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Research Article

External Validity of the Novel Surrogate Measures for Occipitocervical Alignment

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Abstract

Background: Occipitocervical fusion (OCF) is a rare and often challenging surgical procedure. Several methods have been introduced to obtain the best measures for occipitocervical alignment. The mandible-C2 angle was first introduced in 2020. In this study, we aimed to evaluate the out-of-sample validity of these measures.
Methods: We retrospectively studied 274 lateral cervical radiographs of patients aged 1 to 87 years with no cervical pathology evident on X-ray. A board-certified radiologist and a second-year radiology resident performed the measurements on five specific angles as suggested by Bellabarba. The five angles measured consisted of: 1) anterior C2 body/anterior mandible angle (AB/AM), 2) anterior C2 body/posterior mandible angle (AB/PM), 3) posterior C2 body/anterior mandible angle (PB/AM), 4) posterior C2 body/posterior mandible angle (PB/PM), and 5) occipito-C2 angle (OC2A).
Results: Inter-rater correlation data were calculated for single and average measures. The inter-rater agreement for individual angle measures of O-C2A, AB/AM, AB/PM, PB/AM, and PB/PM were 0.49, 0.11, 0.25, 0.33, and 0.49, respectively. The intraclass correlation coefficient (ICC) for average measures of O-C2A, AB/AM, AB/PM, PB/AM, and PB/PM were 0.66, 0.20, 0.40, 0.50, and 0.66, respectively.
Conclusion: Our study did not find statistically significant evidence to confirm that these angles were dependable indicators of occipitocervical alignment, except for the PB/PM angle, which showed a validity comparable to our reference angle.

Keywords: Cervical Vertebrae; Spinal Fusion; Diagnostic Imaging; Radiology

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Background

The occipitocervical junction presents a unique, complex, biomechanical interface between the cranium and the upper cervical spine (1). The gold standard treatment of craniocervical instability is instrumentation and fusion (2). Occipitocervical fusion (OCF) and instrumentation is a technically demanding procedure that has faced several challenges, among which preserving and restoring local anatomy is of utmost importance. Intraoperative restoration of the occipitocervical alignment calls for an adequate knowledge of radiological relations of the anatomical landmarks. Absence or hypoplasia of the occipital bone can make visualization of these landmarks a real challenge (3).

Optimizing the function of gaze, swallowing, and comfort postoperatively necessitates a correct, anatomic occipitocervical alignment. A misaligned relationship of the occiput relative to the cervical spine after fusion can result in disabling complications, including dysphagia, dyspnea, loss of horizontal gaze, and acceleration of adjacent segment degeneration caudal to the stabilization construct. In particular, it has been hypothesized that an occipital-cervical junction fixed in flexion might contribute to severe postoperative dysphagia or lethal dyspnea (4, 5).

Features representing radiographic relations of anatomical landmarks are currently playing a pivotal role in accurately determining occipitocervical alignment and optimizing outcomes. Presently, there is no clear gold standard for establishing the ideal occipitocervical angle for OCF (6).

Occipito-C2 angle (OC2A), owing to its simplicity in terms of measurement method and high reliability, has long been used as a common method for measuring the occipitocervical angle. However, intraoperative visualization of landmarks needed for determining OC2A has been reported to be hindered by the poor intraoperative quality of the imaging (7-10).

Bellabarba et al. have recently introduced new radiographic surrogates for occipitocervical alignment including: 1) anterior C2 body/anterior mandible angle (AB/AM), 2) anterior C2 body/posterior mandible angle (AB/PM), 3) posterior C2 body/anterior mandible angle (PB/AM), 4) posterior C2 body/posterior mandible angle (PB/PM), and 5) OC2A (11).

The performances of these measures in the population from which they were derived have been reported to be excellent. However, no independent study has ever investigated the external validity of those findings. As such, using a sample of the Caucasian population, we aimed to examine the hypothesis that the performance of new measures remains the same across different ethnicities.

Methods

We followed the same methodology as Bellabarba's (11) to make our findings comparable. A few modifications, however, have been applied as follows. We retrospectively studied 274 lateral cervical radiographs of patients aged 1 to 87 years (with 255 aged 18 years or older) without cervical pathology evident on X-ray.

