The Spine Journal 19 (2019) 1362–1368

Clinical Study

Impact of the occiput and external acoustic meatus to axis angle on dysphagia in patients suffering from anterior atlantoaxial subluxation after occipitocervical fusion

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Received 28 January 2019; revised 4 April 2019; accepted 4 April 2019

Abstract

BACKGROUND CONTEXT: Dysphagia is a complication that sometimes occurs after occipitocervical fusion (OCF). An appropriate O-C2 angle (O-C2a) is recognized as a critical factor for preventing dysphagia. The occiput and external acoustic meatus to axis angle (O-EAa) has some advantages over the O-C2a and is now recognized to outperform O-C2a in predicting dysphagia. However, there are no data on this topic from patients with anterior atlantoaxial subluxation (AAS).

PURPOSE: To evaluate the relationship between the O-EAa and dysphagia in patients suffering from AAS after OCF surgery.

STUDY DESIGN: A retrospective clinical study.

PATIENT SAMPLE: Data from 22 consecutive AAS patients who had undergone OCF were reviewed retrospectively.

OUTCOME MEASURES: The outcome measures included the O-EAa, O-C2a, the narrowest oropharyngeal airway space (nPAS), and the morbidity of dysphagia after OCF.

METHODS: Between September 2011 and September 2017, data from 22 consecutive AAS patients who had undergone OCF were reviewed retrospectively. The patients were divided into two groups according to whether they had suffered postoperative dysphagia by face-to-face questioning or telephone interview. Lateral radiographs were analyzed to determine the pre- and postoperative O-EAa, O-C2a, angle formed by the inferior endplate of C2 and the EA-line (C2Ta), and smallest anteroposterior diameter of the oropharynx between the levels of the uvula and the tip of the epiglottis (nPAS).

RESULTS: The incidence of dysphagia after OCF was 18.18% (4/22). The pre- and postoperative mean nPAS values were significantly different between the groups ($p<.05$). The postoperative mean O-EAa of the group with dysphagia was significantly smaller than that of the group without dysphagia ($p<.05$). The mean change in nPAS was significantly larger in the group with dysphagia than that in the group without dysphagia ($p<.05$). The changes in the O-EAa, O-C2a, and nPAS were linearly correlated within patients. The marginal R^2 values for the patients were 0.452 and 0.202 for the O-EAa and O-C2a, respectively.

CONCLUSIONS: The O-EAa impacts dysphagia in patients with AAS after OCF. Measuring this angle intraoperatively may be a simple and effective procedure. The O-EAa may be used as a practical index to avoid postoperative dysphagia in patients with AAS after OCF.

Keywords: Anterior atlantoaxial subluxation; Occipitocervical fusion; Complication; Dysphagia

FDA device/drug status: Not applicable.

Author disclosures: ***TC:*** Nothing to disclose. ***XY:*** Nothing to disclose. ***WK:*** Nothing to disclose. ***ZL:*** Nothing to disclose. ***YS:*** Nothing to disclose.

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https://doi.org/10.1016/j.spinee.2019.04.002

Introduction

Occipitocervical fusion (OCF) can provide immediate rigidity and fusion for the treatment of congenital deformities, trauma, rheumatoid arthritis (RA), and degenerative processes, but this procedure may lead to craniocervical junction instability. This technique provides excellent fixation and has increased fusion rates [1]. However, complications often occur after OCF. Events include surgical site and donor site complications such as hematoma, infection, and pain [2]. Malalignment of the craniocervical junction may result in dysphagia and dyspnea, which influence patients' daily lives and occasionally threaten their survival [3,4]. Previous studies have suggested that the etiology of upper airway obstruction after OCF has multiple aspects, including edema of the airway, reduction of anterior atlantoaxial subluxation (AAS), and a decrease in the O-C2 angle (O-C2a) [5−7]. Oropharyngeal stenosis resulting from a decrease in the O-C2a is considered a major factor [8,9]. Therefore, some scholars believe that maintaining the O-C2a at or above its preoperative value can prevent postoperative dyspnea and dysphagia after OCF [10].

