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Incidence and risk factors for adjacent segment degeneration following occipitoaxial fusion for atlantoaxial instability in non-rheumatoid arthritis

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Abstract

Purpose To investigate the incidence and risk factors for adjacent segment degeneration (ASD) following occipitoaxial fusion (OAF) for atlantoaxial instability (AAI) in non-rheumatoid arthritis (RA).

Methods The study group comprised 41 patients without RA who underwent OAF due to AAI. Fifteen patients with postoperative ASD after OAF were classified as the ASD group, and the other 26 patients without postoperative ASD were included in the non-ASD group. There were 12 men and 3 women with a mean age of 43.52 years in the ASD group, and 19 men and 7 women with a mean age of 45.31 years in the non-ASD group. The mean follow-up period was 6.1 and 5.9 years in the ASD group and non-ASD group, respectively. Clinical outcomes and plain radiographs were retrospectively reviewed and compared between the two groups.

Results The difference between pre- and postoperative O-C2 angles in the non-ASD group was significantly greater than that in the ASD group. The C2–7 angles changed significantly between the pre- and postoperative periods. It was suggested that the small O-C2 angle and large C2–7 angle observed in the early postoperative period were risk factors for the development of ASD. We also demonstrated a high incidence of subaxial subluxation (SAS) and swan neck deformity in the ASD group (27 versus 3.8% and 20 versus 0%, respectively).

Conclusion Under-correction of the O-C2 angle is likely to cause malalignment of the cervical spine, resulting in the development of postoperative ASD, SAS, and swan neck deformity.

Keywords Adjacent segment degeneration · Atlantoaxial · Occipitoaxial · Risk factor

Introduction

Atlantoaxial instability (AAI) can result from different aetiologies, such as traumatic, inflammatory, and congenital abnormalities [1]. It has been reported that the O-C2 angle strongly correlates with the C2–7 sagittal alignment and occipitoaxial fusion (OAF) in hyperextension, resulting in the development of kyphosis of the lower cervical spine [2, 3]. However, there have been no reports showing the correlation between these radiographic parameters and the development of adjacent segment degeneration (ASD), subaxial subluxation (SAS), and iatrogenic swan neck deformity in non-rheumatoid arthritis (RA) patients. In the present study, we evaluated preoperative and postoperative radiographs following OAF for AAI in patients without RA to identify risk factors for the later development of ASD, SAS, and swan neck deformity.

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Materials and methods

Patients

We performed a retrospective study to investigate the incidence of and risk factors for ASD following OAF for AAI in non-RA. The inclusion criteria were in accordance with the Kellgren degeneration grading system as follows [4]: the adjacent segment was grade 0–1 before the procedures, and the follow-up period was more than 5 years and included complete radiological data, including cervical anteroposterior and lateral X-rays preoperatively, early postoperatively, and at the final follow-up, as well as computed tomography (CT) before the procedures and at the final follow-up. The exclusion criteria were the following: degeneration grading ≥ grade 2 before procedures; RA cases; paediatric cases; poor compliance, unavailability of regular follow-up information, and insufficiency of the follow-up materials.

Between June 2002 and August 2011, 41 cases that met both the inclusion and exclusion criteria were retrospectively reviewed. The aetiologies were atlas transverse ligament rupture ($n = 14$), type II fractures of the odontoid process ($n = 12$), os odontoideum (OO) ($n = 7$), occipitalization of the atlas ($n = 4$), and basilar invagination ($n = 4$). All patients had various extents of occipitocervical pain and restricted cervical movement. Other common presenting symptoms included tingling numbness in limbs ($n = 17$) and incomplete paraplegia ($n = 7$).

