



# Association between three-dimensional measurements of the unilateral cleft lip nasal deformity and maxillary alveolar morphology: A retrospective study

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## KEYWORDS

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Cleft lip nose;  
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Plaster model

**Summary Background:** It is debatable whether rhinoplasty is necessary during a primary operation for cleft lip. However, many surgeons believe that rhinoplasty should be performed simultaneously for severe deformities. We investigated whether alveolar cleft severity is involved in nasal deformity.

**Methods:** Forty-three patients were assessed for alveolar cleft severity using maxillary plaster models prepared during primary cheiloplasty. We conducted morphological assessments of nasal deformities using three-dimensional photogrammetry. Patients were divided into two groups according to alveolar cleft severity: group A, overlap of the alveolar segments; group B, nonoverlap of the alveolar segments. Nasal asymmetry was assessed by measuring distances between landmarks around the nostrils and the columellar angle. These measurements were compared between the groups. The correlations between the columellar angle and the ratios of the five cleft side/non-cleft side distances and the correlation of each ratio were analyzed.

**Results:** Groups A and B included 21 and 22 patients, respectively. Group A demonstrated superior deviation of the alar base on the non-cleft side than that of the alar base on the cleft side ( $p < 0.05$ ). No other statistically significant differences were observed. Group A had more severe nasal deformity. Columellar angle and nostril base width demonstrated correlation.

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**Conclusion:** In an uncorrected, unoperated unilateral cleft lip nasal deformity, alar base deformity is affected by deformity of the alveolar segments.

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## Introduction

Preoperative cleft lip anthropometry is important for drafting therapeutic regimens and assessing outcomes. Studies on results require permanent records of preoperative assessments due to the major effects of primary operations.

Cleft lip plaster casts and models have long been used as preoperative three-dimensional (3D) permanent records of cleft lips.<sup>1</sup> Another recording method is the digitalization of cleft lip casts.<sup>2</sup> Recently, 3D imaging systems have become essential for assessing facial structure.<sup>3</sup> Two of the several systems currently available are the 3dMDface system (3dMD Inc., Atlanta, GA, USA) and VECTRA® H1 handheld imaging system (Canfield Imaging Systems, Fairfield, NJ). Many researchers conducting independent studies with various methods have found the 3dMDface system to be precise.<sup>4</sup> The VECTRA®H1, by comparison with 3dMDface, has been shown to yield highly reproducible measurements and is reliable in clinical settings and research.<sup>5</sup>

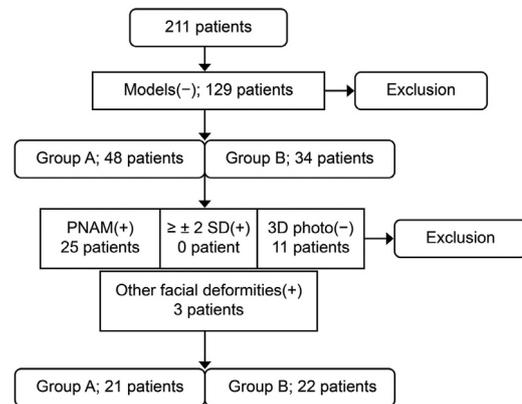
Cleft lip nasal deformities are most likely caused by soft tissue deformities and maxillary hypoplasia on the cleft side.<sup>6</sup> Therefore, preoperative corrections are generally performed to improve these deformities. According to a recent report on relevant surgical techniques, preoperative correction has become more common.<sup>7</sup> Although bilateral symmetry in cleft lip nose deformity is improved by surgical treatment, deformities generally recur as the patient grows. Therefore, assessments of uncorrected hard and soft tissues may help when compared with corrected cases and in treatment that accounts for the patient's growth. In this study, we aimed to assess the relationship between the hard and soft tissues in unoperated, uncorrected unilateral cleft lip nasal deformities.