However, measurement of the O-C2a has some shortcomings that affect its effectiveness. First, the O-C2a consists of McGregor's line and the inferior endplate line of C2. Variation in the endplate line of C2 produces variation in the O-C2a [11]. Second, the O-C2a does not reflect the translation of the cranium in relation to C2 in the process of reducing AAS, and this affects the narrowest oropharyngeal airway space (nPAS), which is defined as the smallest anteroposterior diameter of the oropharynx between the levels of the uvula and the tip of the epiglottis. Increasing or maintaining the O-C2a is not sufficient to predict variation in the nPAS [6]. Morizane et al. suggested that the occipital and external acoustic meatus to axis angle (O-EAa) can compensate for the above shortcomings of the O-C2a [12]. However, data are currently lacking regarding the utility of the O-EAa in the context of AAS. Consequently, the purpose of this investigation was to evaluate the relation between the O-EAa and dysphagia in patients suffering from AAS after OCF surgery via a retrospective clinical study.

Materials and methods

Patients

This study was approved by the ethics board committee of our center. This investigation was a retrospective study of 22 consecutive AAS patients who underwent OCF between September 2011 and September 2017. AAS was defined as an anterior atlantodental interval of 3 mm or more among adults (≥18 year) or 5 mm or more in younger individuals in the neutral or extension position [6,13].

The mean patient age was 47.19 years (range, 14−76 years), and the mean follow-up time was 17.19 months (range, 12−24 months). The comorbidities included basilar invagination (n=15), nonunion of odontoid fractures (n=4), and RA (n=3). Patients with Morquio syndrome, osteogenesis imperfecta, or Down syndrome were excluded from this study. Ventral compression of the cervicomedullary junction was examined in these patients via MRI, and all patients were classified as having a reducible dislocation (Type A) [14]. We routinely used the autologous iliac bones for fusion. The fusion levels were O-C2 (n=17), O-C3 (n=3), and O-C4 (n=2). Dysphagia was defined as patients complaining of swallowing difficulty or swallowing requiring extra effort [15]. Regarding the classification of dysphagia, patients with no complaints of swallowing difficulty were classified as "None"; patients who experienced rare, intermittent episodes of dysphagia were classified as "Mild"; patients with occasional swallowing difficulty for specific foods were classified as "Moderate"; and patients with frequent swallowing difficulties for the majority of solid foods and liquids were classified as "Severe" [16]. Table 1 lists the patients' demographic information. The inclusion criteria were as follows: (1) patients suffered from AAS and were undergoing OCF; (2) complete pre- and postoperative records were available; and (3) the follow-up period was longer than 12 months. The exclusion criteria were as follows: (1) dysphagia was present preoperatively; (2) information on pre- or postoperative swallowing assessment was not provided; (3) patients had a previous history of oropharyngeal surgery; and (4) dysphagia resulting from esophagus injury.

Radiographic measurements

Pre- and postoperative plain lateral radiographs of all patients were examined to calculate the O-EAa, O-C2a, C2Ta, and nPAS (Fig. 1). The O-EAa is defined as the angle formed by the McGregor line and the EA-line, which connects the midpoint of the external acoustic meatuses and the midpoint of the inferior endplate of C2 [12]. The O-C2a is defined as the angle formed by the McGregor line and the inferior endplate of C2. The C2Ta is defined as the angle formed by the inferior endplate of C2 and the EA-line. The nPAS is defined as the smallest anterior-posterior diameter of the oropharynx between the levels of the uvula and the epiglottis tip.

To analyze the correlations of the O-EAa, O-C2a, and C2Ta with the nPAS, we calculated the dO-EAa, dO-C2a, dC2Ta, and dnPAS as follows: dO-EAa=postoperative

Table 1
Patient demographic data

	Dysphagia	No dysphagia	p Value
Number of cases	4	18	
Gender(M/F)	1:3	7:11	.503*
RA:non-RA	2:2	1:17	.073*
Follow-up (mo)	18.75±3.77	16.83±3.78	.300†
Mean age (y)	51.25±8.85	46.28±19.21	.774†

* Fisher's exact probability test.
† Mann-Whitney *U* test.