Surgical procedure

No patients underwent emergency surgery. Cranial traction was applied in other patients and the traction weight ranged from 3 to 5 kg. Neurological function was monitored by intraoperative somatosensory-evoked potentials. After endotracheal intubation and general anaesthesia, the patient was placed in the prone position with the head immobilized and slightly flexed by a Mayfield head holder. Following routine disinfection and draping, a standard midline incision was made from the inion to the C4 spinous process to expose the posterior arch, the lamina, and the pedicle of C2. For patients with compression of the foramen magnum, the posterior margin of the foramen magnum and fused posterior arch were removed. Pedicle screws were implanted into bilateral C2. An occipital plate was applied by drilling three holes into the midline keel of the occiput beneath the external occipital protuberance and fixed using bi-cortical screw purchase. Then, the occipital plate was attached to bilateral pre-curved titanium rods. As demonstrated by C-arm, the internal fixation was well placed and the atlantoaxial joint was sufficiently reduced. After irrigation with saline solution, autologous structural and cancellous bone grafting was performed between the decorticated occiput and C2 spinous process. Rigid cervical collar protection was maintained for approximately 8–12 weeks postoperatively. When the bony fusion was displayed on X-ray imaging, the cervical collar was removed. Postoperatively, bony fusion was confirmed by the absence of a solid or translucent line around the grafting area and lack of instrument failure on dynamic radiographs. Furthermore, the surgical variables included operative time and blood loss.

Functional assessment

The Neck Disability Index (NDI) and Japanese Orthopaedic Association (JOA) score were used to evaluate the functional result before and after the operations. The formula for the JOA improvement rate, indicating neurological function, was the following: postoperative improvement rate = (postoperative score − preoperative score)/(17 − preoperative score) × 100%.

Radiological evaluation

The angle between the occipital bone and the axis (abbreviated O-C2 angle) was determined by the McGregor line and the inferior surface of C2. The C2–7 angle was measured using the Cobb method between the inferior end plate of the axis and that of C7 (Fig. 1). Angular motion at the adjacent segment was measured between the inferior endplate line of the upper vertebral body and superior endplate line of the lower vertebral body on flexion–extension lateral plain film. Disc degeneration on X-ray was rated from grade 0 to 4 using the Kellgren classification system, and ASD was defined as a grade ≥ 2. Patients who displayed slippage of more than 3 mm from the neighbouring vertebral body were considered to have SAS. In addition, the incidence of swan neck deformity was also recorded. Data measurements were

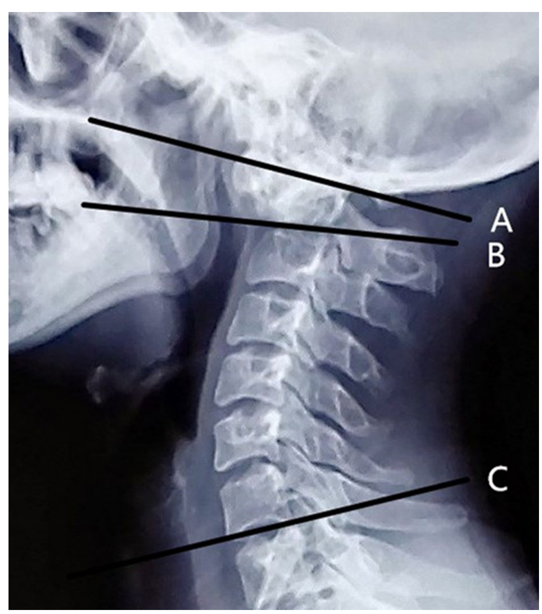

Fig. 1 Evaluation of radiological parameters. ∠AB, O-C2 angle; ∠BC, C2–7 angle

performed three times by the first and second authors independently, and the mean value was used for analysis.

According to the occurrence of ASD during the follow-up period, patients were divided into two groups: non-ASD group and ASD group.

Statistical analysis

Continuous data are expressed as the mean ± standard deviation, and categorical data are reported as the frequency and percentage. Paired and unpaired *t* tests were used to compare changes in the same group or between different groups with parametric values. The χ^2 test and Fisher's exact test or Mann–Whitney *U* test were employed for nonparametric comparisons. To confirm the correlations between different parameters, Pearson's correlation coefficient (*r*) was selected. The *t* test, χ^2 test, and Mann–Whitney *U* test were considered significant if the *P* value was less than 0.05. Statistical analyses were performed using SPSS 20.0 software.