## Patients and methods

### Patients

The present study was approved by the Showa University Fujigaoka Hospital Institutional Review Board (F2017C79). Written informed consent was obtained from the families for publication of the case and accompanying images. The authors confirm that they adhered to the STROBE guidelines.

Our hospital performs cheiloplasty for infants aged 3-6 months with unilateral cleft lip, alveolus, and palate (UCLP) and unilateral cleft lip and alveolus (UCLA). From December 2014 to February 2018, more than 200 infants with unilateral cleft lip underwent primary repair in our department. We constructed maxillary plaster models and captured images with a 3D camera for patients undergoing primary operation for cleft lip while they were under general anesthesia. After excluding patients for whom either photograph



**Figure 1** Patient background of unilateral cleft lip, alveolus, and palate (UCLP) and unilateral cleft lip and alveolus (UCLA), which could be tracked by medical record information. Models(-); not sampled plaster or difficult to measure due to breakage of plaster. PNAM(+); underwent presurgical nasoalveolar molding. 3D photo(-); photograph was not taken. Other facial deformities(+); syndromic infants.

or maxillary plaster models were not generated, patients with growth curves  $\geq \pm 2$  standard deviations, patients with other facial deformities, and patients who underwent presurgical nasoalveolar molding (PNAM), we included the remaining 43 non-syndromic infants with unilateral cleft lip whose parents or caregivers agreed to participate in this study. **Figure 1** indicates the selection of patients for the study.

### 3D anthropometry

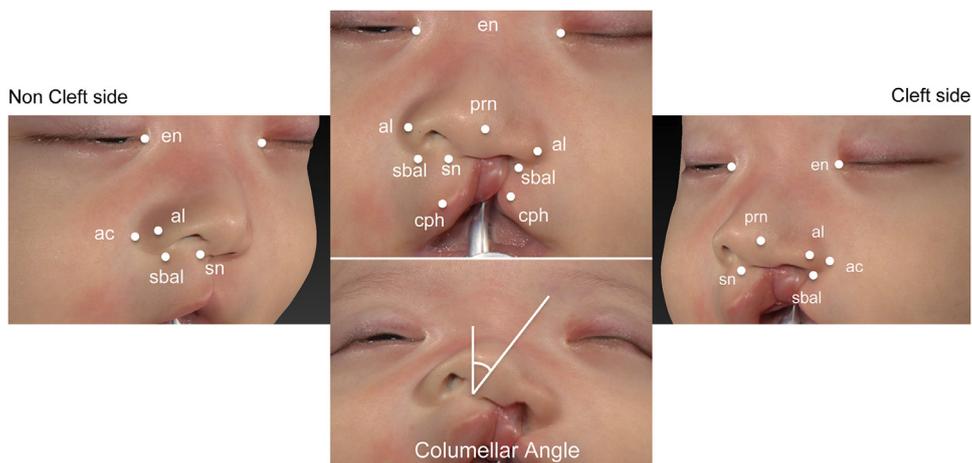
Patients were analyzed under general anesthesia administered just prior to surgery. We constructed maxillary plaster models and performed morphological assessments of nasal deformities using a handheld 3D imaging system (Vectra H-1, Canfield Scientific Co. Ltd., Fairfield, NJ).

Analysis of the 3D images was performed with a software application (Mirror, Canfield Scientific Co. Ltd, Fairfield, NJ) in accordance with the method reported earlier.<sup>8</sup>

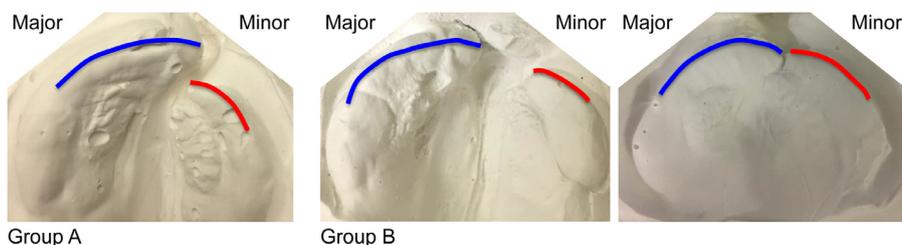
Nasal asymmetry was assessed using anthropometric landmarks (en, ac, sbal, al, sn, prn, and cph) (**Figure 2**).<sup>9,10</sup> Linear distances (en-ac, en-sbal, sbal-sn, and sbal-cph), surface distance (prn-al-ac), and columellar angle were also measured. Measurements were recorded by two observers.