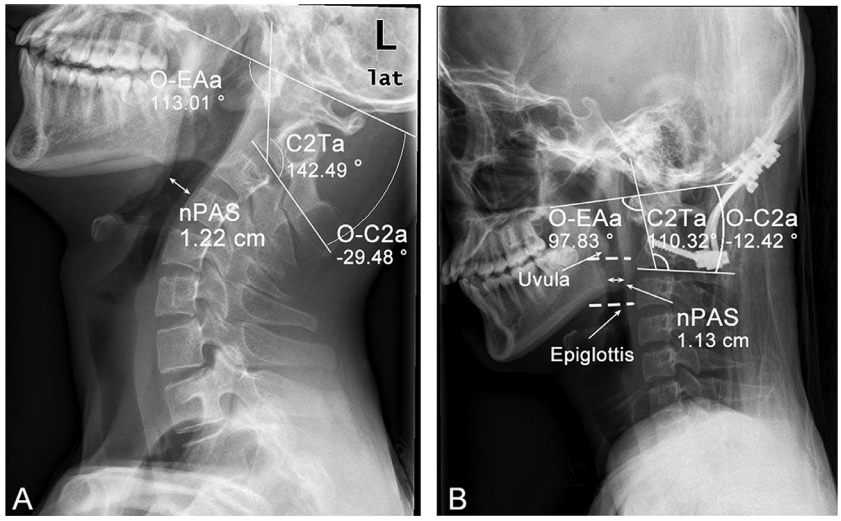

Fig. 1. Representation of radiographic measurements. O-EAa is defined as the angle formed by McGregor's line and the line connecting the external acoustic meatus to the midpoint of the caudal endplate of C2. C2Ta is defined as the angle formed by the inferior endplate of C2 and a line connecting the external acoustic meatus with the midpoint of the caudal endplate of C2. O-C2a is defined as the angle formed by McGregor's line and a line extending through the caudal endplate of C2. nPAS is defined as the narrowest anteroposterior distance from the posterior pharyngeal wall to the back of the tongue, between the tip of the uvula and the tip of the epiglottis. In this case, the preoperative (A) and postoperative (B) values of O-EAa were 113.01° and 97.83°, those of O-C2a were −29.48° and −12.42°, those of C2Ta were 142.49° and 110.32°, those of nPAS were 1.22 cm and 1.13 cm, respectively.

O-EA angle−preoperative O-EA angle, dO-C2a=postoperative O-C2 angle−preoperative O-C2 angle, dC2Ta=postoperative C2T angle−preoperative C2T angle, and dnPAS=postoperative nPAS−preoperative nPAS.

Statistical analysis

The Mann-Whitney *U* test was used to compare continuous variables, and Fisher's exact probability test was used to compare categorical variables. We used simple linear regression to determine whether the dO-EAa and dO-C2a were correlated with the dnPAS. SPSS 19.0 software (SPSS Inc., Chicago, IL) was used for the statistical analysis. A p value <.05 was considered to indicate statistical significance.

Results

Retrospective analysis of medical records during hospitalization revealed that among the 22 included patients, 4 patients developed different degrees dysphagia at 1 to 2 weeks after OCF. Thus, the incidence of dysphagia was 18.18% (4/22). Table 1 lists the patients' demographic data. No significant difference was observed between the two groups in terms of gender, age, the proportion of patients with RA, or the average follow-up period (p>.05). Table 2 lists the clinical data of patients with postoperative dysphagia. According to the dysphagia scale, two patients had "Moderate" dysphagia, one patient had "Severe" dysphagia, and one patient had "Mild" dysphagia. In two patients, the dysphagia remitted at 4 months and 6 months after OCF. The other two patients continued to complain of swallowing difficulty, but the dysphagia was considered "Mild" at follow-up. Table 3 lists the pre- and postoperative radiographic parameters. There were significant differences between the two groups in terms of preoperative nPAS, postoperative O-EAa, postoperative nPAS, dnPAS, dC2Ta, and dO-C2a (p<.05). The mean pre- and postoperative nPAS values were significantly lower in the group with dysphagia than in the group without dysphagia. The postoperative mean