Results

General characteristics and surgical results

Differences in the demographic characteristics and surgical results between the two groups were not significant. The above-mentioned results are displayed in Table 1. No significant complications occurred during the surgery, i.e., vertebral artery (VA) or spinal cord injuries. Similarly, all patients achieved osseous fusion after 6 months, and no instrumental failure occurred during the entire follow-up period. No patient underwent a reoperation due to ASD, SAS, or swan neck deformity.

Functional results

Both groups showed a significant improvement in neurological function postoperatively or at the final follow-up compared with the preoperative values. The immediate postoperative JOA scores were not significantly different between the two groups. Although no significant difference was seen between the two groups in terms of JOA scores at the final follow-up, significance was almost reached (Fig. 2). However, the improvement rates in the JOA score were 46.7 ± 1.6 and 47.4 ± 1.8% at the final follow-up in the ASD and non-ASD groups, respectively, and no significant difference was observed between the two groups ($P = 0.21$). Compared with that of the ASD group, the NDI of the non-ASD group did not differ before and after the operation. However, at the final follow-up, the NDI between the two groups was significantly different (Fig. 3).

Table 1 General characteristics of and surgical variables in the two groups

	ASD group ($n = 15$)	Non-ASD group ($n = 26$)	*P* value
Age (year)	43.52 ± 6.31	45.31 ± 5.75	0.36
Gender ratio (male/female)	12/3	19/7	0.62
Follow-up period (year)	6.1 ± 0.3	5.9 ± 0.4	0.10
Operation time (min)	141 ± 59	124 ± 54	0.35
Blood loss (ml)	295 ± 48	270 ± 42	0.09

Radiological results

The ASD developed at a mean follow-up of 38 months (range 4–72 months) after OAF, and the disc degeneration rating significantly increased in the ASD group. A significant difference was observed between the two groups in terms of the angular motion of C2–3 at the final follow-up. In addition, 4 of the 15 cases developed SAS and 3 patients

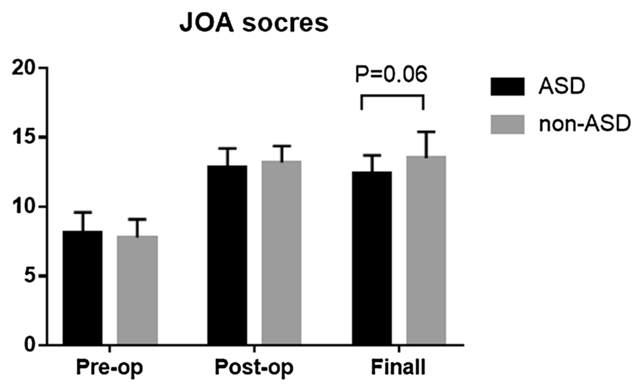

Fig. 2 Neurological outcomes in the two groups

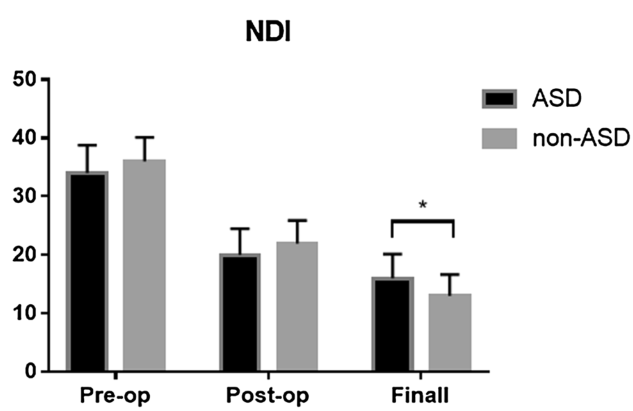

Fig. 3 NDI in the two groups

Table 2 Comparison of radiographic parameters between the ASD and non-ASD groups

	ASD group ($n = 15$)	Non-ASD group ($n = 26$)	P value
Preoperative Kellgren grade of X-ray			
Grade 0/1/2/3/4	10/5/0/0/0	20/6/0/0/0	0.60
Kellgren grade of X-ray at final			
Grade 0/1/2/3/4	0/0/3/9/3	14/12/0/0/0	< 0.001
Angular motion of C2–3 (°) at final	1.4 ± 1.6	3.5 ± 1.8	< 0.001
The incidence of SAS	27%	3.8%	0.03
The incidence of swan neck deformity	20%	0.0%	< 0.001

