Relationship between the morphology of the atlanto-occipital joint and the radiographic results in patients with atlanto-axial subluxation due to rheumatoid arthritis

Haku Iizuka · Yasunori Sorimachi · Tsuyoshi Ara ·
Masahiro Nishinome · Takashi Nakajima ·
Yoichi Iizuka · Kenji Takagishi

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Abstract The upper cervical spine is a common focus of destruction from rheumatoid arthritis (RA). Atlanto-axial subluxation (AAS) presents with marked frequency among patients with instability. However, there are occasional patients who show no motion between the occipital bone and atlas on a dynamic cervical radiograph in AAS patients. This study investigated the morphology of the atlanto-occipital joint (AOJ) in AAS patients due to RA using computed tomography, and examined the relationship between its morphology and other radiographic results. Twenty-six consecutive patients with AAS due to RA treated by surgery were reviewed. The subjects included 18 females and 8 males. The average patient age was 59.3 years. The mean duration of RA was 14.3 years. In all the patients, the AOJ was morphologically evaluated using sagittal reconstruction view on computed tomography before surgery. Moreover, the ADI value was investigated at the neutral and maximal flexion position, and atlanto-axial angle (AAA) at the neutral position in preoperative lateral cervical radiographs. The morphology of the AOJ on a CT sagittal reconstruction view was classified into three types as follows: a normal type which showed a maintenance of the joint space, a narrow type which showed a disappearance of the joint space and a fused type which showed the fusion of the AOJ. The pre-

operative CT sagittal reconstruction image of the AOJ demonstrated a normal type bilaterally in six cases (Group A). In 15 cases (Group B), CT image demonstrated narrowing on at least one side of the AOJ. In five cases (Group C), CT images demonstrated fusion on at least one side of the AOJ. The average ADI value at the flexion position was 10.7 mm in Group A, 11.7 mm in Group B, and 12.6 mm in Group C. There was no significant difference among those groups. The average ADI value at the neutral position before surgery was 2.8 mm in Group A, 5.9 mm in Group B, and 10.4 mm in Group C. There was no significant difference between Group A and B ($P > 0.105$), and Groups B and C ($P > 0.032$), however, there was a significant difference between Groups A and C ($P < 0.004$). The average AAA value was 25.3° in Group A, 19.3° in Group B and 3.4° in Group C. There was no significant difference between Groups A and B ($P > 0.230$), however, there was a significant difference between Groups A and C ($P < 0.002$), and Groups B and C ($P < 0.007$). This study showed that fusion or ankylosis of the AOJ induced an enlargement of the 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. This morphology may progress to SAS and VS due to AOJ after atlanto-axial arthrodesis.

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H. Iizuka (✉) · Y. Sorimachi · T. Ara · M. Nishinome ·
T. Nakajima · Y. Iizuka · K. Takagishi
Department of Orthopaedic Surgery,
Gunma University Graduate School of Medicine,
3-39-22, Showa, Maebashi, Gunma 371-8511, Japan
e-mail: ihaku@showa.gunma-u.ac.jp

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Introduction

The upper cervical spine is a common focus of destruction from rheumatoid arthritis (RA). The common features of this instability consist of atlanto-axial subluxation (AAS)

Fig. 1 There was no detectable movement between the occipital bone and atlas, despite reduction of atlanto-axial subluxation observed on an extension radiograph. **a** Neutral, **b** flexion, **c** extension

and vertical subluxation (VS), or a combination of both. AAS presents with marked frequency among patients with instability. It seems that the atlanto-axial joint (AAJ) plays an important role in the occurrence of AAS. Previous authors have described the histological features of the facet joint of upper cervical spine in RA. Eurderink et al. [2] performed a detailed pathological analysis of the facet joint of the cervical spine in RA patients, then described that AAJ lesions were observed in 44%. O'Brien et al. [5] identified potential clinical or radiographic correlations with the histology of AAJ, and noted that mechanical degeneration rather than an inflammatory rheumatoid process seems responsible for the deteriorating neurologic structure in chronic RA at the craniocervical junction.

However, there are occasional patients who show no motion between the occipital bone and atlas on a dynamic cervical radiograph in AAS patients (Fig. 1). Eurderink et al. [2] also described that atlanto-occipital joint (AOJ) lesions were demonstrated in 42% of patients. Moreover, the degree of displacement of the atlas against the axis usually appears quite variable on a lateral cervical radiograph in the neutral position. This study investigated the morphology of the AOJ in AAS patients due to RA using computed tomography, and examined the relationship between its morphology and other radiographic results.

