Characteristics of idiopathic atlanto-axial subluxation: a comparative radiographic study in patients with an idiopathic etiology and those with rheumatoid arthritis

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Abstract

Objective Atlanto-axial subluxation (AAS) is caused by multiple conditions; however, idiopathic AAS patients without RA, upper-cervical spine anomalies or any other disorder are rarely encountered. This study retrospectively investigated the radiographic findings in idiopathic AAS patients, and clarified the differences between those AAS patients and those due to RA.

Methods Fifty-three patients with AAS treated by transarticular screw fixation were reviewed. The subjects included 8 idiopathic patients (ID group) and 45 RA patients (RA group). The study investigated the atlanto-dental interval (ADI) value and space available for spinal cord (SAC) at the neutral and maximal flexion position.

Results The average ADI value at the neutral position in the ID and RA groups before surgery was 7.8 and 7.2 mm, respectively ($p > 0.74$). The average ADI value at the flexion position in the two groups was 10.3 and 11.7 mm, respectively ($p > 0.06$). The average SAC value at the neutral position in the two groups was 12.0 and 17.1 mm, respectively ($p < 0.01$). Finally, the average SAC value at the flexion position in the two groups was 10.7 and 13.5 mm, respectively ($p < 0.01$).

Conclusions The SAC value at both the neutral and flexion positions in idiopathic AAS patients was significantly smaller than those values in RA-AAS patients. This may be because the narrowing of the SAC in the idiopathic

group easily induces cervical myelopathy. Furthermore, surgery was often recommended to RA patients, because of the neck pain induced by RA-related inflammation of the atlanto-axial joint, regardless of any underlying myelopathy.

Keywords Atlanto-axial subluxation · Idiopathic · Rheumatoid arthritis · Radiographic study

Introduction

Atlanto-axial subluxation (AAS) is caused by multiple conditions, such as an upper cervical spine anomaly, rheumatoid arthritis (RA), Down syndrome, infections and other conditions. RA, in particular, is a common cause for such instability, and some authors [5, 6, 10] have reported their recent clinical results after atlanto-axial arthrodesis. However, idiopathic AAS patients without RA, upper-cervical spine anomaly or any other disorder are rarely encountered. Greenberg [2] classified AAS into two groups, including an incomplete odontoid process and an incomplete transverse atlantal ligament. Furthermore, the latter group was divided into a traumatic group, hyperemic group and congenital group which all included idiopathic type of AAS. However, the characteristics of idiopathic AAS have not been described elsewhere. This study retrospectively investigated the radiographic findings in idiopathic AAS patients, with the goal of clarifying the differences between the AAS patients and those due to RA.

Materials and methods

Fifty-three patients with AAS treated by surgery at the university hospital between 2001 and 2010 were reviewed.

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The subjects included 8 idiopathic patients (ID group) and 45 RA patients (RA group). AAS was diagnosed by a lateral cervical radiograph showing an anterior atlanto-dental interval (ADI) on a flexion radiograph of 3 mm or more. None of the patients in the ID group had any systemic inflammatory disease and history of trauma, and none of the patients demonstrated any bony anomaly of the cervical spine like os odontoideum, atlas assimilation or Klippel–Feil syndrome. The duration of RA in the RA group ranged from 3 to 33 years, and the mean was 14.3 years. RA patients showing a Ranawat [9] value less than 13 mm were not included in this series in order to exclude the effects of vertical subluxation induced by RA inflammation.

Surgery was usually indicated in patients with cervical myelopathy with atlanto-axial instability. In addition, patients presenting with only neck pain after unsuccessful adequate conservative treatment were also recommended for surgery. Selective atlanto-axial arthrodesis (transarticular screw fixation) [3, 7] were performed for all patients, and none required more extensive surgery, such as occipito-cervical fusion.

The Ranawat [9] value and the atlanto-axial angle (AAA) at the neutral position were investigated in preoperative lateral cervical radiographs (Fig. 1a, b). The ADI value and space available for spinal cord (SAC) were also investigated at the neutral and maximal flexion positions (Fig. 1c, d). All parameters were analyzed using the *t* test, the Mann–Whitney *U* test, and the Chi-square test for independence. A value of $p < 0.05$ was considered to be significant.

Results

The ID group included four males and four females with an average age of 64.3 years (45–77 years). The RA group included 13 male and 32 female patients. The average patient age was 61.0 years (34–78 years). There were no significant differences between the two groups in age ($p > 0.42$) or gender ($p > 0.23$) (Table 1).

The average Ranawat value in the ID group and RA group before surgery was 16.0 and 15.1 mm, respectively ($p > 0.27$). The average AAA value in the two groups was

Fig. 1 Method used for radiological measurement. **a** Ranawat value: the distance from the center of the pedicle axis to the line connecting the center of the anterior and posterior arches. **b** The atlanto-axial angle (AAA): the angle determined by measuring the angle formed by two lines extending from the inferior border of the C2 vertebral body and the line connecting the center of the anterior and posterior arches. **c** The atlanto-dental interval (ADI) and space available for spinal cord (SAC) at the neutral position measured on the line connecting the center of the anterior and posterior arches. **d** ADI and SAC at the maximal flexion position

Table 1 Comparison of status between the ID group and RA group

	ID group (8 cases)	RA group (45 cases)	
Age	64.3 ± 10.6	61.0 ± 10.5	$p > 0.42$
Gender (M:F)	4:4	13:32	$p > 0.23$