SAS subaxial subluxation

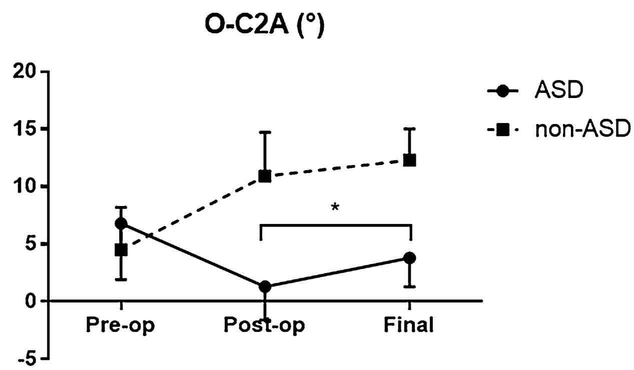

Fig. 4 O-C2 angles in the two groups

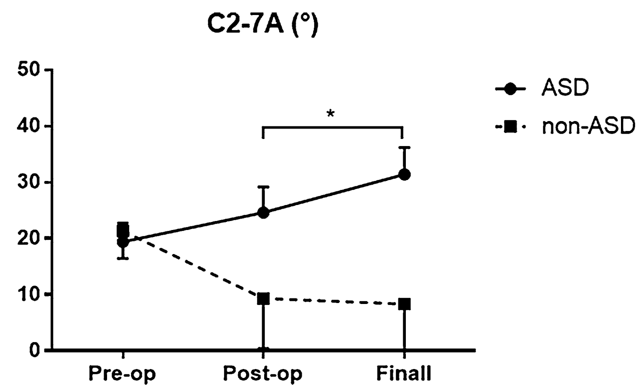

Fig. 5 C2–7 angles in the two groups

had swan neck deformity during the follow-up period. However, only one patient had symptoms (neck pain). This incidence was significantly different from that in the non-ASD group. The data mentioned above are displayed in Table 2.

The relationship between the O-C2 angle and cervical lordosis in the entire group at the time of the final follow-up was assessed. There was a negative correlation between the O-C2 angle and C2–7 angle at the last follow-up (Pearson correlation coefficient $R = -0.49$, $P < 0.001$). Then, we evaluated whether the degree of O-C2 correction would influence the cervical alignment in the long term postoperatively. Interestingly, there was a correlation between the correction of the O-C2 angle and changes in the C2–7 angle (preoperative versus final follow-up) (Pearson correlation coefficient $R = -0.64$, $P = 0.03$).

The baselines of the O-C2 angle in the two groups were not significantly different. The average postoperative O-C2 angle was significantly smaller in the ASD group. Furthermore, the value of the O-C2 angle at the final follow-up was significantly different from the immediately postoperative values in the ASD group (Fig. 4).

The average preoperative C2–7 angle in the non-ASD group decreased significantly and the postoperative C2–7 angle in the non-ASD group was significantly smaller than those in the ASD group. In contrast, the C2–7 angle increased significantly and differed from preoperative values in the ASD group. At the final follow-up, the C2–7 angles between the two groups were still significantly different (Fig. 5).

