



# Influence of Atlantoaxial Fusion on Sagittal Alignment of the Occipitocervical and Subaxial Spines in Os Odontoideum with Atlantoaxial Instability

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**Study Design:** Retrospective case analysis.

**Purpose:** We hypothesized that larger the C1–C2 fusion angle, greater the severity of the sagittal malalignment of C0–C1 and C2–C7.

**Overview of Literature:** In our experience, instances of sagittal malalignment occur at C0–C1 and C2–C7 following atlantoaxial fusion in patients with Os odontoideum (OO).

**Methods:** We assessed 21 patients who achieved solid atlantoaxial fusion for reducible atlantoaxial instability secondary to OO. The mean patient age at the time of the operation was 42.8 years, and the mean follow-up duration was 4.9 years. Radiographic parameters were preoperatively measured and at the final follow-up. The patients were divided into two groups (A and B) depending on the C1–C2 fusion angle. In group A (n=11), the C1–C2 fusion angle was  $\geq 22^\circ$ , whereas in group B, it was  $< 22^\circ$ . The differences in the radiographic parameters of the two groups were evaluated.

**Results:** At the final follow-up, the C1–C2 angle was increased. However, this increase was not statistically significant ( $18^\circ$  vs.  $22^\circ$ ,  $p=0.924$ ). The C0–C1 angle ( $10^\circ$  vs.  $5^\circ$ ,  $p<0.05$ ) and C2–C7 angle ( $22^\circ$  vs.  $13^\circ$ ,  $p<0.05$ ) significantly decreased. The final C1–C2 angle was negatively correlated with the final C0–C1 and C2–C7 angles. The final C0–C1 angle ( $4^\circ$  vs.  $6^\circ$ ,  $p<0.05$ ) and C2–C7 angle ( $8^\circ$  vs.  $20^\circ$ ,  $p<0.05$ ) were smaller in group A than in group B. After atlantoaxial fusion, the C0–C1 range of motion (ROM;  $17^\circ$  vs.  $9^\circ$ ,  $p<0.05$ ) and the C2–C7 ROM ( $39^\circ$  vs.  $31^\circ$ ,  $p<0.05$ ) were significantly decreased.

**Conclusions:** We found a negative association between the sagittal alignment of C0–C1 and C2–C7 after atlantoaxial fusion and the C1–C2 fusion angle along with decreased ROM. Therefore, overcorrection of C1–C2 kyphosis should be avoided to maintain good physiologic cervical sagittal alignment.

**Keywords:** Os odontoideum; Atlantoaxial instability; Atlantoaxial fusion; Sagittal alignment

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## Introduction

The surgical treatment of patients with C1–C2 instability with Os odontoideum (OO) is proven successful when using a combination of fusion and internal fixation technique [1-4]. Indications for surgical treatment can be simply considered in conjunction with the existence of OO and OO associated with occipitocervical pain and/or with neurologic signs and symptoms. Other factors that may help determine the need for stabilization and/or decompression include C1–C2 instability, associated deformities, and spinal cord compression.

Several techniques have been used to stabilize the C1 and C2 vertebrae in patients with OO [1-11]. The early surgical techniques of C1–C2 fusion include posterior sublaminar wiring techniques, such as the Gallie technique and Brooks–Jenkins technique [5-7]. **The transarticular screw fixation technique**, introduced by Magerl and Seemann [8], is most typically relied upon because it provides immediate rigid fixation postoperatively and has higher fusion rates than posterior sublaminar wiring techniques [9]. **Recently, another C1–C2 posterior fixation technique** that uses a C1 lateral mass screw with C2 pedicle screw fixation, introduced by Goel and Laheri [10] and modified by Harms and Melcher [11], has become widely accepted as a standard treatment.

Although the clinical results for atlantoaxial instability (AAI) are satisfactory, some patients show postoperative reduction in occipitocervical and subaxial cervical lordosis. Several studies have reported that the appearance of occipitocervical and subaxial sagittal malalignments may be attributable to hyperlordotic C1–C2 fusion [12-14]. However, the condition of most patients in these studies was complicated with the presence of rheumatoid arthritis (RA). **Occipitocervical and subaxial sagittal malalignments** may develop as part of the natural course in this kind of disease. Therefore, the treatment results in non-rheumatoid arthritic patients who underwent posterior C1–C2 fusion require further investigation.