Table 2 Comparison of radiographic data between the ID group and RA group

	ID group	RA group	
Ranawat value (mm)	16.0 ± 1.3	15.1 ± 2.3	$p > 0.27$
AAA (°)	23.1 ± 10.8	18.8 ± 11.8	$p > 0.33$
ADI at neutral (mm)	7.8 ± 5.1	7.2 ± 4.5	$p > 0.74$
ADI at flexion (mm)	10.3 ± 4.3	11.7 ± 2.4	$p > 0.06$
SAC at neutral (mm)	12.0 ± 2.3	17.1 ± 5.3	$p < 0.01$
SAC at flexion (mm)	10.7 ± 1.8	13.5 ± 3.0	$p < 0.01$

23.1° and 18.8°, respectively ($p > 0.33$). The average ADI value at the neutral position in the ID and RA groups before surgery was 7.8 and 7.2 mm, respectively ($p > 0.74$). The average ADI value at the flexion position in the two groups was 10.3 and 11.7 mm, respectively; there was no difference between the two groups ($p > 0.06$). The average SAC value at the neutral position in the two groups was 12.0 and 17.1 mm, respectively; there was a significant difference between the two groups ($p < 0.01$). Finally, the average SAC value at the flexion position in the two groups was 10.7 and 13.5 mm, respectively, which was also significantly different ($p < 0.01$) (Table 2).

Case studies

Case 1

A 57-year-old male (belonging to the ID group, Fig. 2) who complained of neck pain was treated with atlanto-axial arthrodesis at the author's hospital. The ADI and SAC values in the neutral position before surgery were 10.6 and 12.0 mm, respectively. The ADI and SAC values at the maximal flexion position before surgery were 11.3 and 10.9 mm, respectively.

Case 2

A 65-year-old female (belonging to the RA group, Fig. 3) who complained of neck pain was also treated. The ADI and SAC values at the neutral position before surgery were 9.5 and 15.7 mm, respectively. The ADI and SAC values at

the maximal flexion position before surgery were 10.5 and 15.5 mm, respectively.

Discussion

Greenberg [2] classified AAS and noted that dislocations due to congenital incompetence of the transverse atlantal ligament can be divided into those due to no known cause and those found in association with Down syndrome. He also noted that the idiopathic AAS group is really one of exclusion: no history of trauma, infection, RA and associated congenital anomaly that might place excessive stress on the ligament. Therefore, the ID group in the current study was classified into this group. Hensinger [4] reported that the patients showed congenital laxity of the transverse atlantal ligament and noted that this is a diagnosis of exclusion suggested by the clinical occurrence of chronic AAS, without a predisposing cause such a history of trauma, congenital anomaly, infection or RA to account for the radiographic findings. We also considered that the ID group in the current study should be classified into this group. Wadia [11] investigated 28 cases of congenital AAS, and also classified those into two groups, including an odontoid process attached group and an odontoid detached or absent group. Furthermore, the former group was divided into a group with occipitalization of the atlas and that without occipitalization, which included all idiopathic type AAS. He also noted that, while the transverse ligament is supposed to be the most important ligament in maintaining the odontoid process in the normal position, in the random autopsy sectioning of the transverse ligament, it was found to be impossible to dislocate the atlanto-axial joints even using considerable manual force. Therefore, he suggested that the stability of the atlanto-axial joint was maintained by all of the ligaments, including the fibrous joint capsules, working as a whole. Fielding et al. [1] also noted that after rupture of the transverse ligament, the alar ligaments are usually inadequate to prevent further displacement of the atlanto-axial joint when a force similar to that which ruptured the transverse ligament is applied.

The current study found no differences in the average age and gender between the ID and RA groups. There were also no differences between the groups in the ADI value at the neutral and flexion position between the two groups. These results suggest that there was no difference between the two groups in the degree of instability between C1 and C2. However, the SAC value at both the neutral and flexion position in the ID group was significantly smaller than those in the RA group. Moreover, Oda et al. [8] evaluated the diagnostic validity of SAC at the C1 level for myelopathy in patients with RA and noted that a value of 14 mm or less was

Fig. 2 Case 1: ID group, a 57-year-old male who complained of neck pain and numbness in his hands.

a Neutral position, **b** flexion position, **c** extension position and **d** MRI T2 image

recommended as a cutoff point for SAC for screening patients with a high risk for myelopathy. This suggests that narrowing of the SAC in the ID group may induce the occurrence of cervical myelopathy. The inflammation of the atlanto-axial joint often induces severe neck pain in patients with AAS due to RA, thus surgery was more likely to be performed due to the presence of neck pain only without any accompanying cervical myelopathy. Consequently, the differences in the surgical indications between these two groups may account for these results. However, one limitation of this study is that the degree of severity of the cervical myelopathy was not evaluated. In RA patients, joint destruction often induces deterioration in the activities of daily life; therefore, we considered that it was not suitable to compare the pre- and

postoperative neurological status between the groups. Furthermore, RA patients frequently have a retro dental pannus contributing to the SAC narrowing, and thus have MRI stenosis in the neutral, and particularly in the flexion position, which is usually greater than radiographic stenosis.

In conclusion, the SAC value at both the neutral and flexion positions in idiopathic AAS patients was significantly smaller than those values in RA-AAS patients. This difference may be because the narrowing of the SAC in idiopathic group easily induces cervical myelopathy. Furthermore, RA patients often underwent surgery because of the neck pain induced by RA-related inflammation of the atlanto-axial joint, regardless of any underlying myelopathy.

Fig. 3 Case 2: RA group, a 65-year-old female who complained of neck pain. **a** Neutral position, **b** flexion position, **c** extension position and **d** MRI T2 image

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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