Discussion

The prevalence of SAS among the operated patients and the high incidence (15–76.5%) of newly developed SAS in RA patients after occipitocervical fusion have been reported [5–7]. However, a few studies have focused on the development of ASD after O-C2 arthrodesis for AAI. The previous studies have shown that there is a linear correlation between an increase in O-C2 lordosis and a decrease in C2–7 lordosis after O-C2 arthrodesis [2, 8]. These studies suggested that occipitocervical fixation in the hyperlordotic position leads to postoperative subaxial kyphosis. Our results are consistent with those of previous reports that describe a negative linear correlation between the perioperative change in the O-C2 and C2–7 sagittal angles. In our study, 15 of 41 cases (36.6%) developed ASD after OAF. A previous study demonstrated that 67% of the patients (8 of 12) had C2–3 spontaneous fusion on postoperative radiographs after C1–2 arthrodesis [9]. To exclude the effect of inflammatory processes, we did not include RA patients. In addition, the patients in our study were younger than those in the previous studies. Thus, we speculate that the occurrence rate of ASD after surgery is mainly caused by mechanical changes in cervical alignment and leading to lower incidence than in the previous studies. Notably, ASD and swan neck deformity did not result in neurological deficits but rather a decrease in NDI in our study. However, this may reduce quality of life. Moreover, the difference in JOA scores between the two groups nearly reached statistical significance, which may need longer follow-up period to verify the difference.

To our knowledge, this study is the first to demonstrate that an increase in postoperative lordosis resulting from under-correction of the O-C2 angle during OAF causes postoperative ASD. In the previous studies, the optimal angle of OAF and the relationship between the postoperative O-C2 angle and the cervical sagittal alignment have been mentioned [2, 3, 10]. Passias et al. recommended that the O-C2 angle should be corrected to approximately 15°, as overcorrection possibly causes subaxial kyphosis. In our study, the postoperative O-C2 angle was approximately 2°, which is significantly less than 15°. This small angle may cause compensatory hyperlordosis in C2–7. Because a negative correlation was observed between the O-C2 angle and C2–7 angle, the appropriate OAF angle should be carefully determined preoperatively [3, 11].

In the C1–2 arthrodesis for AAI, some investigators have speculated that the decrease in the C2–7 angle resulting from hypercorrection of the AA angle may cause postoperative SAS in patients with RA [12, 13]. In our study, under-correction of the O-C2 angle may also have led to the development of SAS, and the rate in the ASD group was 27%. Furthermore, three patients developed swan neck deformity. It is considered that C2–3 spontaneous fusion with OAF imposes higher stress on the adjacent segment, resulting in the development of SAS. Interestingly, a recent study showed that, after correction of the O-C2 kyphosis, the C2–7 angle was compensated for over the course of several years [11]. This finding indicated that swan neck deformity is reversible. In addition, the previous studies have shown that segmental cervical spine unit angular motion decreases with severe degeneration [14, 15]. In our studies, angular motion of C2–3 in the ASD group was significantly decreased at the final follow-up.

Interestingly, one patient in the present study had an operative history of C2, and the graft bone was extended to C3. Postoperatively, the patient developed C2–3 union and swan neck deformity (Fig. 6). Thus, we speculated that both the O-C2 angle and extended bone graft contributed to the development of ASD and swan neck deformity. Two previous studies support this hypothesis [9, 16]. In addition, chronic dislocation may cause contracture of cervical muscles. Thus, when the dislocation in vertical direction is reduced, higher tension of neck muscles may result. The combination of high cervical muscle tension, swan neck deformity, and OAF may impose more stress on the anterior edge of the intervertebral disc, and cause the difference in height between the anterior and posterior intervertebral discs (Fig. 6c).

Posterior atlantoaxial fixation has contributed greatly to the improvement of clinical symptoms and the increase in fusion rate due to the use of new and reliable surgical procedures and implants. Oshima et al. [17] believed that, in the Magerl technique, the fixation angle strongly depends