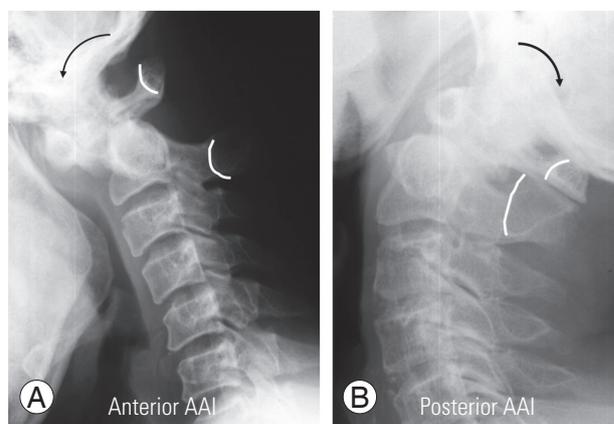
Therefore, we performed retrospective evaluation of the influence of atlantoaxial fusion on the sagittal alignment of the occipitocervical and subaxial spines in OO patients with AAI.

## Materials and Methods

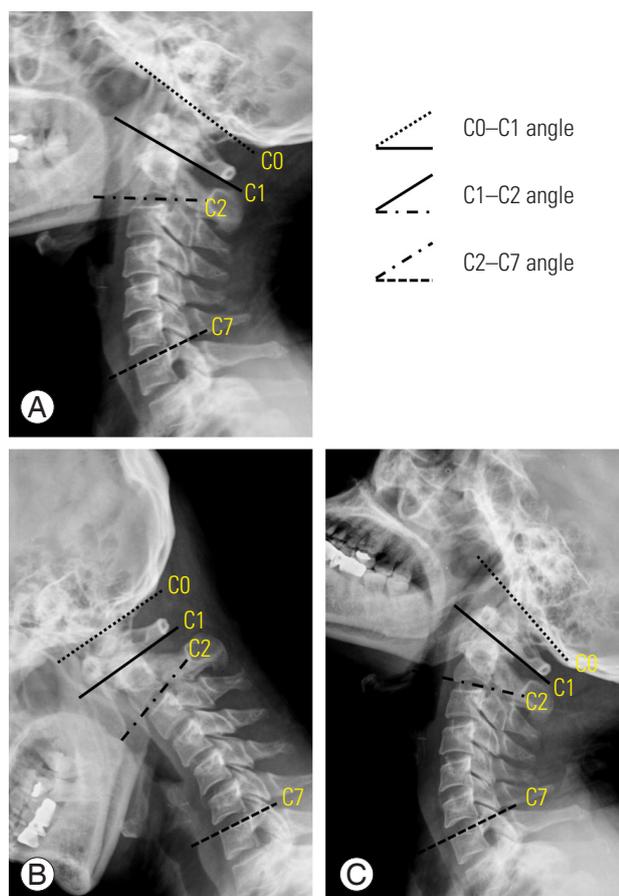
We retrospectively analyzed 21 patients (10 women and

11 men) who achieved solid fusion for OO with reducible AAI and were followed up for at least 24 months. The criteria for C1–C2 fusion were as follows: (1) difference of  $<2^\circ$  between the flexion and extension lateral radiographs, (2) formation of a bony bridge, (3) no findings of implant failure, and (4) radiolucency in  $<50\%$  of the tissue around the implant [15]. The mean age at the time of operation was 42.8 years (range, 5–73 years), **whereas the mean follow-up duration was 4.9 years (range, 2–12 years)**. Eight patients complained of intractable occipitocervical pain, and 13 complained of myelopathy. All the patients with myelopathy showed intramedullary signal change and retrodental reactive lesions (two cystic and 11 fibrocartilaginous lesions) on magnetic resonance imaging. Magnetic resonance images were produced using a 1.5-tesla unit (Siemens, Munich, Germany) in 13 myelopathy patients. Subjects who (1) had RA, (2) underwent combined surgery on the subaxial cervical spine, (3) had non-union, (4) underwent revision surgery, and (5) had combined cervical deformity of  $\leq C2$  level were excluded.