All parameters needed to measure radiographic features were calculated using the standard measuring tools provided by PACS software. Each reviewer measured the parameters of the interest independently without having any knowledge of the other reviewer's data on the same set of subjects.

A board-certified radiologist and a second-year radiology resident performed the measurements pertaining to five specific angles as suggested by Bellabarba et al. (11). The five angles measured consisted of AB/AM, AB/PM, PB/AM, PB/PM, and OC2A.

The OC2A was measured as a gold standard against which to compare the performance of the other suggested radiographic features of the occipitocervical alignment.

In radiographs with a head rotation where shadows for the right and left mandible were separately visualized, angles were calculated for both shadows, with the average being reported. Angles directed cranially were considered positive, while those with caudal direction were considered negative. The number of radiographs where landmarks of interest could not be visualized accurately was registered for each feature.

Data are presented as either mean and standard error (SE) or frequency (%) for continuous and categorical data, respectively. The survey analysis method was followed to take into account the intra-class correlation of data measured by each reviewer. Intraclass correlation coefficient (ICC) was used to measure absolute agreement between individual ratings as well as between the average ratings over different raters. Inter-rater agreement was calculated by developing a series of analyses of variance (ANOVA). P-values denote testing the hypothesis that ICC = 0. P-values under 0.05 were considered significant.

This study was approved by the Ethical Committee of Shahid Beheshti University of Medical Sciences, Tehran, Iran (ethical code: IR.SBMU.REC.1400.148) and written informed consent was obtained from the participants. The authors confirm that all methods were performed following institutional ethical standards and the Declaration of Helsinki.

Results

The mean, SE, and 95% confidence intervals (CIs) of the five angles, including the standard O-C2A, AB/AM, AB/PM, PB/AM, and PB/PM, in the total population and for the patients aged ≥ 18 years, between the two reviewers are summarized in table 1.

Table 1. Mean, standard error (SE), and 95% confidence interval (CI) of the average angle values for the observers in total population and patients aged ≥ 18 years

Angle	Group	Mean (degree)	Linearized	
			SE	95% CI
OC2A	Total population	25.1	0.5	18.6, 31.6
	Patients aged ≥ 18 years	25.0	0.5	18.2, 31.9
AB/AM	Total population	11.0	0.2	8.6, 13.4
	Patients aged ≥ 18 years	10.8	0.3	6.9, 14.7
AB/PM	Total population	10.8	1.0	-1.9, 23.5
	Patients aged ≥ 18 years	10.6	1.2	-4.1, 25.2
PB/AM	Total population	13.5	0.4	8.3, 18.6
	Patients aged ≥ 18 years	13.3	0.5	6.9, 19.7
PB/PM	Total population	17.2	1.8	-5.8, 40.1
	Patients aged ≥ 18 years	16.9	1.9	-6.7, 40.4

OC2A: Occiput-C2 angle; AB/AM: Anterior C2 body/anterior mandible angle; AB/PM: Anterior C2 body/posterior mandible angle; PB/AM: Posterior C2 body/anterior mandible angle; PB/PM: Posterior C2 body/posterior mandible angle; SE: Standard error; CI: Confidence interval

Inter-rater correlation data were calculated for single and average measures. The inter-rater agreement indices for individual angle measures of O-C2A, AB/AM, AB/PM, PB/AM, and PB/PM were 0.49, 0.11, 0.25, 0.33, and 0.49, respectively. Average measures were more reliable than individual measures. For average measures, calculated inter-rater agreement data were rated as excellent if they were between the 0.75 and 1.00 range. The ICC for average measures of O-C2A, AB/AM, AB/PM, PB/AM, and PB/PM were 0.66, 0.20, 0.40, 0.50, and 0.66, respectively. In addition, Cronbach's alpha, another statistical reliability score, demonstrated good results for OC2A, acceptable results for AB/AM, AB/PM, and PB/AM, and questionable results for PB/PM. These measures are summarized in table 2.

