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Ⅲ　研究成果の刊行物・別刷

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Sagittal Alignment of the Subaxial Cervical Spine After C1-C2 Transarticular Screw Fixation in Rheumatoid Arthritis

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Abstract: Several articles reported the association between the development of subaxial kyphosis and the hyperlordotic fixation of C1-C2. However, their patients were heterogeneous in both primary disease and operative procedure. Transarticular screw fixation has become a popular procedure for C1-C2 arthrodesis instead of wiring techniques in which C1-C2 is difficult to fix in the intended alignment. Furthermore, in rheumatoid arthritis (RA) patients, subaxial lesions play an important role in potential subaxial alignment changes. The subaxial influences after C1-C2 transarticular screw fixation in patients with RA are unclear. To investigate the radiographic features of the subaxial cervical spine after C1-C2 transarticular screw fixation for RA, we reviewed 28 cases of C1-C2 transarticular screw fixation for rheumatoid atlanto-axial subluxation. The sagittal alignment of C1-C2 and the subaxial cervical spine was measured and the factors that affect subaxial alignment were investigated. Subaxial alignment became less lordotic in the postoperative course. The C1-C2 fixation angle and subaxial alignment showed a negative linear correlation. However, no significant correlation was found between changes in the C1-C2 angle and changes in the subaxial alignment. Four patients had a postoperative kyphotic subaxial deformity. Neurologic deterioration recurred in 4 patients, because of the postoperative development of subaxial subluxation. Common radiographic changes included an increase in C1-C2 lordosis, constant inclination of C1, an anterior shift of C2, and a decrease in C2-C7 lordosis. Many factors, not only C1-C2 angle, are associated with subaxial sagittal alignment change after C1-C2 transarticular screw fixation.

Key Words: rheumatoid arthritis, C1/C2, atlanto-axial subluxation, fusion, Magerl procedure, subaxial alignment, subaxial subluxation

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The upper cervical spine is well known to be affected by various kinds of pathologies and often must be fused surgically to relieve the cord compression and/or intractable pain. Despite excellent results of C1-C2 posterior arthrodesis, several authors[1,2] have reported postoperative malalignment of the subaxial cervical spine after the procedure. Yoshimoto et al[2] reported that 42% of the patients who underwent C1-C2 arthrodesis showed the progression of kyphosis in the subaxial cervical spine, and that C1-C2 fixation in a hyperlordotic position led to subaxial kyphosis after the surgery. This conclusion, however, is of limited significance, because these previous studies involved a variety of surgeries and diseases including rheumatoid arthritis (RA). Posterior wiring techniques were formerly the standard procedures to fuse C1-C2.[3–6] Tightening of a wire passed around the laminae of C1 and C2 tended to approximate these laminae and often impose a hyperlordosis. Recently, however, transarticular screw fixation has become a popular procedure for C1-C2 arthrodesis in place of wiring techniques.[7,8] C1-C2 can be aligned more freely by this procedure than by wiring techniques. In patients with RA, subaxial cervical alignment is affected not only by C1-C2 fusion but also by subaxial lesions including subaxial subluxation (SAS), commonly recognized and often progressive among patients with RA. There has been no detailed study of subaxial changes after C1-C2 transarticular screw fixation in patients with RA. We retrospectively investigated subaxial changes after C1-C2 transarticular screw fixation exclusively in patients with RA.

MATERIALS AND METHODS

Between 1995 and 2001, 35 patients with rheumatoid atlanto-axial subluxation underwent C1-C2 transarticular screw fixation supplemented with posterior wiring or lamina clamping. Patients who underwent concomitant surgery on the subaxial region were excluded from the study. Two patients died within 2 years after the