All the surgeries were performed by a single senior surgeon. The direction of AAI was defined as the direction of displacement of the spinolaminar line during flexion and extension (Fig. 1). Eleven patients with anterior AAI underwent posterior sublaminar wiring alone, and 10 patients with combined (anterior+posterior) AAI underwent posterior sublaminar wiring and transarticular screw fixation. Autogenous iliac bone graft was used for all patients. The operative techniques used were as described. The patients were placed in the prone position with the skull fixed with a Mayfield clamp. The neck was positioned to



**Fig. 1.** (A, B) The direction of the AAI was defined as the direction of displacement of the spinolaminar line during flexion and extension. AAI, atlantoaxial instability.



**Fig. 2.** The C0–C1, C1–C2, and C2–C7 angles were measured on neutral (A) lateral radiographs. The C0–C1 angle was measured between the McRae line and the line passing through the centers of the C1 anterior and posterior arches. The C1–C2 angle was measured between the line passing through the centers of the C1 anterior and posterior arches and line tangential to the inferior border of the C2 body. The C2–C7 angle was measured using the Cobb angle between the line on the inferior endplate of C2 and superior endplate of C7. The C0–C1, C1–C2, and C2–C7 range of motions were measured on flexion (B) and extension (C) lateral radiographs.

optimize access by flexing it slightly at the occiput while extending the subaxial spine. We attempted to reduce the C1–C2 articulation during this positioning process. The procedures were assisted with fluoroscopy. A midline incision was made over C1–C2 but distally extended to allow the required drill angulation, and the posterior aspects of C1 and C2 were fully exposed. Subperiosteal exposure of the C1 arch and C2 posterior elements was performed. Cannulated screws were placed for bilateral transarticular fixation under lateral fluoroscopic guidance. Following screw placement, the modified sublaminar wiring method was used to fix an iliac bone graft. A Philadelphia collar or Halovest was applied for 12 weeks after the procedure.

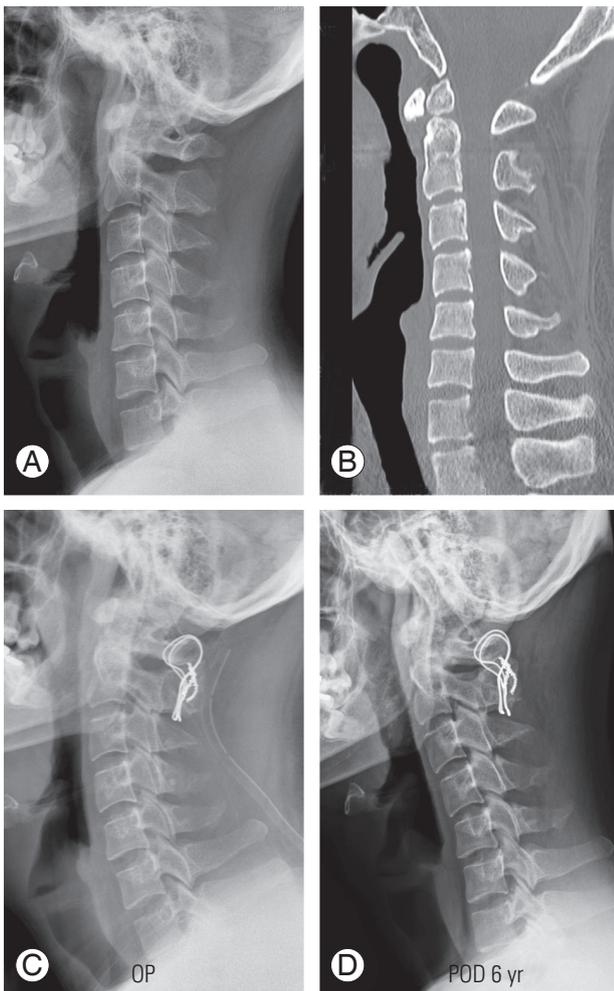
Plain anteroposterior and lateral radiographs of the cervical spine were recorded preoperatively, postoperatively, and at the final follow-up with subjects in the upright neutral, flexion, and extension positions. Radiographic parameters were measured preoperatively and at the final follow-up. The C0–C1, C1–C2, and C2–C7 angles were measured on neutral lateral radiographs. The C0–C1, C1–C2, and C2–C7 range of motions (ROMs) were measured on flexion and extension lateral radiographs (Fig. 2). The differences in the radiographic parameters from before the surgery to that at the final follow-up were evaluated using paired *T*-test. The patients were divided into two groups as per the C1–C2 fusion angle. In group A ( $n=11$ ), the C1–C2 fusion angle was  $\geq 22^\circ$ , whereas that in group B was  $< 22^\circ$  (Table 1). The differences in the radiographic parameters of the two groups were evaluated using independent *T*-test. Correlation between the C1–C1, C0–C1, and C2–C7 angles was analyzed using Pearson test. A *p*-value of  $< 0.05$  was considered statistically significant.

