Hip-Spine Syndrome: the Coronal Alignment of the Lumbar Spine and Pelvis in Patients with Ankylosed Hips

Tadatsugu Morimoto, Motoki Sonohata, Masaru Kitajima, Tomohito Yoshihara, Hirohito Hirata, and Masaaki Mawatari

Department of Orthopaedic Surgery, Saga University, 5-1-1 Nabeshima, Saga, Japan

Name: Tadatsugu Morimoto
Address: Nabeshima 5-1-1, Saga 849-8501, Japan
Tel: +81-952-34-2343
E-Mail: sakiyuki0830@gmail.com

All authors declare that there are no conflicts of interest
There is no conflict of interest with any financial organization regarding the material discussed in this manuscript.

Tadatsugu Morimoto wrote and prepared the manuscript, Motoki Sonohata participated in the writing or technical editing of the manuscript, Masaru Kitajima, Tomohito Yoshihara, and Hirohito Hirata collected data, and Masaaki Mawatari served as a scientific advisor.

This study was approved by the institutional ethics committee of Saga University Hospital (Approval code: 2015-12-13).

None

This is a retrospective, observational study carried out using the opt-out method via our hospital website.
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Abstract

Introduction

Fixed abduction and/or adduction deformities of the hip joint may cause pelvic obliquity with subsequent development of secondary lumbar scoliosis. However, the relationships between the magnitude of a fixed angle (either abduction or adduction) of the hip and the direction of pelvic tilt and lumbar scoliosis remain unclear. The purpose of this study was to investigate the coronal alignment of the lumbar spine and pelvis in patients with ankylosed hips.

Methods: A total of 56 patients were analyzed, including 17 males and 39 females, with an average age of 65 years (range: 45 to 80 years). Regarding the coronal spinopelvic alignment, the following parameters were measured: the degree of lumbar scoliosis (LS; Cobb angle), pelvic obliquity (PO), and ankylosed hip angle (AHA). The PO and AHA were defined as the angle between the inter-teardrop line and a horizontal line, respectively, and the long axis of the femur on the side of the ankylosed hip. For each parameter, correlations between the parameters were evaluated using a regression analysis. A P value of <0.05 was considered significant.

Results: Positive linear correlations were observed between the AHA and direction of the PO angles ($r = 0.831$, $p < 0.01$), the AHA and direction of the LS angles ($r = 0.770$, $p < 0.01$), and the directions of the PO and LS angles ($r = 0.832$, $p < 0.01$).

Conclusions: This study provides evidence to suggest that, in patients with ankylosed hips, the abduction position is positively correlated with the downward PO and the convexity of the LS toward the AH side.
contrast, the adduction position is positively correlated with these results on the opposite side.

Key words: Hip-spine syndrome, Spino-pelvic alignment, Ankylosed hip, Lumbar scoliosis, Pelvic obliquity
INTRODUCTION

Fixed adduction deformity due to contracture resulting from hip osteoarthritis may cause pelvic obliquity (PO) with the subsequent development of secondary lumbar scoliosis. This well-known phenomenon was first defined by Offierski and MacNab as secondary hip-spine syndrome¹. However, the relationships between the magnitude of a fixed hip angle (either abduction or adduction) and the impact of pelvic tilt and lumbar scoliosis remain unclear. Although ankylosed hips are associated with better outcomes than hip joints with contracture, as an ankylosed hip joint is painless and fused, no studies have investigated these associations in patients with ankylosed hips. The objectives of this study were to investigate the coronal alignment of the lumbar spine and pelvis in patients with ankylosed hips.

MATERIALS AND METHODS

Patients

Seventy-two patients (75 hips) treated with total hip arthroplasty for an ankylosed hip at Saga University Hospital between January 2003 and March 2012 were registered in this study. An ankylosed hip was defined as the bony union of the hip joint with trabeculae crossing the resection space and/or transformation of bone architecture, resulting in complete immobility. Sixteen patients (19 hips) were deemed ineligible due to a lack of evaluable data resulting from inadequate radiographs (4 hips in 4 patients), the presence of bilateral ankylosed hips (8 hips in 5 patients) or a history of total or bipolar hip arthroplasty on the opposite side (6 hips in 6 patients and 1 hip in 1 patient, respectively). Therefore, a total of 56 eligible patients (17 males, 39 females) were analyzed.

The affected side was the right in 23 patients and the left in 33 patients, with no significant difference in the side of the ankylosed hip. The mean age of participants was 65 years (range, 45–80 years), and all had a history of spontaneous (n = 24) or surgical (n = 32) fusion of the hip joint. The initial diagnosis was tuberculosis in 13 cases, bacterial infection in 9 cases, post-trauma in 11 cases, developmental hip dysplasia in
20 cases, idiopathic osteonecrosis of the femoral head in 2 cases, and a slipped capital femoral epiphysis in 1 case. The mean duration of ankylosis prior to total hip arthroplasty was 38 years (range, 7–70 years).