### Measurement using plaster models

We constructed maxillary plaster cast models after capturing the 3D images. Based on the classifications by Pruzansky



**Figure 2** Facial landmarks. Abbreviations: en, endocanthion; ac, alar curvature; sbal, subalare; al, alare; sn, subnasale; prn, pronasale; cph, crista philtri. Lateral alar base height; en-ac, alar base height; en-sbal, nostril base width; sn-sbal, alar length; prn-al-ac, lateral lip height; sbal-cph.



**Figure 3** Severity of the alveolar cleft. Purzansky classification. Blue lines indicate the major segment (non-cleft side). Red lines indicate the minor segment (cleft side). Group A: the major segment overlaps the minor segment. Group B: the major segment does not overlap the minor segment.

et al.,<sup>11</sup> patients were divided into two groups according to alveolar cleft severity: group A demonstrated overlap of the alveolar segments, and group B demonstrated approximation of the alveolar segments without contact or approximation of the alveolar segments with an end-to-end contact (Figure 3).

Groups A and B were compared with regard to nasal asymmetry. The correlations between the columellar angle and the ratios of the five respective cleft side/non-cleft side distances were also analyzed.

### Statistical analysis

The utility of the measurements by the two observers was assessed using the Pearson correlation coefficient, intraclass correlation coefficient, and Wilcoxon signed rank test. The Mann-Whitney U test was used for continuous variables, and Fisher exact test was used for categorical variables. Statistical analysis was performed using IBM SPSS Version 21 (IBM Corp., Armonk, N.Y.). A p-value of  $<0.05$  was considered statistically significant.

## Results

### Patient demographics

Groups A and B consisted of 21 (15 boys, 6 girls) and 22 (15 boys, 7 girls) patients, respectively. In group A, the average age was  $4.8 \pm 1.2$  months, and the cleft was on the right side in five patients and on the left side in 16, and in group B, the average age was  $4.5 \pm 1.1$  months, and the cleft was on the right side in 11 patients and on the left side in 11. In group A, 17 patients had UCLA and four patients had UCLP, and in group B, 11 patients had UCLA and 11 patients had UCLP (Table 1). Although more patients in group A had UCLA than those in group B, this difference was not statistically significant ( $p = 0.07$ ).

### Differences between the measurements by observers

A significant difference was observed in prn-ac on the non-cleft side ( $p = 0.001$ ), representing the shortest surface distance (prn-al-ac). Compared to the nasal alar on the cleft side, that of the non-cleft side showed more undulation and

**Table 1** Patient demographics ( $n = 43$ ).

	Group A (%)	Group B (%)	$p^*$
No.	21	22	
Gender			1.000
Female	6 (28.6)	7 (31.8)	
Male	15 (71.4)	15 (68.2)	
Mean age $\pm$ SD, months	$4.8 \pm 1.2$	$4.5 \pm 1.1$	0.508
Cleft side			0.144
Right	5 (23.8)	11 (50.0)	
Left	16 (76.2)	11 (50.0)	
Type			0.07
Unilateral cleft lip and alveolus	17 (81.0)	11 (50.0)	
Unilateral cleft lip, alveolus, and palate	4 (19.0)	11 (50.0)	

\* Mann-Whitney U test was used for continuous variables and Fisher exact test was used for categorical variables.

