Effect of a reduction of the atlanto-axial angle on the cranio-cervical and subaxial angles following atlanto-axial arthrodesis in rheumatoid arthritis

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Abstract

Purpose We retrospectively investigated the radiographic findings in patients with atlanto-axial subluxation (AAS) due to rheumatoid arthritis, and clarified the effect of reduction of the atlanto-axial angle (AAA) on the cranio-cervical and subaxial angles.

Methods Forty-one patients, consisting of 29 females and 12 males, with AAS treated by surgery were reviewed. The average patient age at surgery was 61.0 years, and the average follow-up period was 4.0 years. We investigated the AAA at the neutral position in lateral cervical radiographs before surgery and at the last follow-up. In addition, we also investigated the clivo-axial angle (CAA) and the subaxial angle (SAA) at the neutral position before and after surgery.

Results Due to pre-operative AAA, the patients were classified into three groups as follows: (1) the kyphotic group (K group), (2) the neutral group (N group), and (3) the lordotic group (L group). The average AAA values at the neutral position in the K group before and after surgery were 6.0° and 18.1°, respectively ($P < 0.001$). In the N group 19.7° and 21.7°, respectively ($P < 0.05$), and in the L group 31.6° and 27.0°, respectively ($P < 0.01$). However, no significant differences in the average CAA values were found before and after surgery in all groups. Furthermore, no significant differences in the SAA values were seen before and after surgery in all groups.

Conclusions A proper reduction of the AAA did not affect the cranial angles or induce kyphotic malalignment of the subaxial region after atlanto-axial arthrodesis. However, if we can obtain a significant and large reduction of AAA in patients showing kyphosis before surgery, then this reduction will be offset in the atlanto-occipital joint and we should therefore pay special attention to its morphology after surgery.

Keywords Atlanto-axial subluxation · Rheumatoid arthritis · Atlanto-axial angle · Reduction

Introduction

The pathophysiology of rheumatoid arthritis (RA) involves inflammation of the synovial membrane. In particular, the atlanto-axial complex is frequently affected. Paus et al. [1] assessed the mortality rate in a cohort of operated and non-operated patients, and noted that RA with neck involvement is a progressive and serious condition with reduced lifetime expectancy. The common features of this instability consist of atlanto-axial subluxation (AAS) and vertical subluxation (VS), or a combination of both. In particular, AAS is the most frequent deformity, representing approximately 65 % of all cervical subluxations [2]. Matsunaga et al. [3] noted that all of the irreducible AAS patients who did not undergo surgical treatment were bedridden within 3 years after the onset of myelopathy, and the survival rate was 0 % in the first 8 years. In AAS patients, the degree of displacement between the atlas and axis appears quite variable in lateral radiographs taken at the neutral position. Oda et al. [4] noted that upper cervical lesions progressed from reducible AAS to irreducible AAS with VS. They also noted that this extent of progression

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was different with the RA subset, which was also related to the development of subaxial subluxation. Iizuka et al. [5] noted that fusion or ankylosis of the atlanto-occipital joint (AOJ) induced an enlargement of the atlanto-dental interval (ADI) and anterior inclination of the atlas in the neutral position, despite the fact that normal findings of AOJ showed a slight displacement of the atlas to axis in RA patients showing AAS involvement. Sorimachi et al. [6] noted that a destructive abnormality of subchondral bone in the atlanto-axial joint (AAJ) induced an enlargement of the ADI and anterior inclination of the atlas in AAS patients with RA.

Reducible AAS without any other cervical instabilities, such as VS or subaxial subluxations (SAS), can be managed successfully by atlanto-axial arthrodesis. Atlanto-axial transarticular [7, 8] screw fixation has been the gold standard for treatment of this condition. At present, a combination of lateral mass screws for C1 and pedicle screws for C2 [9, 10] are becoming popular. However, in either method, it is important to acquire reduction of displacement between C1 and C2. In C1–C2 transarticular screw fixation, performing reduction before screw placement and fusion is essential, whereas in C1–C2 lateral mass, performing pedicle screw construct reduction before placing the screw is not an essential prerequisite and can be done after placing the screw as well. Regarding the relationship between the atlanto-axial angle (AAA) and subaxial alignment following atlanto-axial arthrodesis, some authors [11–15] noted that fixation of the AAA in a hyperlordotic position will lead the lower cervical spine to a kyphotic sagittal alignment after surgery. Ishii et al. [11] noted that a decrease in post-operative lordosis resulting from excessive correction of the AAA in patients with atlanto-axial arthrodesis causes post-operative subaxial subluxation requiring additional long fusion surgery. However, the effect of reduction of the AAA to the lower upper cervical spine in atlanto-axial arthrodesis has not yet been fully described. Furthermore, its effect on the skull region has not been described elsewhere. This study retrospectively investigated radiographic findings in patients with AAS due to RA and clarified the effect of reduction of the AAA on the cranio-cervical and subaxial angles.