Table 2. Interobserver statistical reliability in the total population and patients aged ≥18 years

Angle	Group		Inter-rater agreement	95% CI	P-value	Cronbach's alpha
OC2A	Total population	Individual	0.49	0.39, 0.58	< 0.001	0.70
		Average	0.66	0.56, 0.74		
	Patients aged ≥18 years	Individual	0.47	0.37, 0.57	< 0.001	0.69
		Average	0.64	0.54, 0.72		
AB/AM	Total population	Individual	0.11	-0.01, 0.23	0.039	0.68
		Average	0.20	-0.03, 0.38		
	Patients aged ≥18 years	Individual	0.11	-0.02, 0.23	0.048	0.98
		Average	0.20	-0.04, 0.38		
AB/PM	Total population	Individual	0.25	0.13, 0.36	< 0.001	0.60
		Average	0.40	0.24, 0.53		
	Patients aged ≥18 years	Individual	0.26	0.14, 0.37	< 0.001	0.67
		Average	0.41	0.24, 0.54		
PB/AM	Total population	Individual	0.33	0.21, 0.44	< 0.001	0.60
		Average	0.50	0.35, 0.61		
	Patients aged ≥18 years	Individual	0.34	0.22, 0.45	< 0.001	0.61
		Average	0.51	0.37, 0.62		
PB/PM	Total population	Individual	0.49	0.36, 0.60	< 0.001	0.56
		Average	0.66	0.53, 0.75		
	Patients aged ≥18 years	Individual	0.55	0.40, 0.66	< 0.001	0.57
	Total population	Individual	0.49	0.39, 0.58	< 0.001	0.70

Cronbach's alpha: a ≥0.9: Excellent; 0.8 ≤a < 0.9: Good; 0.7 ≤a < 0.8: Acceptable; 0.6 ≤a < 0.7: Questionable; 0.5 ≤a < 0.6: Poor; a < 0.5: Unacceptable

OC2A: Occiput-C2 angle; AB/AM: Anterior C2 body/anterior mandible angle; AB/PM: Anterior C2 body/posterior mandible angle; PB/AM: Posterior C2 body/anterior mandible angle; PB/PM: Posterior C2 body/posterior mandible angle; CI: Confidence interval

Table 3. The frequency of angles within different ranges from the median

Percentiles	Angle			
	AM/AB (n = 271)	PM/AB (n = 272)	AM/PB (n = 271)	PM/PB (n = 272)
1% (smallest)	1 (0.5)	0.5 (0.3)	0.5 (0.2)	0.6 (0.5)
5% (smallest)	1.4 (0.5)	1 (0.5)	1.6 (0.5)	1.7 (0.5)
10% (smallest)	2.4 (1)	1.6 (0.5)	2.8 (0.5)	3 (0.6)
25% (smallest)	4.9 (1)	3.4 (0.6)	5.6 (0.5)	7.3 (0.6)
50%	9.5	8.45	11	15
75% (largest)	16.1 (35)	13.35 (31.6)	20.5 (42)	22.1 (40)
90% (largest)	22 (40)	21 (32)	30 (42)	27 (42)
95% (largest)	26.8 (41)	25 (34)	34 (43)	33 (42)
99% (largest)	40 (42)	32 (35)	42 (44)	42 (47)
Mean ± SD	11.22 ± 8.17	9.79 ± 7.52	13.88 ± 10.22	15.39 ± 9.69
Variance	66.76944	56.55945	104.4831	93.81234
Skewness	1.086118	1.023126	0.828939	0.518461
Kurtosis	4.190436	3.590758	2.917562	2.835416

AM/AB: Anterior mandible angle/anterior C2 body; PM/AB: Posterior mandible angle/anterior C2 body; AM/PB: Anterior mandible angle/posterior C2 body; PM/PB: Posterior mandible angle/posterior C2 body

Due to the possible use of these measurements during surgery in patients whose baseline alignment was unknown, we investigated the frequency with which our measurement fell within different ranges from the median, and because the data were positively skewed, we reported skewness and kurtosis (Table 3). This is also represented graphically for the five angles (Figure 1).