Materials and methods

Twenty-six consecutive patients with AAS due to RA treated by surgery at the university hospital between 2001 September and 2006 December were reviewed. The subjects included 18 females and 8 males. The average patient age was 59.3 years (34–73 years). The duration of RA ranged from 3 to 33 years, and the mean was 14.3 years. Atlanto-axial subluxation was diagnosed by a lateral cervical radiograph showing an anterior atlanto-dental interval (ADI) on a flexion radiograph of 3 mm or more. The

average ADI values in neutral and flexion position before surgery was 6.1 and 11.6 mm, respectively. In patients with cervical myelopathy with atlanto-axial instability, surgery was usually indicated as well as when the patients complained of only neck pain, it is adapted after unsuccessful adequate conservative treatment. We performed selective atlanto-axial arthrodesis (transarticular screw fixation) for all the patients and none of them required more extensive surgery, such as occipito-cervical fusion.

In all the patients, the AOJ was morphologically evaluated using sagittal reconstruction view (slice thickness 2 mm) on computed tomography (CT: Light Speed Qx-I, GE, slice thickness 1.25 mm) before surgery. Moreover, the ADI value was investigated at the neutral and maximal flexion position and atlanto-axial angle (AAA) at the neutral position in preoperative lateral cervical radiographs (Fig. 2). The morphology of the AOJ was examined by the second author (Y. S) who did not perform these operations and by the first author (H. I). The morphology of the AOJ on a CT sagittal reconstruction view was classified into three types as follows: a normal type which showed a maintenance of the joint space, a narrow type which showed a disappearance of the joint space and a fused type which showed the fusion of the AOJ (Fig. 3a, b, c). The ADI and AAA values were analyzed using ANOVA and Bonferroni's/Dunn's post-test for an analysis of variance and the post hoc test.

Fig. 2 Method of radiological measurement. **a** Atlanto-dental interval (ADI), **b** Atlanto-axial subluxation (AAA)

Fig. 3 Classification of the morphology of the atlanto-occipital joint. **a** Normal type, **b** narrow type, **c** fused type

Results

The pre-operative CT sagittal reconstruction image of the AOJ demonstrated a normal type bilaterally in six cases (23.1%, Group A). In 15 cases (57.6%, Group B), CT image demonstrated narrowing on at least one side of the AOJ (one side: 3 cases, both sides: 12 cases). In five cases (19.2%, Group C), CT images demonstrated fusion on at least one side of the AOJ (one side: 3 cases, both sides: 2 cases). These findings are summarized Table 1. The average ADI value at the flexion position was 10.7 mm in Group A, 11.7 mm in Group B, and 12.6 mm in Group C. There was no significant difference among those groups ($P > 0.551$). The average ADI value at the neutral position before surgery was 2.8 mm in Group A, 5.9 mm in Group B, and 10.4 mm in Group C (Table 2). There was a significant difference among those groups ($P < 0.012$). There was no significant difference between Groups A and B ($P > 0.105$), and Groups B and C ($P > 0.032$), however, there was a significant difference between Groups A and C ($P < 0.004$). The average AAA value was 25.3° in Group A, 19.3° in Group B and 3.4° in Group C (Table 2). There was a significant difference among those groups ($P < 0.006$). There was no significant difference between Groups A and B ($P > 0.230$), however, there was a significant difference between Groups A and C ($P < 0.002$), and Groups B and C ($P < 0.007$).

Case series

Case 24

A 65-year-old female was treated with atlanto-axial arthrodesis at this hospital. No severe displacement was detected between C1 and C2 on a lateral cervical radiograph in the neutral position (Fig. 4a). The CT image before surgery demonstrated the maintenance of the joint space of the AOJ (Fig. 4b).

Table 1 Patient characteristics

No	Age	Sex	ADI		AAA	AOJ	
			Flexion	Neutral		Rt	Lt
1	59	M	8	1	26	Normal	Normal
2	54	F	8	2	23	Normal	Normal
3	61	F	11	1	18	Narrow	Narrow
4	64	F	14	1	36	Narrow	Narrow
5	65	F	12	10	0	Fused	Narrow
6	38	F	12	7	10	Fused	Fused
7	47	F	12	11	20	Narrow	Narrow
8	56	F	10	7	18	Normal	Narrow
9	34	M	12	7	20	Narrow	Narrow
10	66	M	15	2	29	Normal	Normal
11	69	F	17	16	-5	Normal	Narrow
12	63	F	8	5	15	Narrow	Narrow
13	71	F	13	11	0	Fused	Fused
14	53	M	15	13	-2	Fused	Narrow
15	56	F	11	11	9	Fused	Narrow
16	60	F	11	3	38	Normal	Normal
17	61	F	12	3	33	Narrow	Narrow
18	37	F	10	8	18	Normal	Normal
19	69	M	12	11	15	Narrow	Narrow
20	69	F	10	2	32	Narrow	Narrow
21	58	F	10	6	36	Narrow	Narrow
22	62	M	13	9	-2	Narrow	Narrow
23	72	F	9	7	19	Narrow	Normal
24	65	F	12	1	18	Normal	Normal
25	73	M	12	2	18	Narrow	Narrow
26	59	M	13	1	17	Narrow	Narrow

Case 21

A 58-year-old female underwent transarticular screw fixation at this hospital. The hyperextension of the AAA was demonstrated pre-operatively (Fig. 5a). The CT image demonstrated disappearance of the joint space (Fig. 5b).