Plain radiographic evaluations

Antero-posterior and lateral radiographs of the pelvic and thoracic and lumbar spine were obtained for all patients in the erect posture at our institute. During the radiographic examination, all patients were instructed to stand upright in a relaxed, natural posture.

The parameters measured were lumbar scoliosis (LS), PO and the ankylosed hip angle (AHA) in the coronal plane (Fig. 1 a, b). The degree of LS was measured using the Cobb method in the coronal plane, with LS defined as a curve measuring greater than 10°. The apical level, defined as the point of the most laterally deviated vertebra in a scoliosis curve, was determined. LS was marked as positive in calculations when the convexity faced the ankylosed hip side and negative when the convexity faced contralaterally. The degree of PO was defined as the angle between the horizontal line and the line passing through the inferior tip of the bilateral pelvic teardrops (also known as the U-figure). PO was marked as positive in calculations when the downward direction was toward the ankylosed hip side and negative when the downward direction was toward the opposite side. The AHA in the coronal plane was defined as the angle between the perpendicular angle of the line passing through the inferior tip of bilateral pelvic teardrops and the long axis of the femur on the side of the ankylosed hip. This variable was calculated with abduction regarded as a positive angle and adduction regarded as a negative angle.

In this study, we diagnosed “windswept hip-spine deformity” when both the abduction position correlated with a downward motion of PO and convex LS was noted toward the ankylosed side (Fig. 2 a) or when the adduction position correlated with these findings on the opposite side (Fig. 2 b).
The sensitivity and likelihood ratios were determined to correspond with windswept hip-spine deformity when limited to either an abduction or adduction of >10° in the AHA. The positive likelihood ratio (LR+) was defined as “sensitivity / (1 – specificity)” and reflected an increase in the odds of having windswept hip-spine deformity when the value was positive (LR+ >1). The negative likelihood ratio (LR−) was defined as “(1 – sensitivity) / specificity” and reflected a decrease in the odds of having windswept hip-spine deformity when the value was negative (LR− of 0–1). Tests with LR+ >10 or LR− < 0.1 are usually considered suitable for application in routine practice2).

Statistical analyses

The Statistical Package for Social Sciences (SPSS) software program, version 19 (IBM SPSS, Chicago, IL, USA) was used for the statistical analyses of data. For each parameter, correlations between parameters were evaluated using Student’s $t$-test, the chi-squared test and Pearson correlation coefficients. Statistical significance was set at a value of $p < 0.05$. Correlation coefficients were evaluated as follows: <0.25, little-to-no significant correlation; 0.25–0.5, relatively weak correlation; >0.5, relatively strong correlation3,4).

Measurements were obtained twice, once each by two assessors on separate days. The intra-class correlation coefficient (ICC) was used to assess the intra- and inter-examiner reliability of the LS, PO and AHA values.

Estimates of the mean intraobserver reliability for measurements of the LS, PO, and AHA were 0.72, 0.83 and 0.82, respectively. The mean interobserver reliability for measurements of LS, PO, and AHA were 0.71, 0.84 and 0.78, respectively. ICC values were considered poor for values <0.40, good for values of 0.40–0.75, and excellent for values > 0.75 [3,4]. The intra- and interobserver reliability ranged from good to excellent, so parameters rated by one investigator in a single evaluation of all patients were used in this study.
RESULTS

Lumbar scoliosis

Twenty-nine patients (52%) had scoliosis, with a single curve in the thoracolumbar range in 27 patients and a double curve in 2 patients. When such a double curve existed, the largest curve was considered in our analysis. Of the 29 patients with scoliosis, the apical vertebra of the largest curve varied from L1 to L4, with L3 as the median apical vertebra; the apex was L1 in 3 patients, L2 in 10 patients, L3 in 11 patients, and L4 in 5 patients.

Correlations between coronal parameters

Positive linear correlations were observed between the AHA and the direction of PO ($r = 0.831$, $p < 0.01$; Fig. 3a), the AHA and the direction of LS ($r = 0.770$, $p < 0.01$; Fig. 3b) and the directions of PO and LS ($r = 0.832$, $p < 0.01$; Fig. 3c).

In short, a larger AHA value on abduction was associated with larger PO and LS values on the ankylosed side. In contrast, a larger AHA value on adduction was associated with larger PO and LS values on the opposite side. In addition, the PO value was proportional to the LS value on both the ankylosed side and contralaterally.