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surgery: one from myocardial infarction and the other from brain infarction. Five patients were lost to the minimum 2-year follow-up; thus, the remaining 28 patients (4 men and 24 women) provided the data in this study. A monocortical autograft from the iliac crest was used in each patient. The mean age of patients at surgery was 60.2 years (range, 34 to 74 y) and the mean follow-up period was 6.0 years (range, 2 to 10 y) after the surgery. All patients fulfilled the American Rheumatism Association criteria for RA. The indication for surgery was intolerable neck pain and/or cervical myelopathy caused by C1-C2 subluxation. Blinded measurement of sagittal spinal curve was conducted by one of the authors (Y.M.), who was not directly involved in the surgeries nor in the follow-up of patients using a modified Harrison posterior tangent method.[9] Radiographs, taken with the patient in the upright or sitting position, were scanned using a high quality image scanner and the digital data were transmitted to a computer work station, where measurement was performed using a software program (PopImaging Version 3.5; Digital Being Kids, Kanagawa, Japan). The bony landmarks to be identified on lateral neutral radiographs were all postero-superior and postero-inferior vertebral body corners of C2-C7, the mid-C1 anterior arch, and the mid-C1 posterior arch. Relative rotational angle (RRA_{Cm-Cn}) is the segmental angle derived from posterior vertebral tangents passing through the 2 points of the posterior vertebral body corners of Cm and Cn (Fig. 1). RRA_{C1-C2} was obtained from the angle between the posterior vertebral tangent of C2 and the perpendicular to the line through the centers of the anterior and posterior arches of C1 (Fig. 1). RRA was defined to have a negative (−) value in segmental lordosis and a positive (+) value in segmental kyphosis. The absolute rotation angle (ARA_{C2-C7}) was defined as the summation of the angles from RRA_{C2-C3} to RRA_{C6-C7}. The atlas plane angle (APA) was the angle derived from the horizontal line and the line passing through the centers of the anterior and posterior arches of C1 (Fig. 2). Negative (−) APA meant C1 in an extension position with respect to the horizontal plane. The horizontal translation of C2 from C7, which was defined as the distance from the postero-superior body corner of C2 to a vertical line through the postero-inferior body corner of C7, was termed Tz_{C2-C7} (Fig. 2). The RRA from C1-C2 to C6-C7, the ARA_{C2-C7}, the APA, and the Tz_{C2-C7} were measured on lateral neutral radiographs before surgery, 1 month after surgery, 1 year after surgery, and at the final follow-up. Subaxial vertebral slippage (≥ 3 mm) was regarded as significant SAS if accompanied by rheumatoid lesions such as erosive changes in facet joints, intervertebral discs, endplates, or spinous processes.

Neurologic impairment was evaluated using the Ranawat classification (class I, no neural deficit; class II, subjective weakness with hyperreflexia and dysesthesia; class IIIA, objective weakness and long tract signs but ambulating; and class IIIB, objective weakness and long tract signs and not ambulating).[10] Neck pain was also classified using Ranawat grading (0, none; 1, mild; 2, moderate; and 3, severe).[10] Clinical assessment was conducted just before the surgery, 1 year after the surgery, and at the final follow-up. The relationship between clinical status and the change in subaxial cervical alignment was also investigated.

Paired *t* test, Student *t* test, analysis of variance, and correlation coefficient methods were used for statistical analysis. Probability values less than 0.05 were considered statistically significant. Analyses were performed using JMP statistical computer software, version 5.0 (SAS Institute, Cary, NC).

RESULTS

Radiographic Evaluation

C1 Inclination

The mean APA before surgery was -11.2 ± 12.0 degrees; 1 month after surgery, it was -15.9 ± 8.8 degrees; 1 year after surgery, it was -12.9 ± 8.5 degrees; and at the final follow-up, it was -12.0 ± 9.2 degrees. The C1 inclination came into a more extended position with respect to the horizontal line just after the surgery and it returned to its preoperative inclination subsequently. However, there was no statistical difference among these values.