**Table 1.** Demographic and radiological data according to final C1–2 fusion angle

Variable	Group A (C1–2 angle $\geq 22^\circ$ , N=11)	Group B (C1–2 angle $< 22^\circ$ , N=10)	<i>p</i> -value
Age (yr)	40.98 $\pm$ 9.2	44.8 $\pm$ 14.5	0.36
Sex (male/female)	7/4	3/7	0.56
Operation method (PSLW/PSLW+TASF)	5/6	6/4	0.41
Preop C0–C1 angle ( $^\circ$ )	8.4 $\pm$ 4.4	11.2 $\pm$ 6.7	0.08
Preop C1–C2 angle ( $^\circ$ )	17.5 $\pm$ 5.6	19.3 $\pm$ 6.2	0.72
Preop C2–C7 angle ( $^\circ$ )	22.2 $\pm$ 3.4	21.3 $\pm$ 3.4	0.45
Final C0–C1 angle ( $^\circ$ )	3.8 $\pm$ 1.2	6.2 $\pm$ 3.4	$< 0.05$
Final C1–C2 angle ( $^\circ$ )	15.78 $\pm$ 3.8	28 $\pm$ 5.4	$< 0.05$
Final C2–C7 angle ( $^\circ$ )	7.7 $\pm$ 9.9	20.0 $\pm$ 7.2	$< 0.05$

Values are presented as mean $\pm$ standard deviation or number.

PSLW, posterior sublaminar wiring; TASF, trans-articular screw fixation using Magerl's technique; Preop, preoperative.

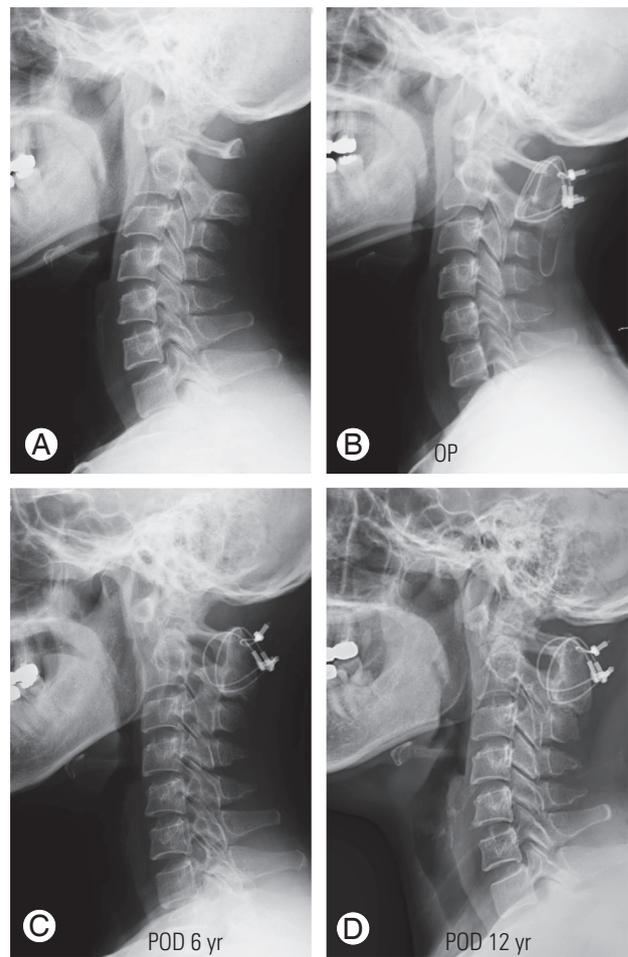


**Fig. 3. (A–D)** A 44-year-old man with Os odontoideum underwent C1–C2 posterior sublaminae wiring and fusion. The C1–C2 angle was overcorrected from 23° preoperatively to 30° at the final follow-up. The C0–C1 angle decreased from 10° to 9° preoperatively, whereas the C2–C7 angle decreased from 10° to –5° at 2 years postoperatively. OP, operative; POD, postoperative day.