Among the entire cohort, 11 cases with PO and 10 cases with LS did not meet the criteria for windswept hip-spine deformity. However, using the cut-off point of abduction or adduction AHA $> 10^\circ$, only 1 case with PO and no cases with LS did not meet the criteria for windswept hip-spine deformity. Regarding the association between windswept hip-spine deformity and the AHA, when limited to abduction or adduction of the AHA $>10^\circ$, the values of LR$^+$ were 10 and 4.38 and LR$^-$ were 0.36 and 0.16, respectively, in patients with abduction and adduction (Table 1, 2).
Representative case: Ankylosed hip on abduction (Fig. 1 a, b)

The patient was a 77-year-old man with a chief complaint of lumbago and knee pain who was found to have an ankylosed hip on abduction due to infection. The LS, PO and AHA values were 20°, 12° and 28°, respectively. Convex lumbar scoliosis and downward pelvic tilt were observed toward the ankylosed side.

DISCUSSION

The etiology and biomechanics of PO and LS derived from hip deformities are multifactorial. These conditions can be caused by contractures of the hip, leg length discrepancy, muscle imbalances, and uneven weight-bearing due to hip pain, particularly while walking. With regard to contractures of the hip, the effects of either abduction or adduction contractures on both the affected and contralateral hips must be taken into account. In addition, knee flexion of the longer limb and/or plantar flexion of the foot of the shorter limb can arise as compensatory mechanisms. Furthermore, the biomechanics of PO and LS derived from hip deformities are often complicated by the coexistence of these conditions. Therefore, although previous studies have reported that contractures of the hip are often accompanied by pelvic tilt and LS, evidence regarding a relationship between these factors is contradictory and inconclusive\(^5\sim\!^9\).

Regarding patients with neuromuscular disorders, Letts et al.\(^6\) and Abel et al.\(^7\) reported that a subluxed hip was more likely to occur on the elevated side of the pelvis, opposite the apex of scoliosis. However, Hodgkinson et al.\(^8\) and Lonstein et al.\(^9\) found no relationships between hip subluxation, PO and scoliosis. The above studies were associated with several limitations in assessing the relationship between the magnitude of a fixed hip angle (either abduction or adduction) and the direction of PO and LS, as contractures of the hip joint are often painful, and the joint is not fused. The present study, therefore, included patients with unilateral anklylosed hips because of the lack of pain and presence of fusion, excluding those with bilateral...
The present study provides evidence suggesting that the abduction position correlates positively with a downward motion of PO and convex LS toward the ankylosed side, while the adduction position correlates positively with these results on the opposite side. This study is the first to validate the occurrence of secondary hip-spine syndrome, in which fixed deformities of the hip joint result in PO with subsequent development of secondary lumbar scoliosis, as proposed by Offierski and MacNab in 1983. We defined this phenomenon as “windswept hip-spine deformity”.

When limited to abduction or adduction AHA >10°, only one case did not meet the criteria for windswept hip-spine deformity. Both LR+ and LR− in patients with abduction or adduction >10° in the AHA showed reliable values. Therefore, the cut-off point for abduction or adduction >10° in the AHA might be a strong factor influencing the occurrence of windswept hip-spine deformity. However, in patients with abduction or adduction <10° AHA, other factors, such as pre-existing spinal deformity, leg length discrepancy, lumbago or knee pain, or internal or external rotation of the AHA, might have a greater influence on the occurrence of windswept hip-spine deformity than the abduction or adduction of the AHA.

Windswept hip-spine deformity correlated well with the clinical impression that an inadequate position in patients with an ankylosed hip results in compensatory PO and LS in order to maintain proper body balance. If such conditions are induced by continuous PO and LS due to an inadequate position of an ankylosed hip, the patient may develop structural changes in the spine and pelvis, inducing severe low back pain and leading to secondary conversion to total hip arthroplasty (THA). Performing total hip arthroplasty in patients with ankylosed hips, especially those with malposition (either excessive abduction or adduction), may therefore help prevent degenerative spinal changes and relieve low back pain. Eguchi et al. pointed out that improvements in PO and LS following THA in patients with severe hip osteoarthritis may be
related to improvements in back pain following THA.

In contrast, some patients with structural scoliosis may develop problems with balance after THA for ankylosed hips. Abe et al.\(^1\) reported that coronal balance in flexible scoliosis in patients with degenerative hip osteoarthritis, even in some ridged scoliosis cases, tended to improve after THA. The long-term effects of THA on ankylosed hips should be investigated in future studies.