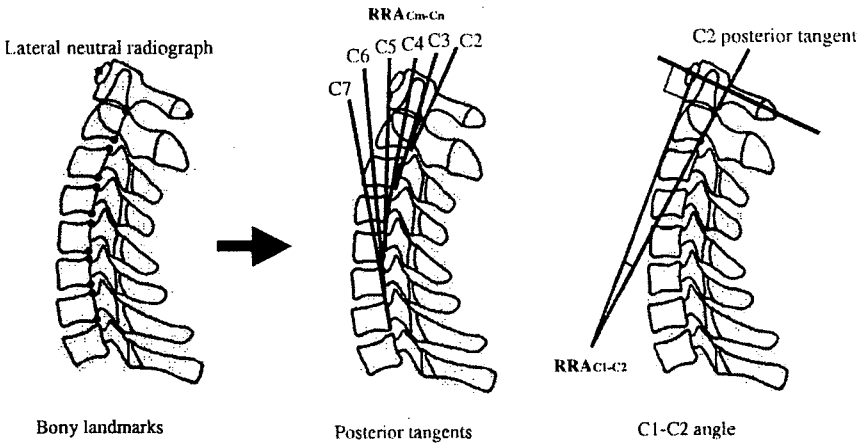

FIGURE 1. Relative rotational angle (RRA_{Cm-Cn}).

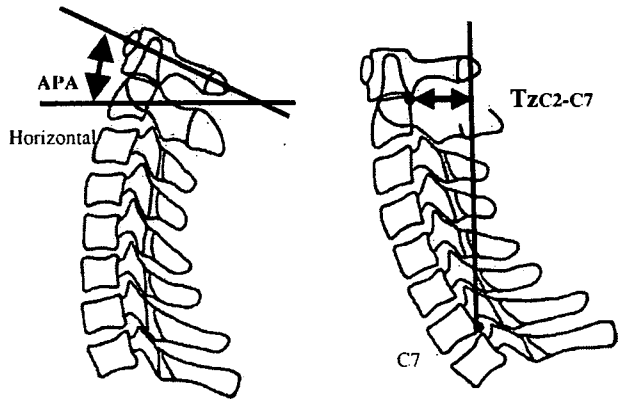

FIGURE 2. Atlas plane angle (APA) and horizontal translation ($Tz_{C2\text{-}C7}$).

C1-C2 Alignment

The mean $RRA_{C1\text{-}C2}$ before surgery was -6.0 ± 11.9 degrees; 1 month after surgery, it was -13.1 ± 9.8 degrees; 1 year after surgery, it was -12.7 ± 9.5 degrees; and at the final follow-up, it was -13.5 ± 9.4 degrees. The lordosis in C1-C2 was significantly increased by surgery (-6.0 vs. -13.1 degrees, $P = 0.0016$) and showed no significant change in the postoperative course. There was a positive linear correlation between preoperative and postoperative $RRA_{C1\text{-}C2}$ ($R = 0.535$, $P = 0.0028$) (Fig. 3A); the more lordotic the preoperative C1-C2 alignment was, the more lordotic the C1-C2 fixation angle became.

Horizontal Translation of C2

The mean horizontal translation ($Tz_{C2\text{-}C7}$) before surgery was 3.3 ± 13.0 mm; 1 month after surgery, it was 8.4 ± 12.4 mm; 1 year after surgery, it was 9.7 ± 12.4 mm; and at the final follow-up, it was 12.9 ± 15.0 mm. C2 progressively shifted forward with respect to C7 postoperatively ($P = 0.0030$).

Subaxial Alignment

No patients had SAS and only 1 patient had a positive value of $ARA_{C2\text{-}C7}$ on a lateral neutral radiograph preoperatively. The mean $ARA_{C2\text{-}C7}$ before surgery was -20.2 ± 13.2 degrees; 1 month after surgery, it was -19.5 ± 13.6 degrees; 1 year after surgery, it was -15.5 ± 13.0 degrees; and at the final follow-up, it was -13.3 ± 15.6 degrees. The subaxial cervical spine became less lordotic with increasing postoperative time ($P = 0.0293$). The $RRA_{C1\text{-}C2}$ one month after surgery, which was also the C1-C2 fixation angle, and the $ARA_{C2\text{-}C7}$ one year after surgery showed a negative linear correlation ($R = -0.516$, $P = 0.0043$) (Fig. 3B); the more lordotic the alignment in which C1-C2 was fixed by surgery, the less lordotic the subaxial alignment became postoperatively. However, no significant correlation was found between the operative changes of $RRA_{C1\text{-}C2}$ and the postoperative changes of $ARA_{C2\text{-}C7}$ 1 year after surgery ($R = -0.180$, $P = 0.362$) (Fig. 4A) or at the final follow-up ($R = 0.070$, $P = 0.731$) (Fig. 4B) (Fig. 5). Postoperative positive conversion of $ARA_{C2\text{-}C7}$, which indicates the development of subaxial kyphosis, was found in 4 patients (14%) during the follow-up. In 3 of them, the kyphosis was closely related to the deterioration of preoperative rheumatoid subaxial lesions, such as erosive changes of the facet or endplate. Postoperative development of SAS was noted in 5 patients (18%), all of whom had some degree of preoperative subaxial lesions preceding subluxation (Fig. 6). No statistical association was found between postoperative $RRA_{C1\text{-}C2}$ and the postoperative development of SAS.