Materials and methods

Forty-five patients with AAS treated by surgery at the university hospital between 2001 and 2010 were reviewed. Two patients died of causes unrelated to surgery within 1 year. One patient developed pseudoarthrosis and required revised surgery. Forty-one of the remaining 42 cases were thus reviewed, consisting of 29 females and 12 males. The duration of RA ranged from 3 to 39 years, with a mean

duration of 16.2 years. The average patient age at surgery was 61.0 years (range 34–78 years). The average follow-up period was 4.0 years (range 1–10 years). We included the patients showing reducible AAS; therefore, patients with irreducible AAS were excluded. We did not encounter any major complications in the patients without pseudoarthrosis, as described previously. AAS was diagnosed by a lateral cervical radiograph showing an anterior atlanto-dental interval (ADI) on a flexion radiograph of 3 mm or more. In patients with cervical myelopathy with atlanto-axial instability, surgery was usually indicated. In addition, when patients complained of only neck pain after unsuccessful adequate conservative treatment, they were also recommended to undergo surgery. The research protocol of this study was approved by our Institutional Review Board.

We performed selective atlanto-axial arthrodesis (transarticular screw fixation) for all patients. Regarding our strategy to reduce AAA, several investigators [12, 14] have mentioned that the optimum AAA for C1–2 arthrodesis should be approximately 20°, we therefore considered $20^\circ \pm 5^\circ$ to be acceptable. Thereafter, patients were classified into three groups as follows: (1) the kyphotic group (K group), in which the AAA was less than 15° (15 patients); (2) the neutral group (N group), in which the AAA ranged between 15 and 25° (15 patients); and (3) the lordotic group (L group), in which the AAA was greater than 25° (11 patients). These findings are summarized in Table 1. In general, in K group, we attempted to acquire a reduction of the displacement between the atlas and axis during surgery; in N group, we attempted to maintain the AAA; in L group, we attempted to reduce, or at least maintain the AAA, since the hyperlordotic fixation of the AAA has been reported to change the kyphotic lower cervical alignment [11–15]. We attempted to acquire a suitable AAA using an image intensifier before inserting the screw.

In all patients, we retrospectively investigated the AAA at the neutral position in lateral cervical radiographs before surgery and at the last follow-up (Fig. 1a). We also investigated the clivo-axial angle (CAA) (Fig. 1b), and the subaxial angle (SAA) (Fig. 1c) before and after surgery. All parameters were analyzed using the paired *t* test, Wilcoxon signed-ranks test, ANOVA, Kruskal–Wallis test, and Sheffe's *F* test. A value of $P < 0.05$ was considered to be significant.

Results

The pre-operative average CAA value in the K, N, and L groups was 151.7°, 154.0°, and 152.3°, respectively. The pre-operative average SAA value in those groups was 20.7°, 14.5°, and 18.4°, respectively (Table 2). There was

Table 1 Summary of the background data of 3 groups

	Group K (<i>n</i> = 15)	Group N (<i>n</i> = 15)	Group L (<i>n</i> = 11)
Gender (female/male)	10/5	13/2	9/4
Age	63.4 ± 9.7	56.2 ± 12.0	64.1 ± 5.0
Duration of RA (years)	17.2 ± 8.5	15.2 ± 10.4	16.1 ± 4.2
ADI (mm)	12.8 ± 2.6	10.8 ± 2.3	11.7 ± 2.3
Duration of follow-up (years)	3.9 ± 1.8	4.5 ± 2.5	3.4 ± 1.7

no difference among the three groups in pre-operative CAA ($P > 0.55$) or SAA values ($P > 0.45$).

The average AAA values at the neutral position in the K group before and after surgery were 6.0° and 18.1°, respectively. There was a significant difference in the angles before and after surgery ($P < 0.001$). Those values

in the N group were 19.7° and 21.7°, respectively, which were also significantly different before and after surgery ($P < 0.05$). The L group ranged from 31.6° and 27.0° before and after surgery, respectively. There was a significant difference between the angles before and after surgery ($P < 0.02$).

The average CAA values at the neutral position in the K group before and after surgery were 151.7° and 155.1°, respectively ($P > 0.08$); in the N group 154.0° and 157.7°, respectively ($P > 0.07$); and in the L group 152.3° and 153.0°, respectively ($p > 0.45$). None of the average CAA values were significantly different before and after surgery.

The average SAA values at the neutral position in the K group before and after surgery were 20.7° and 17.6°, respectively ($P > 0.08$); in the N group 14.5° and 12.4°, respectively; ($P > 0.35$); and in the L group 18.4° and 16.4°, respectively ($P > 0.57$). None of the average SAA values were significantly different before and after surgery.