Table 2
Clinical data of patients with postoperative dysphagia

Case no.	Age (y)	Gender	Original morbidity	Dysphagia scale	Duration (mo)	Dysphagia scale at the final follow-up	Po-O-EAa (°)	Po-nPAS (cm)
1	39	M	BI	Moderate	15	Mild	94.88	0.70
2	60	F	BI	Moderate	4	None	93.47	0.73
3	54	F	RA	Mild	6	None	96.62	0.71
4	52	F	RA	Severe	18	Mild	92.76	0.64

BI, basilar invagination; RA, rheumatoid arthritis; Po-O-EAa, postoperative O-EA angle; Po-nPAS, postoperative minimum width of the oropharyngeal airway space.

Patients with no complaints of swallowing difficulty were classified as "None." Patients who experienced rare, intermittent episodes of dysphagia were classified as "Mild." Patients with occasional swallowing difficulty for specific foods were classified as "Moderate." Patients with frequently swallowing difficulties for the majority of solid foods and liquids were classified as "Severe."

Table 3
Patient radiographic outcomes (mean±SD)

	Dysphagia (n=4)	Without dysphagia (n=18)	p Value
Pre-O-EAa (°)	106.89±4.76	108.15±4.67	.594
Pre-C2Ta (°)	97.69±3.9	111.89±13.69	.066
Pre-O-C2a (°)	9.2±7.88	−3.74±11.48	.141
Pre-nPAS (cm)	1.2±0.12	1.72±0.34	.007*
Po-O-EAa (°)	94.43±1.7	97.63±3.16	.010*
Po- C2Ta (°)	93.33±7.93	91.71±7.17	.195
Po-O-C2a (°)	1.1±7.24	5.93±8.67	.141
Po-nPAS (cm)	0.7±0.04	1.4±0.3	.000*
dO-EAa (°)	−12.46±5.6	−10.51±6.25	.837
d C2Ta (°)	−4.36±8.25	−20.18±12.08	.001*
dO-C2a (°)	−8.1±7.3	9.67±11.49	.000*
dnPAS (cm)	−0.5±0.08	−0.32±0.17	.019*

SD, standard deviation; Pre-O-EAa, preoperative O-EA angle; Pre-O-C2a, preoperative O-C2 angle; Pre-nPAS, the preoperative narrowest oropharyngeal airway space; Po-O-EAa; postoperative O-EA angle; Po-O-C2a, postoperative O-C2 angle; Po-nPAS, postoperative minimum width of the oropharyngeal airway space; dO-EAa, the difference between postoperative and preoperative O-EA angle; dO-C2a, difference between the postoperative and preoperative O-C2 angles; dnPAS, the difference between postoperative and preoperative minimum width of the oropharyngeal airway space.

* p<.05.

O-EAa value (mean±standard deviation, 94.43°±1.7°) of the group with dysphagia was significantly lower than that of the group without dysphagia (mean±standard deviation, 97.63°±3.16°). The mean dnPAS of the group with dysphagia was significantly greater than that of the group without dysphagia.

A linear correlation was found between the dO-EAa and dnPAS (R=0.672, R^2=0.452, p=.001), as shown by the distributions in Fig. 2. Additionally, as shown in Fig. 3, a linear correlation was found between the dO-C2a and dnPAS (R=0.450, R^2=0.202, p=.036). The R value of the former correlation was greater than that of the latter.

Case presentation

A 52-year-old female with comorbid RA presented with headache and gradual motor weakness in the bilateral lower and upper extremities. Plain radiographs demonstrated irreducible AAS (Fig. 4). MRI displayed cervical cord compression at the level of the posterior arch of C1. The preoperative O-EAa and nPAS were 113.06° and 1.04 cm, respectively on plain lateral radiographs (Fig. 4A). The patient underwent intraoperative reduction, decompression of the spinal cord by resection of the posterior arch of the atlas, and OCF with an iliac bone graft; the joint was fixed with a screw-rod instrument attached to the C2 to C3 lateral masses. After surgery, the patient's headache and motor weakness symptoms were relieved, but she developed dysphagia, which continued throughout the 1-year follow-up period. The postoperative O-EAa and nPAS were 92.76° and 0.64 cm, respectively (Fig. 4B).