Clinical Evaluation

Preoperatively, 25 patients (89%) had neck pain; all of them indicated pain relief by 1 or 2 grades of Ranawat category 1 year after the surgery. During the follow-up period, neck pain deteriorated in 5 patients; in 2 of these patients, the development of subaxial kyphosis was noted.

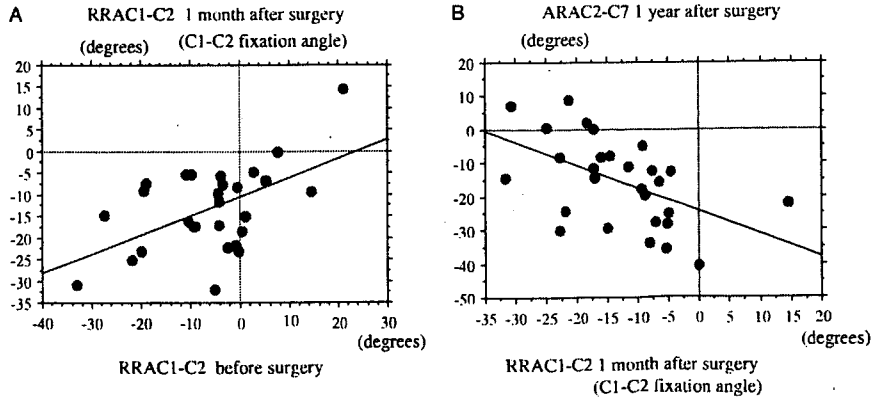

FIGURE 3. A, Correlation between preoperative and postoperative C1-C2 angles ($R = 0.535$, $P < 0.01$). B, Correlation between C1-C2 fixation angle and subaxial alignment 1 year after surgery ($R = -0.516$, $P < 0.01$).

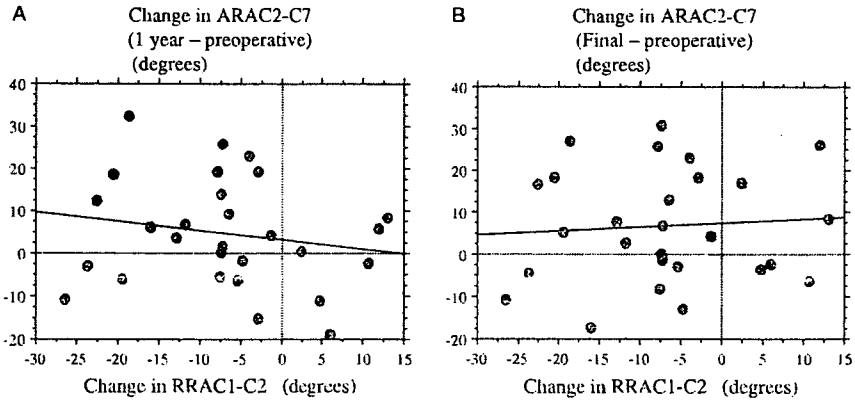

FIGURE 4. A, Correlation between the operative changes of C1-C2 alignment and the changes of subaxial alignment before surgery and 1 year after surgery ($R = -0.180$, $P = 0.362$). B, Correlation between the operative changes of C1-C2 alignment and the changes of subaxial alignment before surgery and at the final follow-up ($R = 0.070$, $P = 0.731$).