Fig. 1 Method used for radiological measurement.
a Atlanto-axial angle (AAA);
b clivo-axial angle (CAA);
c subaxial angle (SAA)

Table 2 Summary of the radiological data of 3 groups

	Group K	Group N	Group L
AAA			
Pre-op	$6.0^{\circ} \pm 8.2^{\circ}$	$19.7^{\circ} \pm 2.6^{\circ}$	$31.6^{\circ} \pm 4.3^{\circ}$
Post-op	$18.1^{\circ} \pm 5.0^{\circ}$	$21.7^{\circ} \pm 2.9^{\circ}$	$27.0^{\circ} \pm 2.6^{\circ}$
P value	<0.001	<0.05	<0.02
CAA			
Pre-op	$151.7^{\circ} \pm 10.2^{\circ}$	$154.0^{\circ} \pm 8.7^{\circ}$	$152.3^{\circ} \pm 9.5^{\circ}$
Post-op	$155.1^{\circ} \pm 9.5^{\circ}$	$157.7^{\circ} \pm 10.3^{\circ}$	$153.0^{\circ} \pm 5.8^{\circ}$
P value	>0.08	>0.07	>0.45
SAA			
Pre-op	$20.7^{\circ} \pm 16.7^{\circ}$	$14.5^{\circ} \pm 12.0^{\circ}$	$18.4^{\circ} \pm 14.0^{\circ}$
Post-op	$17.6^{\circ} \pm 16.4^{\circ}$	$12.4^{\circ} \pm 13.0^{\circ}$	$16.4^{\circ} \pm 15.0^{\circ}$
P value	>0.08	>0.35	>0.45

Discussion

Toyama et al. [14] reported that 33 % of all patients who underwent atlanto-axial arthrodesis developed post-operative kyphosis or swan-neck deformity of the lower cervical spine, which was attributable to fixation of the atlanto-axial joint in a hyperextended position. Yoshimoto et al. [15] also noted that C1–C2 fixation in the hyperlordotic position would lead the subaxial cervical spine to kyphotic alignment; therefore, the greatest care must be taken to accurately determine the C1–C2 fixation angle during surgery. Furthermore, Ishii et al. [11] noted that excessive correction of the AAA in atlanto-axial arthrodesis is likely to cause a loss of cervical lordosis, thus resulting in the development of post-operative SAS. Nojiri et al. [16] evaluated 330 asymptomatic volunteers to establish standard values for the normal alignment of the upper cervical spine and noted that the mean AAA was $26.5 \pm 7^{\circ}$ in males and $28.9 \pm 6.7^{\circ}$ in females. Yoshimoto et al. [15] noted that the optimal C1–2 fixation angle may be quite variable among individuals because physiologic sagittal alignment of the cervical spine differs from person to person; however, some authors [12, 14] reported that a post-operative AAA of 20° may thus be a landmark angle.

According to these reports, we have given special attention to determining the suitable AAA in atlanto-axial arthrodesis. In the current study, although we tried to maintain the AAA after surgery in the N group, post-operative AAA was significantly increased compared with before surgery. We could not completely control AAA, in particular, we sometimes encountered a loss of reduction when inserting the screw. However, the mean values after surgery were mostly similar to the angle some authors advocated, and in fact did not affect post-operative CAA and SAA. In the K group, we acquired a significant and large reduction of the AAA after surgery, however, the

CAA and SAA did not show significant differences after surgery. Particularly in relation to the skull, this result indicates that reduction of the AAA can offset the atlanto-occipital joint (Fig. 2). Grob et al. [17] noted that once atlanto-axial arthrodesis was achieved in AAS patients, the inflammatory process with destruction of the lateral masses of the atlas can prevent further deterioration with vertical cranial migration. However, Iizuka et al. [5] demonstrated a case showing ankylosis of the atlanto-occipital joint after transarticular screw fixation. Therefore, we should pay special attention to the effects of this offset in the atlanto-occipital joint. However, in the patients showing ankylosis in the atlanto-occipital joint before surgery [5], this offset mechanism cannot work. Furthermore, we suggest that the proper correction of the AAA in the K group did not induce a loss of cervical lordosis, although hyperlordotic fixation of the AAA has been reported to change the kyphotic lower cervical alignment [11–15]. Finally, in the L group, we acquired a significant reduction of the AAA after surgery; however, this reduction did not affect the skull and lower cervical spine alignment.

This study is associated with some limitations that are worth noting. Due to the small number of sample cases, this study lacks a powerful statistical analysis, and the results might therefore be influenced. Moreover, this study was retrospective, not prospective.

In conclusion, the proper reduction of the AAA did not affect the cranial angles or induce kyphotic malalignment of the subaxial region after atlanto-axial arthrodesis. However, if we can obtain a significant and large reduction of AAA in patients showing kyphosis before surgery, then this reduction will be offset in the atlanto-occipital joint; and we should therefore pay special attention to its morphology after surgery.

Fig. 2 Mechanism of offset of the atlanto-occipital joint after reduction of the AAA. Although the AAA was reduced after surgery, the CAA was maintained due to offset of the atlanto-occipital joint. **a** Before surgery; **b** after surgery

Conflict 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.

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