Several limitations associated with the present study must be noted. First, this cross-sectional study was limited with respect to distinguishing \textit{de novo} scoliosis from preexisting deformities and, therefore, did not allow for analyses of the temporal sequence of events. Second, although the director of impetus for PO and/or LS may derive from shortening of the hip abduction, this study did not assess the effects of leg-length discrepancy because measuring leg-length discrepancy is difficult in patients with ankylosed hips, particularly those with malposition and knee flexion and/or plantar flexion of the foot as a compensatory mechanism. Third, this study did not evaluate the sagittal spino-pelvic alignment, which has recently been essential to such studies. We neglected to do so because it was difficult to assess the center of the hip accurately in most ankylosed hip cases. A final potential limitation associated with this study is the fact that, although we used thoracic, lumbar and pelvic X-ray findings, we did not assess the overall spinal alignment with whole-spine radiographs. While the most frequently involved level of spinal deformities derived from hip disease and/or PO may be at the lumbar level, because the most apical vertebra of the curve varied from L1 to L4 in our study, exploring this issue in greater detail will require future studies to assess the effects of hip disease on the lumbar and overall spinal alignment using whole-spine radiographs.

\textbf{CONCLUSION}

This study provides evidence suggesting that, in patients with AH, the abduction position correlates positively
with downward PO and convexity of the LS toward the AH side. In contrast, the adduction position correlates positively with these results on the opposite side.

Conflicts of interest

No benefits in any form have been received or will be received from a commercial party related directly or indirectly to the subject of this article.
REFERENCES


FIGURES AND TABLE LEGENDS

Table 1. Likelihood ratios for windswept hip-spine deformity in cases of abduction over 10°

Table 2. Likelihood ratios for windswept hip-spine deformity in cases of adduction over 10°

Figure 1. A 77-year-old man with right ankylosed hip on abduction due to infection.

a) X-ray of the lumbar spine.

b) X-ray of the pelvis and femur.

Convex lumbar scoliosis and downward pelvic obliquity are observed toward the ankylosed side.

*Pelvic obliquity, defined as the angle between the horizontal line and the line passing through the inferior tip of bilateral pelvic teardrops (white arrow). **Ankylosed hip angle, defined as the angle between the perpendicular angle of the line passing through the inferior tips of the bilateral pelvic teardrops (white arrow) and the long axis of the femur on the side of the ankylosed hip.

Figure 2. “Windswept spine-hip deformity” illustration

a) Abduction ankylosed hip case (white arrow; ankylosed hip)

b) Adduction ankylosed hip case (white arrow; ankylosed hip)

“Windswept hip-spine deformity” was defined when either the abduction position correlated with a downward motion of pelvic obliquity and convex lumbar scoliosis was noted toward the ankylosed side (a) or when the adduction position correlated with these findings on the opposite side (b).
Figure 3. a) The correlation between the ankylosed hip angle and the direction of pelvic obliquity. b) The correlation between the ankylosed hip angle and the direction of lumbar scoliosis. c) The correlation between the direction of pelvic obliquity and the direction of lumbar scoliosis. The correlation coefficients between the ankylosed hip angle and direction of pelvic obliquity (a), ankylosed hip angle and direction of lumbar scoliosis (b) and direction of pelvic obliquity and direction of lumbar scoliosis (c) were 0.831, 0.770 and 0.832, respectively (P < 0.01). Pelvic obliquity was marked as positive in calculation when the downward direction was toward the ankylosed hip side and negative when the downward direction was toward the opposite side. The ankylosed hip angle was calculated with abduction regarded as a positive angle and adduction as a negative angle.
Table 1. Likelihood ratios for windswept hip-spine deformity in cases of abduction over 10°

<table>
<thead>
<tr>
<th>AHA&lt;-10° (n = 18)</th>
<th>Likelihood ratios</th>
</tr>
</thead>
<tbody>
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<td>number</td>
<td>Sensitivity</td>
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<tr>
<td>Abduction (n = 35)</td>
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</tbody>
</table>

CI, confidence interval; AHA, ankylosed hip angle
Table 2. Likelihood ratios for windswept hip-spine deformity in cases of adduction over 10°

<table>
<thead>
<tr>
<th>Adduction (n = 21)</th>
<th>Number</th>
<th>Sensitivity</th>
<th>LR+ (95% CI)</th>
<th>LR− (95% CI)</th>
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</thead>
<tbody>
<tr>
<td></td>
<td>7</td>
<td>88%</td>
<td>4.38 (1.69–11.36)</td>
<td>0.16 (0.04–0.68)</td>
</tr>
</tbody>
</table>

CI, confidence interval; AHA, ankylosed hip angle
Fig. 3a

Ankylosed hip angle

Pelvic Obliquity

$r = 0.831$

$P < 0.01$

Fig. 3a
Fig. 3b

Lumbar scoliosis vs. Ankylosed hip angle

$r = 0.770$

$P < 0.01$
Pelvic obliquity

Lumbar scoliosis

Fig. 3c

r 0.832
P < 0.01