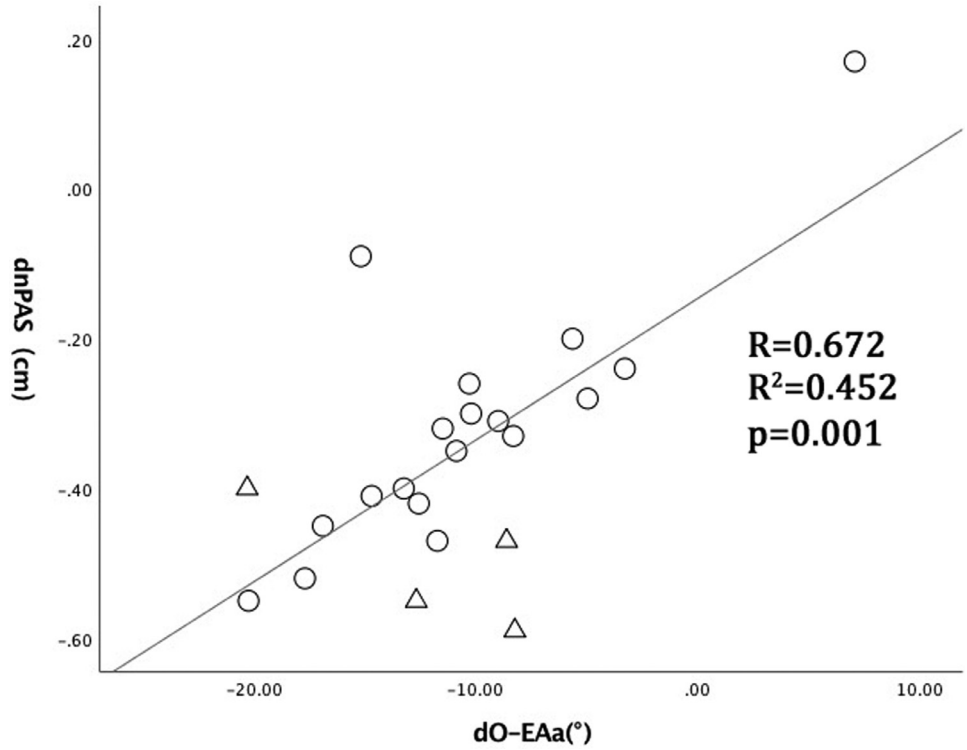

Fig. 2. The distribution of dO-EAa and dnPAS. The black-lined triangles represent cases of dysphagia, and the black-lined circles represent cases without dysphagia. dO-EAa, difference between the postoperative and preoperative O-EA angles; dnPAS, difference between the postoperative and preoperative minimum widths of the oropharyngeal airway space.