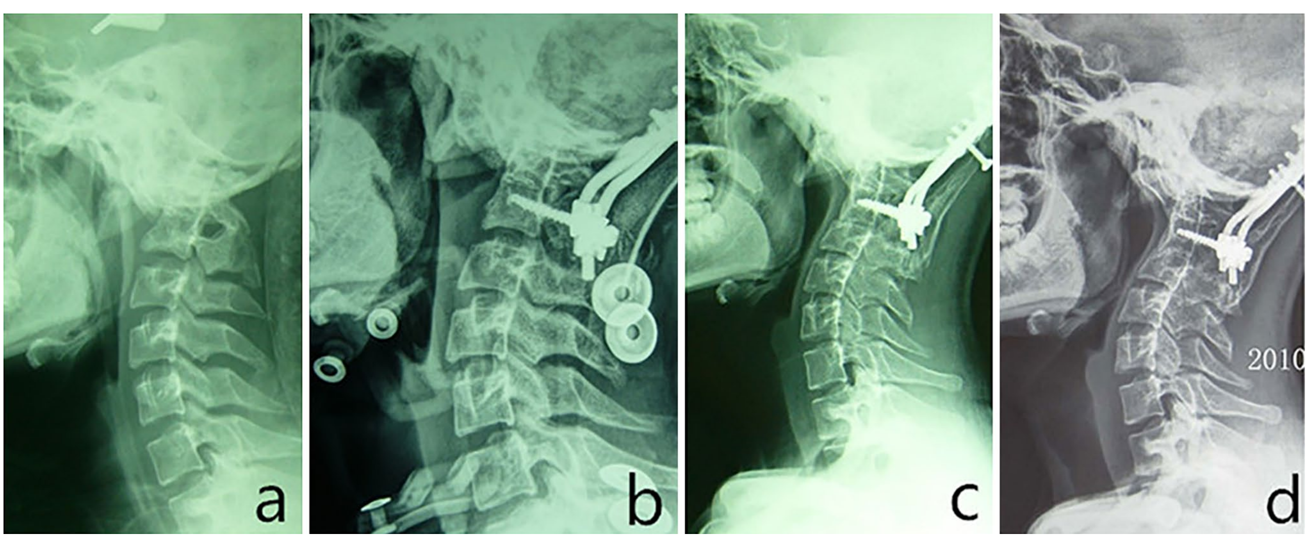

Fig. 6 21-year-old male with non-RA AAI. **a** Preoperative lateral plain films. **b** Postoperative lateral plain films showed that there was atlantoaxial reduction and no ASD. The immediately postoperative O-C2 angle was 5.5°. **c** Lateral plain films 1 year after surgery showed C2–3 degeneration. **d** Lateral plain films 55 months after surgery showed C2–3 union and swan neck deformity

on the preoperative neck position, whereas the C1 lateral mass screw procedure is effective in controlling C1–2 sagittal alignment during surgery. Furthermore, the posterior wiring technique used to supplement the Magerl procedure depends primarily on the compression force between the grafted bone and laminae, which has a tendency to fix C1–2 in the hyperlordotic position [18]. It is also possible that the surgical procedures, such as the insertion of the wire under the C2 lamina or the insertion of transarticular screws, may cause C2–3 interlaminar and/or facet damage, resulting in spontaneous adjacent segment fusion. Thus, the C1 lateral mass screw procedure can maintain normal cervical alignment and may prevent ASD and swan neck deformity. In our study, OAF was not involved in the C1–2 facet. For cervical spine procedures, multilevel fusion could lead to a higher incidence of clinical adjacent segment pathology [19, 20]. However, based on the above results, whether OAF may lead to a higher incidence of ASD than atlantoaxial fusion, especially when C1 lateral mass screws are used, requires further research.

Based on the results of the present study, we recommend that to prevent the development of SAS and swan neck deformity, surgeons should avoid under-correction of the O-C2 angle and extensive exposure of the surgical field following extensive bone graft procedures.

This study has several weaknesses and limitations. First, our population size was relatively small. Second, this study was retrospective. Third, several types of diseases were represented in the patient population. Fourth is the two-dimensional analysis based on lateral radiographs. Nevertheless, the results of our study provide information relevant to a very rare complication and should stimulate further research.

Conclusion

The results of the present study suggested that under-correction of the O-C2 angle in OAF was strongly correlated with the development of postoperative ASD, SAS, and a high incidence of swan neck deformity.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval Ethical approval for this study was obtained from local Medical Ethics Committee.

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