Preoperative neurologic status was class I in 5 patients, class II in 14 patients, class IIIA in 5 patients, and class IIIB in 4 patients. Of the 9 patients with neurologic status in class IIIA or class IIIB, 7 showed postoperative improvement by one or more grades of Ranawat class, although the remaining 2 with class IIIB status had no neurologic improvement. Neurologic status deteriorated in 4 patients during the postoperative follow-up, because of the development of SAS. Postoperative alignment change did not result in neurologic worsening by itself, unless SAS was also present.

DISCUSSION

RA commonly involves the cervical spine with potential risks for pain or progressive myelopathy in patients with long-standing RA. Disorders related to RA occur predominantly in the upper cervical region, where surgical treatments have been extensively documented.[11–13] C1-C2 arthrodesis has been traditionally performed using posterior wiring techniques, which may have some difficulties with nonunion or loss of correction between C1 and C2.[3–6] Recent articles[14] report high fusion rates with few complications associated with transarticular screw fixation procedure introduced by Magerl and Seemann.[7] However, we often encounter a subaxial alignment change, which can be a cause of neurologic impairment after C1-C2 arthrodesis. Previous studies on subaxial changes after C1-C2 fusion are of little use in predicting those changes for patients with RA, because those studies enrolled patients with various etiologies of C1-C2 instability. Subaxial lesions can develop not only as a natural course of RA[15–17] but also as a consequence of upper cervical fusion or disruption of the extensor muscles involved in posterior cervical surgery. For these reasons, we chose to focus our study

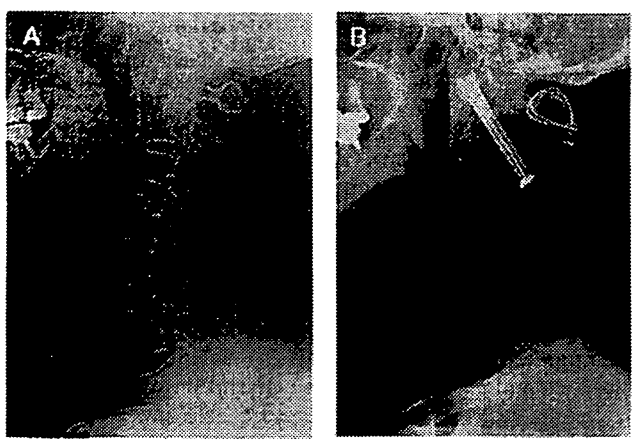

FIGURE 5. A, Preoperative lateral neutral radiograph of 52-year-old woman. B, C1-C2 angle decreased by 7 degrees postoperatively. Ten years after surgery, there was only 7-degree increase in $ARA_{C2\text{-}C7}$.

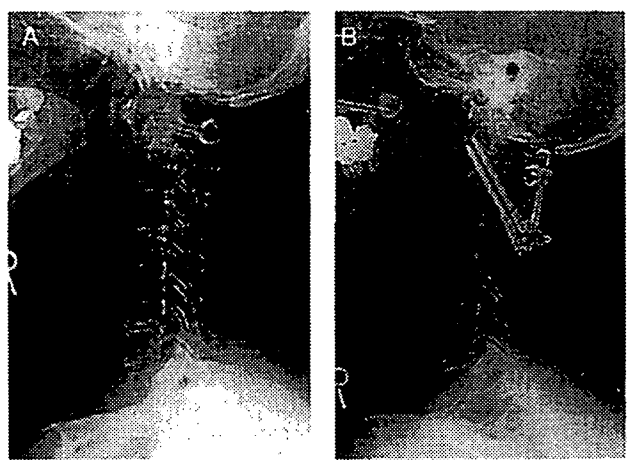

FIGURE 6. A, Preoperative lateral neutral radiograph of 56-year-old woman. Slight subluxation and facet erosion were found at C4-C5. B, C1-C2 angle decreased by 7 degrees postoperatively. Four years after surgery, marked subluxation and angular deformity were developed at C4-C5.