**Table 2** List of anthropometric measurements ( $n = 43$ ).

Anthropometric notation	Definition	Measurement	Cleft side (mean $\pm$ SD, mm)	Non-cleft side (mean $\pm$ SD, mm)
en-ac	Linear distance	Lateral alar base height	$21.7 \pm 1.3$	$19.5 \pm 1.3$
en-sbal	Linear distance	Alar base height	$25.9 \pm 1.5$	$24.0 \pm 1.4$
prn-al-ac(prn-ac)	Surface distance	Nasal alar length	$18.5 \pm 1.3$	$20.4 \pm 1.7$
sn-sbal	Linear distance	Nostril base width	$16.2 \pm 2.7$	$7.2 \pm 1.1$
sbal-chp	Linear distance	Lateral lip height	$6.8 \pm 1.4$	$9.8 \pm 1.3$

was different due to a slight misalignment of the al. However, this difference was not substantial, and the correlation was high. No significant differences were found between the two evaluators with regard to all 10 parameters measured.

### Differences in conditions during imaging

3D photography was performed before and after intubation. Therefore, it was necessary to consider the changes in morphology due to intubation; however, in our previous report, no significant difference was observed in the shape of the nasolabial before and after intubation.<sup>12-14</sup> Regarding the potential effect of measurement after intubation, we determined that intubation did not affect the nasal anthropometric measurements.

### Anthropometric measurements

Anthropometric notations, definitions, and measurements in all patients ( $n = 43$ ) are shown in Table 2. Cleft side values that were larger than non-cleft side values showed abnormalities in the vertical position of the alar base height and lateral alar base height (en-ac, en-sbal) and a right-left difference in the width of the nostril base (sn-sbal). Furthermore, cleft side values that were smaller than non-cleft side values showed hypoplasia of the cleft side nasal alar length (prn-al-ac) and lateral lip height (sbal-cph). These results

suggest a conventional uncorrected, unoperated cleft lip nose deformity.

Table 3 shows the results of comparison between groups A and B. The two groups did not differ significantly in terms of nostril base width, alar width, or cleft side alar base. However, group A had a significantly shorter non-cleft side lateral alar base height (en-ac: group A, mean 19.1 mm; group B, mean 20.0 mm,  $p < 0.05$ ) and non-cleft side alar base height (en-sbal: group A, mean 23.3 mm; group B, mean 24.6 mm,  $p < 0.05$ ). In group A, the alar base was deformed in a superior direction (Figure 4).

Table 4 shows correlations of columellar angle ( $n = 43$ ) with the ratios of cleft side/non-cleft side nostril base width (sn-sbal) and alar base height (en-ac, en-sbal), non-cleft side/cleft side nasal alar length (prn-ac), and lateral lip height (sbal-cph) and correlation of each ratio. The columellar angle was strongly correlated with the cleft side/non-cleft side nostril base width ratio ( $r = 0.72$ ,  $p < 0.05$ ) and weakly correlated with the nasal alar length ratios and lateral lip height ratio ( $r = 0.37$ ,  $p < 0.05$ ;  $r = 0.43$ ,  $p < 0.05$ ). The nostril base width ratio weakly correlated with the lateral lip height ratio ( $r = 0.5$ ,  $p < 0.05$ ). The lateral alar base height ratio strongly correlated with the alar base height ratio ( $r = 0.35$ ,  $p < 0.05$ ).

As the cleft side/non-cleft side nostril base width ratio increased, the columellar angle increased. The columellar angle also tended to increase as the vertical deviation of the nasal alar length ratios and lateral lip height ratio increased. By contrast, the columellar angle was not correlated with cleft side/non-cleft side ratios for the alar bases.