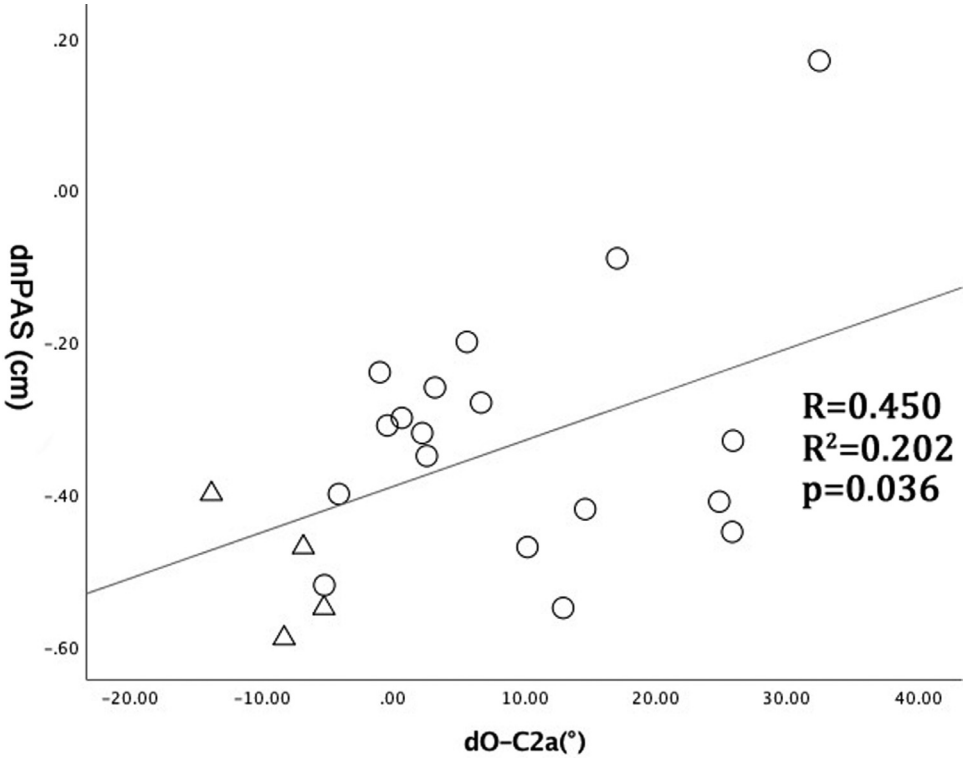

Fig. 3. The distribution of dO-C2a and dnPAS. The black-lined triangles represent cases of dysphagia, and the black-lined circles represent cases without dysphagia. dO-C2a, difference between the postoperative and preoperative O-C2 angles; dnPAS, difference between the postoperative and preoperative minimum widths of the oropharyngeal airway space.

Discussion

OCF can provide immediate rigidity and fusion via the introduction and development of screw-rod instrumentation. The main indication for OCF is occipitocervical junction instability caused by trauma, congenital malformation, tumor, infection, or RA [1]. In our study, OCF was also performed in the case of irreducible atlantoaxial dislocation or resection of the posterior arch of C1. Complications such as postoperative dysphagia and dyspnea often occur after OCF. Dyspnea is one of the most severe complications of OCF. Recent reports in the literature indicate that the incidence of dysphagia after OCF is 15.8% or 17.6% [17,18]. Some patients continue to suffer from dysphagia during the follow-up period. Therefore, the ability to predict the risk of dysphagia in individual OCF recipients would be clinically significant. In our study, four patients complained of dysphagia after surgery. Two patients achieved remission, while the other two patients still suffered from dysphagia at the last follow-up visit.

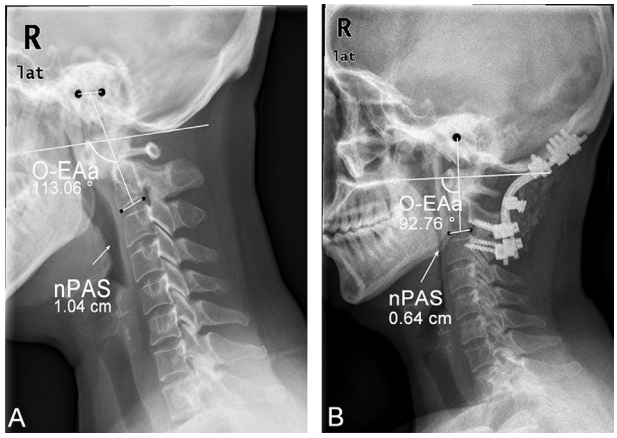

Fig. 4. A case of postoperative dysphagia. (A) The preoperative values of O-EAa and O-C2a were 113.06° and 20.47°, and nPAS was 1.04 cm. (B) The patient suffered from obvious dysphagia after surgery. Postoperative X-rays after surgery showed that O-EAa and O-C2a were 92.76° and 6.64°, respectively. The oropharyngeal space developed obvious stenosis, and nPAS decreased to 0.64 cm. O-EAa, O-EA angle; O-C2a, O-C2 angle; nPAS, the narrowest oropharyngeal airway space.