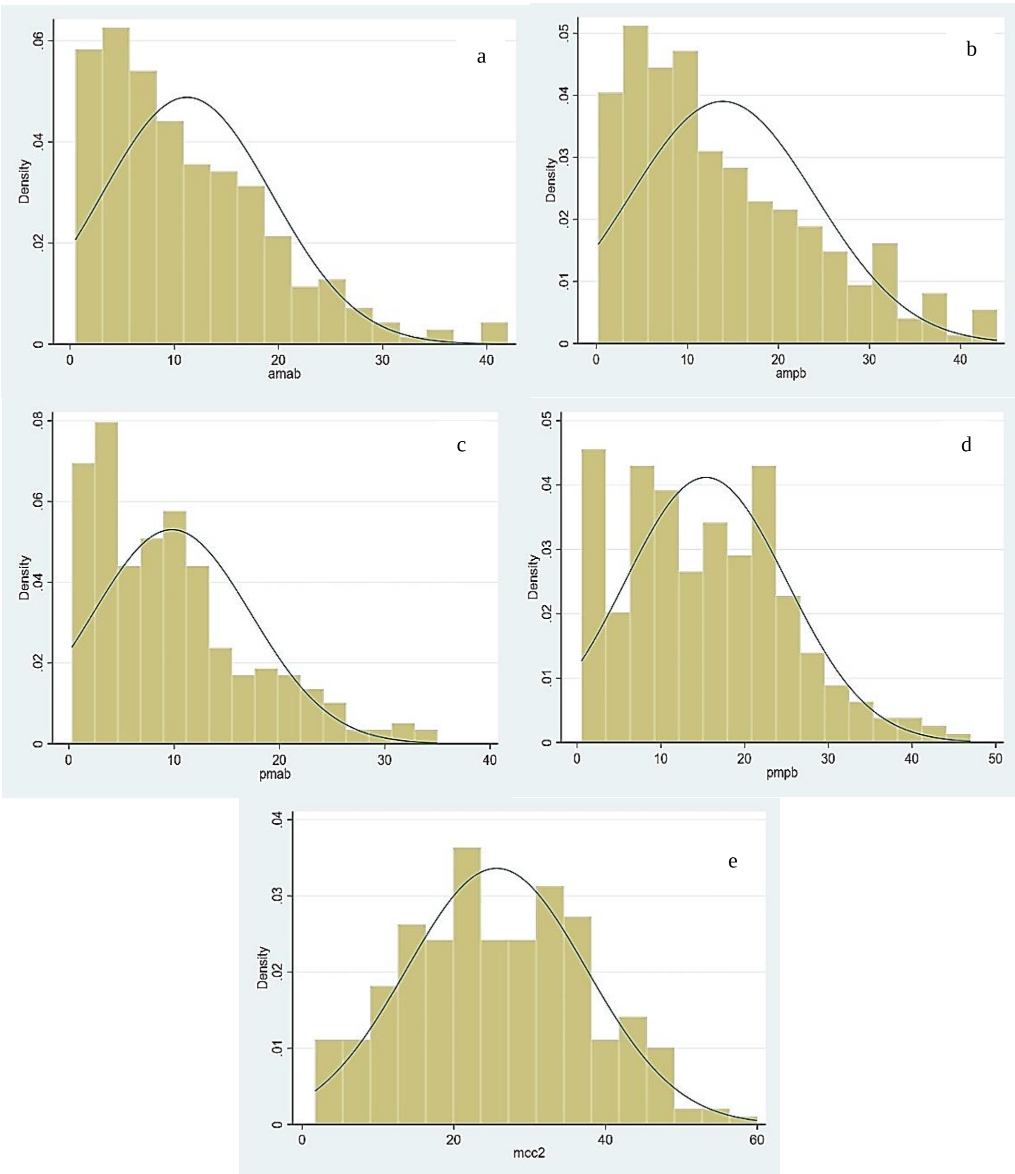

Figure 1. a) Graph demonstrating values for anterior C2 body/anterior mandible angle (AB/AM); b) Graph demonstrating values for posterior C2 body/anterior mandible angle (PB/AM); c) Graph demonstrating values for anterior C2 body/posterior mandible angle (AB/PM); d) Graph demonstrating values for posterior C2 body/posterior mandible angle (PB/PM); e) Graph demonstrating values for occiput-C2 angle (OC2A)