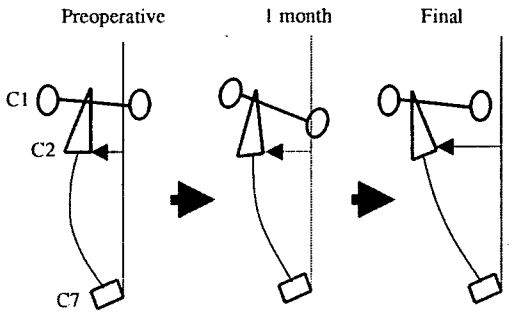

FIGURE 7. Radiographic features after C1-C2 transarticular screw fixation in patients with RA were an increase in C1-C2 lordosis, an anterior shift of C2, a decrease in C2-C7 lordosis, and the tendency of C1 inclination to return to preoperative inclination.

on C1-C2 transarticular screw fixation for rheumatoid atlanto-axial subluxation. There has been no detailed study of the subaxial changes after C1-C2 transarticular screw fixation in patients with RA.

From our study, the radiographic changes after C1-C2 transarticular screw fixation in patients with RA are summarized as follows: (1) an increase in C1-C2 lordosis, (2) a progressive ventral shift of C2 relative to C7, (3) a progressive decrease in C2-C7 lordosis, and (4) a tendency of C1 inclination to return to preoperative inclination (Fig. 7). When radiographs are taken with patients in a neutral position, patients are asked to face straight ahead. Preoperative alignment of the skull-C1 complex in a neutral position may be a natural alignment for individual patient. The postoperative increase of C1-C2 lordosis, which caused an increase in C1 inclination, may have been compensated by a decrease in subaxial cervical lordosis so as to return the C1 inclination to the preoperative one. Actually, in our study, there was a negative linear correlation between the postoperative alignments of C1-C2 and C2-C7, which is consistent with the previous report[2] (Fig. 3B). On the other hand, no significant correlation was found between the operative changes of $RRA_{C1\text{-}C2}$ and the postoperative changes of $ARA_{C2\text{-}C7}$ (Figs. 4A, B). This is opposite to the conventional idea that the more the lordosis in C1-C2 increases from surgery, the more the subaxial kyphosis that develops. The postoperative increase in C1 inclination owing to an increase in C1-C2 lordosis may be counterbalanced by not only the subaxial cervical alignment but also by the thoracolumbar spine alignment to keep the C1 inclination constant, but current study has no data about that.

Postoperative kyphosis in the subaxial cervical spine, which was noted in 4 of the 28 patients (14%), did not develop as often as previously considered and was closely associated with the deterioration of preexisting subaxial lesions. The mean C1-C2 fixation angle of these patients was −20.5 degrees (range, −17 to 25 degrees), which was not excessively lordotic. Although the C1-C2 fixation angle was less lordotic than the preoperative one in 7 patients (25%), 2 of them had a concomitant decrease in subaxial lordosis. In these patients, factors other than the C1-C2 fixation angle might play an important role in subaxial alignment. Previous studies[18,19] have reported that cervical alignment after posterior surgery is significantly affected by detachment of the semispinalis muscles from the spinous process of the axis. Because of the ligament weakness inherent to RA, muscle dissection can also accelerate subaxial lesions and may lead to subaxial kyphosis.

In this study, development of instability at C2-C3, which was adjacent to the fused segment, was not found. The greatest postoperative kyphosis or SAS developed at C4-C5 or C3-C4, accompanied by the deterioration of subaxial lesions.

So far, only the C1-C2 fixation angle has been emphasized as a key to regulate postoperative subaxial alignment in C1-C2 arthrodesis. In patients with RA, however, subaxial alignment is regulated by multiple factors: the C1-C2 angle, the development and/or progression of subaxial lesions, and the disruption of extensor muscles. These factors are difficult to evaluate correctly and subaxial change after C1-C2 arthrodesis for RA is hard to predict. Although we could not demonstrate the optimal C1-C2 fixation angle from this study, we do not necessarily deny the argument that C1-C2 fixation in excessive lordotic or kyphotic alignment should be avoided to prevent subaxial malalignment after the surgery. Regarding neurologic status, alignment change without SAS did not by itself cause neurologic worsening, whereas compression myelopathy caused by SAS was the main cause for postoperative neurologic deterioration.

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