This retrospective study was approved by the Institutional Review Board of the Uijeongbu St. Mary's Hospital (e-IRB UC17RESI0145) and informed consent was waived.

## Results

The C1–C2 angle was increased from  $18.4^{\circ} \pm 5.8^{\circ}$  preoperatively to  $21.6^{\circ} \pm 9.1^{\circ}$  at the final follow-up. However, this improvement was not statistically significant ( $p=0.924$ ). The C0–C1 angle was reduced from  $9.73^{\circ} \pm 7.2^{\circ}$  preoperatively to  $4.9^{\circ} \pm 2.3^{\circ}$  at the final follow-up ( $p<0.05$ ). The C2–C7 angle was also significantly decreased from  $21.8^{\circ} \pm 13.2^{\circ}$  to  $13.3^{\circ} \pm 10.6^{\circ}$  ( $p<0.05$ ). The final C1–C2 angle was negatively correlated with the final C0–C1 angle (correlation



**Fig. 4. (A–D)** A 36-year-old woman with Os odontoideum underwent C1–C2 posterior sublaminae wiring and fusion. The C1–C2 angle was maintained from 20° preoperatively to 20° at 12 years postoperatively. Sagittal alignment was well maintained at a C0–C1 angle of 10° and C2–C7 angle of 22° at 12 years postoperatively. OP, operative; POD, postoperative day.

coefficient,  $-0.547$ ;  $p<0.05$ ) and final C2–C7 angle (correlation coefficient,  $-0.705$ ;  $p<0.01$ ). Following atlantoaxial fusion, the C1–C2 ROM ( $16.6^{\circ} \pm 9.8^{\circ}$  versus  $0^{\circ}$ ,  $p<0.001$ ), C0–C1 ROM ( $16.6^{\circ} \pm 4.4^{\circ}$  versus  $8.7^{\circ} \pm 3.8^{\circ}$ ,  $p<0.05$ ), and C2–C7 ROM ( $38.9^{\circ} \pm 20.6^{\circ}$  versus  $31^{\circ} \pm 15.2^{\circ}$ ,  $p<0.05$ ) were significantly decreased. The final C0–C1 angle ( $3.8^{\circ} \pm 1.2^{\circ}$  versus  $6.2^{\circ} \pm 3.4^{\circ}$ ,  $p<0.05$ ) and the C2–C7 angle ( $7.7^{\circ} \pm 9.9^{\circ}$  versus  $20.0^{\circ} \pm 7.2^{\circ}$ ,  $p<0.05$ ) were smaller in group A than in group B ( $p<0.05$  for both) (Figs. 3, 4, Table 1).

## Discussion

Atlantoaxial articulation is an anatomic feature with a full ROM in the spine that is dependent for its stability on lig-

amentous support and the integrity of the odontoid. AAI etiology includes trauma, congenital malformations, OO, RA, malignancy, and skeletal dysplasia. OO is an anomaly defined as an ossicle with smooth circumscribed margins and no osseous continuity with the C2 body [1,2,4,16]. OO patients with instability can be asymptomatic or can present several symptoms, including occipitocervical pain alone, myelopathy, or intracranial symptoms from vertebrobasilar ischemia [16]. Clinically, patients with OO experience severe neck pain and myelopathy due to craniocervical instability and spinal cord compression. In our study, eight of the 21 subjects complained of neck or suboccipital pain, whereas 13 complained of myelopathy.

The surgical techniques to address OO vary from sublaminar wiring to segmental fixation. The early surgical techniques of Brooks and Jenkins [6] and Gallie [5] achieved variable fusion results [17]. Lowry et al. [7] described a 20% non-union rate with Brooks-type atlantoaxial fusion in patients with OO. To achieve successful fusion, stability reconstruction between C1 and C2 combined with bone grafting is necessary. Transarticular screw fixation with strut grafts produces superior fusion rates than those with sublaminar wiring. Based on a large series of 121 patients, nine of whom had OO, Dickman and Sonntag [17] **showed a 98% fusion rate with this technique** versus a rate of 86% with posterior wiring. Moreover, Farey et al. [18] showed a 33% instability rate with the Gallie technique using immobilization versus 100% with arthrodesis using transarticular screws in 27 patients, six of whom had OO. To our knowledge, only one study has shown good fusion with this technique [19]. The latest generation of posterior spinal instrumentation is the polyaxial screw-rod system. This approach has been applied to atlantoaxial fusion by Harms and Melcher [11] in a series of 6 patients with OO.

