related fear, it is important to focus on educating patients that pain is a common condition and is self-manageable, along with gradual exposure to activities, rather than on imaging findings leading to the development of fear-avoidance behavior. The FABQ enables clinicians to detect patient fear-avoidance beliefs and helps to establish an effective management program to prevent chronic LBP on an individual basis.

Limitations of this study should be noted. The use of the Internet to recruit participants could have contributed to selection bias, although the large sample size that this method allowed is a major strength of this study. However, we must also consider the issue of sample representativeness in this study. Our validation population might not represent a wide range of workers throughout the nation. Workers who can access the Internet might represent a particular socioeconomic status, such as being possibly wealthier, better educated, or relatively younger. We decided to recruit participants using the Internet, taking into account both costs and feasibility. Our strategy may invite criticism regarding generalizability of the results. Although results demonstrating the sufficient psychometric properties of the Japanese version of the FABO as a measure might partly support the adequacy of the sample, it is still possible that good psychometric properties could have been demonstrated in an unrepresentative sample. Concerning the use of online questionnaires, the comparability of online testing to paper and pencil forms in regards to psychometric properties has been demonstrated [30, 31]. It should be noted that test-retest reliability over certain time intervals remains unknown. Also, responsiveness cannot be assessed in this study due to the crosssectional nature of the data. Future studies would be helpful to assess the test-retest reliability and responsiveness of the Japanese version of the FABQ.

Results of the analysis presented here show that the Japanese version of the FABQ is psychometrically reliable and valid as a measure to detect fear-avoidance beliefs in a Japanese population with LBP. The Japanese version of the FABQ has the same domain structure as the original version (work and physical activity subscales), enabling comparisons with data derived using the original version.

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Conflict of interest None.

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