What causes dysphagia after OCF? In the literature, the etiology of OCF is speculated to be closely related to oropharyngeal stenosis [3,4]. The O-C2a is positively linearly correlated with the oropharyngeal airway space and can be a practical predictor of postoperative dyspnea and dysphagia [10]. Ota et al. [7] proposed that as the O-C2a decreases, the mandible shifts posteriorly, resulting in oropharyngeal stenosis and dysphagia. Izeki et al. [6] suggested that reduction of AAS also causes the mandible to move posteriorly and reduces the oropharyngeal space; they suggest the need for a new marker that can indicate not only the variation in the O-C2a but also the translation of C1−C2 at the same time. The novel O-EAa parameter can be used to capture both O-C2a variation and translation of the occipital bone or C1−C2 [12]. The present study showed that the

postoperative mean O-EAa value was significantly smaller in the group with dysphagia than in the group without dysphagia, and the dO-EAa and dO-C2a were linearly correlated with the dnPAS in all AAS patients. In addition, the dO-EAa was superior to the dO-C2a, as evaluated by the marginal R^2. These results imply that the O-EAa improved the prediction of postoperative nPAS in all AAS patients who underwent OCF. The postoperative O-EAa value (mean±standard deviation) was 97.63°±3.16° in the group without dysphagia and 94.43°±1.7° in the group with dysphagia. We can infer that patients with postoperative O-EAa values ranging from 96.13° to 100.79° will not develop dysphagia. In clinical application, we suggest that preserving a postoperative O-EAa value of 100° will help prevent unexpected postoperative dysphagia in patients with AAS after surgery. Notably, the postoperative O-EAa values in this study are larger than the previously reported neutral-position O-EAa values of healthy people (mean 90.0°).

The O-C2a has a limited ability to predict the dnPAS in patients suffering from AAS. There are several reasons for this result. First, the O-EAa is defined as the sum of the O-C2a and C2Ta. That is, the O-C2a is an integral part of the O-EAa. Second, the O-C2a has more interindividual variation than the O-EA, and it does not reflect the process of atlantoaxial reduction when it includes translation of the cranium in relation to C2 [12]. Third, the O-C2a is formed by McGregor's line and the inferior endplate line of C2. The O-C2a decreases significantly with the inclination of C2 in patients with severe atlantoaxial dislocation, which results in inconsistency between the variation of the dO-C2a and that of the dnPAS (Fig. 1). In this study, a positive correlation was found between the dO-EAa and dnPAS, as shown in the scatter plot. Linear regression analysis identified the dO-EAa as the strongest correlate of the dnPAS, showing that the oropharyngeal airway space is reduced as the O-EAa decreases, while only a weak correlation was observed between the dO-C2a and dnPAS. The nPAS was still decreased when the O-C2a was at its largest value during atlantoaxial reduction in patients with AAS [6].

The process of dysphagia following OCF involves the mandible shifting posteriorly, resulting in mechanical stenosis of the oropharyngeal space. Our study showed that the pre- and postoperative mean nPAS values were lower in the group with dysphagia than in the group without dysphagia. There are many reasons for this difference. RA is a considerable risk factor for dysphagia after surgery. Some researchers have reported that the rate of dysphagia after OCF is 60% in patients suffering from RA [10]. Compression of the cervicomedullary intersection may also cause central dysphagia [19]. In the present study, four patients manifested dysphagia after OCF, two patients of whom suffered from RA. The pathomechanism remains unclear. Cricoarytenoid RA, temporomandibular joint destruction, and laryngeal deviation are detected in RA patients [20–22]. These conditions may endanger the airway and render it very difficult to intubate [23]. Some reports have shown that dysmotility of the epiglottis and soft palate may also result in dysphagia after OCF [24].

The limitations of this study are as follows: (1) As the investigation was a retrospective chart review, some collected data such as preoperative information about dysphagia and slight, postoperative swallowing difficulty were not available for all patients. (2) This study had a small sample size. The definitive, ideal O-EAa should be identified by a multicenter retrospective study with a larger sample size.

Conclusion

The O-EAa may be a critical predictor for preventing dysphagia in patients with AAS after OCF. Measuring the O-EAa intraoperatively is a simple and reliable procedure, and maintaining the postoperative O-EAa at approximately 100° is ideal to avoid accidental postoperative dysphagia in AAS patients after OCF.

Acknowledgment

No funds were received in support of this work.

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