**Table 3** Anthropometric measurements of the nose element and comparison between group A and group B.

Anthropometric measurements	Group A (mean±SD)	Group B (mean±SD)	p*
Cleft side (en-ac), mm	21.8 ± 1.4	21.7 ± 1.3	0.54
Cleft side (en-sbal), mm	26.0 ± 1.6	25.9 ± 1.5	0.53
Cleft side (prn-ac), mm	18.4 ± 1.1	18.9 ± 1.6	0.36
Cleft side (sn-sbal), mm	15.9 ± 2.3	15.7 ± 2.7	0.55
Cleft side (sbal-chp), mm	6.7 ± 1.5	6.9 ± 1.3	0.50
Non-cleft side (en-ac), mm	19.1 ± 1.2	20.0 ± 1.1	0.03
Non-cleft side (en-sbal), mm	23.3 ± 1.3	24.6 ± 1.3	0.02
Non-cleft side (prn-ac), mm	19.3 ± 1.5	19.3 ± 1.4	0.29
Non-cleft side (sn-sbal), mm	7.2 ± 1.2	7.5 ± 1.0	0.51
Non-cleft side (sbal-chp), mm	9.9 ± 1.2	9.7 ± 1.3	0.52
Columellar angle, degrees	31.2 ± 8.9	29.3 ± 11.3	0.26

\* Mann-Whitney U test was used for continuous variables.

**Table 4** Correlation of each ratio.

	Correlation Columellar angle (n = 43)
Nostril base width ratio	0.72 <sup>†</sup>
Nasal alar length ratio	0.37 <sup>†</sup>
Lateral alar base height ratio	0.07
Alar base height ratio	0.006
Lateral lip height ratio	0.43 <sup>†</sup>

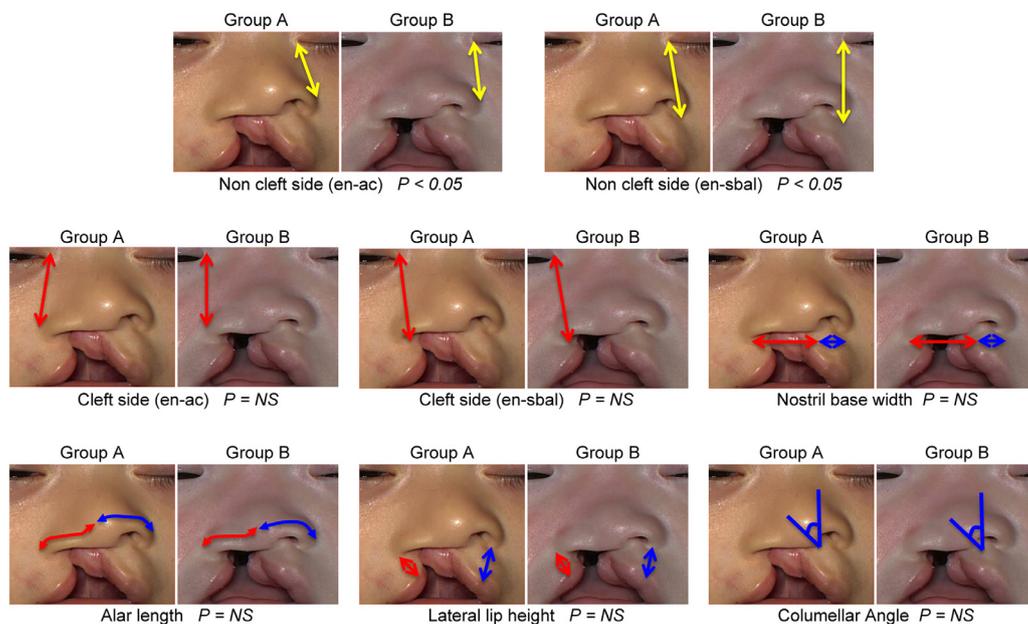
Pearson correlation coefficient.

<sup>†</sup> p < 0.05.

Preoperative orthognathic correction is a concept used to adjust alveolar segments. Methods for preoperative orthognathic correction include passive methods, including the Hotz plate and active methods, including the Latham appliance and the PNAM appliance.<sup>15-17</sup> Correction of the alveolar segments, demonstrated to be involved in cleft lip nasal deformity, is important to achieve bilateral symmetry in cleft lip nasal deformity.<sup>16-18</sup> Barilla et al.<sup>19</sup> compared two groups of patients with UCLP, a group that underwent cleft lip rhinoplasty following the use of a PNAM appliance for 3-4 months (PNAM group) and a group that underwent only surgery without the use of a PNAM appliance (non-PNAM group) in terms of bilateral symmetry in cleft lip nasal deformity during a mean follow-up period of 9 years. Ultimately, bilateral symmetry was achieved in the PNAM group. However, no consensus has been reached regarding the underlying mechanisms by which alveolar cleft severity affect cleft lip nasal deformities.