Discussion

We investigated the out-of-sample validity of the newly introduced measures for occipitocervical alignment by comparing those measures to the standard OC2A and observed that only the PB/PM angle could be a viable alternative. However, the other three measures could not keep pace with the OC2A. We hypothesized that the age difference might explain the differences found in the results of the two studies. Therefore, we performed a subgroup analysis among the patients of the same age as the original paper. However, the results remained essentially the same.

In spite of the fact that several measures are being used to define intraoperative neutral occipitocervical relationship, the prevalence of cervical malposition, which results in unfortunate consequences, has not yet been desirably reduced (12).

The OC2A, created by McGregor's line and the inferior surface of the axis, has been frequently used due to its simple measurement method and high reliability and has been reported to be a good predictor of postoperative dyspnea and dysphagia (6, 13).

In contrast, the measurement of OC2A calls for localizing anatomical structures, which are hard to visualize fluoroscopically during operation. Identifying the inferior endplate of the C2 vertebra can also be difficult in cases with deformation resulting from vertebral body fusion, bone spurs, and bone destruction (7, 14).

To tackle these flaws, other radiographic parameters have been described. Riel et al. hallmarked angle formed by the connecting line between the posterior margin of the facet joints in C3 and C4, and the flat area of the occipital protuberance was counted as a reliable measure to define optimal fusion position (15).

Tan et al. described that mandible cervical distance, placed between the midpoint of the two mandibular angles and the anterior border of the C2 body, could be significantly affected by slight head rotation (14). Yoon et al. labeled the angle forming the line connecting the posterior border of the C4 vertebral body and McGregor's line and reported it as a superior method in inter-observer and intra-observer reliability (16).

Most recently, Bellabarba et al. introduced the mandible-C2 angle and five specific angles, including AB/AM, AB/PM, PB/AM, PB/PM, and OC2A, in 2020 (11). In this retrospective study, 100 lateral cervical radiographs of adults without diagnosed pathology were assessed to measure these angles. It is concluded that the radiographic relationship between the mandible and the body and C2 spine can provide a reliable marker for neutral occipitocervical alignment. To our knowledge, these measures have never been formally examined in a sample different from which the original parameters have been developed. Our study was an attempt to examine the out-of-sample validity of the novel parameters of intraoperative occipitocervical alignment. We observed that the performance of these parameters was not as promising as those reported in the original paper.

While several radiologic parameters have been suggested to define the neutral position for OCF, a combination of these parameters has been introduced because each has its shortcomings, and the criteria by which they are defined are ambiguous. Hence, the best option for identifying neutral cervical alignment is yet to be determined.

The findings of the present study need to be interpreted in light of its strengths and limitations. We conducted an out-of-sample analysis for newly introduced measures with a large sample size, as well as a subgroup analysis. Nonetheless, our limitations are as follows. Firstly, we did not include the anthropometric characteristics of the participants (e.g., body size and weight). Second, the number of patients below 18 years might not have been large enough. Third, it should be noted that in our study, we examined radiographs of patients who had no fracture, malalignment, or any other pathologic findings, which were obtained under optimal circumstances, and measurements may be more challenging in post-traumatic and intraoperative settings. However, this limitation is inherent to these types of studies. Lastly, this study is focused on assessing radiograph measurements, and the relationship of optimal radiographic alignment to clinical outcomes should be examined.

Conclusion

We examined out-of-sample performances of recently introduced radiographic parameters as compared to their more traditional counterpart OC2A. Our investigation did not yield statistically significant results to support the use of these angles as reliable indicators of occipitocervical alignment, with the exception of the PB/PM angle, which demonstrated validity that is relatively comparable to our reference angle. Further research is warranted to validate these findings across diverse populations.

Conflict of Interest

The authors declare no conflict of interest in this study.

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