Although there are several reports on C1–C2 fusion, few studies have focused on the association between C1–2 fixation angle and postoperative subaxial sagittal alignment change. It has been reported that subaxial kyphosis after posterior C1–2 fusion is associated with an increased C1–C2 fusion angle [12,14,19–23]. Yoshimoto et al. [12] documented that in any type of C1–C2 posterior fusion surgery, C1–C2 fixation in a hyperlordotic position led to postoperative subaxial kyphosis. Ishii et al. [20] also found that excessive correction of the C1–C2 angle may cause postoperative hyperlordosis and lead to the development of subluxations of subaxial spine in RA. Huang et

al. [21] investigated the cervical sagittal alignment in non-rheumatoid arthritic patients following posterior C1–C2 fusion and identified a correlation between the C1–C2 fusion angle and postoperative cervical sagittal alignment. They also found that posterior C1–C2 fusion in hyperextension may lead to kyphotic change of the atlanto-occipital alignment and increase the forward tilt of the cervical spine. Our results also showed smaller final C0–C1 and C2–C7 angles in the C1–C2 hyperlordotic group (C1–C2 fusion angle  $\geq 22^\circ$ ) than in the group with C1–C2 fusion angle  $< 22^\circ$ . Further analyses could potentially reveal that increased posterior C1–C2 fusion angle is associated with kyphotic change of occipitocervical and subaxial alignments in patients with OO. Unless the corrected C1–C2 angle is satisfied, surgeons should attempt to make an adjustment during the operation. Matsumoto et al. [22] recommended the application of structural interlaminar spacers instead of autologous bone that can maintain a proper cervical angle.

Thus, there is still no consensus regarding the ideal C1–C2 fusion angle. Nojiri et al. [23] stated that the mean C1–C2 angle in healthy men is  $26.5^\circ$  and that in healthy women is  $28.9^\circ$ . Moreover, several investigators have noted that the optimum C1–C2 angle for C1–C2 fixation should be approximately  $20^\circ$  [14,24]. Kato et al. [24] regarded over-reduction as a C1–C2 angle reduction of  $>20^\circ$  [25]. Huang et al. [21] **also commented the change of postoperative C2–C7 angle between final C1–C2 angle less than  $20^\circ$  group and more than  $22^\circ$  group.** They showed that patients with a correction of  $>20^\circ$  in the C1–C2 angle are more likely to present a significant postoperative loss of cervical lordosis. We believe that in OO patients with AAI, C0–C1 and C2–C7 segments have been adapted to the C1–C1 kyphosis for a long time. In the present study, the preoperative C1–C2 angle was  $18.4^\circ$ . Therefore, an acute overcorrection of C1–C2 kyphosis can negatively affect C0–C1 and C2–C7 in the long-term follow-up. Based on our results, we postulate that fusing the C1–C2 angle at  $<22^\circ$  can decrease the likelihood of sagittal malalignment of C0–C1 and C2–C7.

This study has certain limitations. First, it is a retrospective case series with a relatively small study population owing to the rarity of this pathologic condition. However, to our knowledge, no study has evaluated  $>20$  cases of OO, especially using a traditional sublaminar wiring technique and/or transarticular screw fixation. Second, we did not include a control group for comparison, such

as patients undergoing C1–C2 polyaxial screw fixation. Huang et al. evaluated the relation between the C1–C2 fusion angle and postoperative cervical alignment using C1–C2 segmental fixation [21]. They also reported that over-reduction of the C1–C2 angle and hyperlordotic C1–C2 fusion leads to sagittal malalignment. Therefore, the results of the present study are also applicable for conventional C1–C2 segmental fixation technique.

## Conclusions

We found a negative association between the sagittal alignment of C0–C1 and C2–C7 after atlantoaxial fusion and the C1–C2 fusion angle along with decreased ROM. Thus, overcorrection of C1–C2 kyphosis should be avoided to maintain good physiologic cervical sagittal alignment.

## Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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