### Discussion

The present study is the first to demonstrate that alveolar segment deformation affects vertical deviation of the alar base (lateral alar base height, alar base height).



**Figure 4** Anthropometric measurements of the nose element. Blue lines indicate the non-cleft side. Red lines indicate the cleft side. Yellow lines indicate statistically significant measurements.

Soft tissue in infants with a cleft lip can be assessed preoperatively with maxillary plaster models, cephalometry, and 3D computed tomography (CT). Fisher et al.<sup>20</sup> performed 3DCT in 3-month-old infants with complete cleft lip and discussed its association with soft tissue; they demonstrated that the severity of the cleft (alveolar cleft width) was associated with premaxillary protrusion (S-N-ANS). By comparison with soft tissue, patients with an alveolar cleft width less than 10 mm and those with an alveolar cleft width greater than 10 mm did not demonstrate significant differences in terms of en-en or al-al.<sup>20</sup> In addition, patients with wider clefts tended to demonstrate lateral deviation of the cleft side alar base.<sup>20</sup> However, because of problems such as radiation exposure and body movement, preoperative CT is now almost never performed in infants.<sup>21</sup> Therefore, preoperative and postoperative assessments are performed with maxillary plaster models that pose little risk to infants. Preoperative dislocation of alveolar segments corresponds to the asymmetry of the piriform aperture and is useful for grasping the overall image of maxillary hypoplasia.<sup>22</sup> Therefore, we assessed soft tissue with maxillary plaster models rather than exposing patients to radiation.

The columellar angle and nostril width ratio are correlated in preoperative assessments of soft tissue in cleft lip nasal deformities.<sup>23</sup> This finding was supported by the results of the present study; we found a strong correlation between the cleft side/non-cleft side nostril base width ratio (sn-sbal) and columellar angle. Mercan et al.<sup>24</sup> found that the columellar angle, nostril width ratio, and lateral lip height ratio correlated with cleft lip nasal deformity severity and affected esthetic outcomes. Regarding the association between columellar angle and alveolar cleft, wider alveolar clefts tend to be associated with more lateral deviation of the alar base (i.e., the nostril base width ratio increases).<sup>20</sup> Therefore, columellar angle deformity is suggested to be affected by alveolar cleft width.

In this study, it may not be sufficient to identify other anatomical relationships (maxilla, cartilages, and muscles). Moreover, 3DCT is superior to cephalometry and maxillary plaster models to understand complex maxillary hypoplasia in cleft lip patients and to assess the hard tissue.

Notably, the measurement of the alar base was affected by the cleft alveolus. This study is only a preliminary study to consider the prognosis of cleft lip. As with morphological studies of cleft lip, many measurement points exist, and important measurement points have been narrowed down<sup>23-25</sup> by establishing measurements peripheral to cleft lip nasal deformities and overall measurements demonstrated by classification of alveolar cleft deformities as reference values for uncorrected cases. Future studies on esthetic assessments are warranted, including comparison with preoperatively corrected cases and comparison based on alveolar growth and surgical techniques.

## Conclusion

Patients with overlapped alveolar segments demonstrated superior deviation of the non-cleft side lateral alar base height and the non-cleft side alar base height and advanced nasal deformity. Surgeons should consider the alveolar

segments during preoperative planning and postoperative assessment.

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No funding was received for this article.

## Conflict of interest

We have no conflicts of interest to declare.

## Supplementary material

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.bjps.2019.04.009](https://doi.org/10.1016/j.bjps.2019.04.009).

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