Table 2 Summary of the radiological data

	Group A (<i>n</i> = 6)	Group B (<i>n</i> = 15)	Group C (<i>n</i> = 5)
ADI			
Flexion (mm)	10.7 ± 2.7	11.7 ± 2.2	12.6 ± 1.5
Neutral (mm)	2.8 ± 2.6	5.9 ± 4.5	10.4 ± 2.2
AAA	25.3° ± 7.6°	19.3° ± 12.0°	3.4° ± 5.6°

Fig. 4 Case 24: A 65-year-old female with AAS. A lateral cervical radiograph in the neutral position showing minimal displacement between C1 and C2. Sagittal reconstruction view on CT before surgery showing the maintenance of the joint space. **a** Lateral radiograph, **b** sagittal reconstruction view **Fig. 5** Case 21: A 58-year-old female with AAS. Lateral cervical radiograph showing a slight enlargement of the ADI and an increase of the AAA. Sagittal reconstruction view on CT showing disappearance of the joint space. **a** Lateral radiograph, **b** sagittal reconstruction view

Case 14

A 53-year-old male showed AAS involvement. A significant displacement of the atlas was demonstrated pre-operatively (Fig. 6a). The CT image showed fusion of the AOJ on the right side of the facet joint (Fig. 6b).

Fig. 6 Case 14: A 53-year-old male with AAS. A lateral cervical radiograph showing severe displacement between C1 and C2. Sagittal reconstruction view showing fusion of the AOJ. **a** Lateral radiograph, **b** sagittal reconstruction view

Discussion

Eurderink et al. [2] noted that a bony ankylosis of the AOJ in RA patients was demonstrated in 15%, and in some cases this ankylosis showed complete bony union. This suggests that the morphological fused type indicates pathological fusion or ankylosis. Actually, it was possible to acquire the extension of the atlas through extension of the head during surgery in Group C, although this was not possible in other group since the AOJ often offset this extension.

In the current study, enlargement of the ADI at the neutral position was demonstrated in Group C showing fusion of the AOJ at least one side, although it was close to the normal value in Group A showing normal type bilaterally. Toyama et al. [6] noted that the normal angle of the AAA was 25° after evaluating the AAA in a normal population. Although, the AAA value in Group A in the present series was similar to this value, a severe anterior inclination of the atlas was demonstrated in Group C. Therefore, this demonstrated that either fusion or ankylosis of the AOJ at least on one side induced an enlargement of the ADI and anterior inclination of the atlas, despite the fact that the patients with a normal type bilaterally showed a slight displacement between the atlas and axis. However, patients showing an enlargement of the ADI and anterior inclination of the atlas was also demonstrated in Group B. In fact, it was thought that the bony spur of the dens and anterior arch of the atlas induced enlargement of the ADI in case 19 (Fig. 7). The average ADI and AAA in Group B both showed intermediate values between those of Groups A and B. The average ADI value at the neutral position did not show a significant difference in comparison to those values in the other group, however, the average AAA value was significantly different from the value of Group C.

Fig. 7 Case 19: Midline Sagittal reconstruction view. A spur of the odontoid process and the anterior arch of C1 that induced an enlargement of ADI is clearly seen

Fig. 8 Case 22: The occurrence of ankylosis in the AOJ was clearly observed 1 year after surgery. **a** Preop, **b** 1 year postop, **c** Preop AOJ, **d** 1 year postop AOJ

Regarding the surgical results in rheumatoid patients with upper cervical instability, Casey et al. [1] noted that there is evidence that an early operation reduces the incidence of VS and SAS. O'Brien et al. [5] also noted that consideration should be given to preemptive surgery for

decompression and stabilization before the onset of severe myelopathy. Moreover, Grob et al. [4] 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, Fujiya et al. [3] noted in 57 patients showing isolated atlanto-axial instability or in combination with mild vertical subluxation treated by atlanto-axial arthrodesis, that superior migration of the odontoid process occurred in four patients (7%), and two patients required occipital-cervical fusion as a second procedure. Regarding this issue, in Group C, there is no need for concern. However, in another Group, especially in Group B, the occurrence of any involvement including vertical subluxation due to the AOJ after atlanto-axial arthrodesis should also be considered (Case 22, Fig. 8). Furthermore, the occurrence of SAS, especially C2 spondylolisthesis, after atlanto-axial arthrodesis in Group C should also be considered, since fusion was demonstrated from the occipital bone to C2.

In conclusion, this study showed that fusion or ankylosis of the AOJ induced an enlargement of the 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. This morphology may progress to SAS and VS due to AOJ after atlanto-axial arthrodesis.

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