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Original Research

# Inclination of the Lip and Nose during Resting and Lip Protrusion in Children with Unilateral Cleft Lip

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**Abstract**

***Objective*:** To examine morphological changes of the lip and nose during resting and lip protrusion in patients with unilateral cleft lip.

***Methods*:** A total of 41 patients with unilateral cleft lip who underwent primary lip repair were included in the study population. The distortion of the lip and nose was measured and evaluated using photographs of the preoperative and postoperative resting lips and the postoperative protruding lips.

***Results*:** The position of the labial commissure on the affected side was cranially deviated before lip repair, and the deviation remained after surgery. The degree of cranial deviation was increased by lip protrusion. The position of the alar base on the affected side was caudally deviated before lip repair and improved after surgery; however, it was displaced cranially by lip protrusion. Horizontal displacement of the lip and nose was improved after surgery, and there was no change during lip protrusion.

***Conclusions*:** The characteristic facial distortion in patients with unilateral cleft lip were confirmed. Patients with cleft lip had distortions in the labial commissure and the nose. One of the important future challenges in cleft lip treatment is to focus on changes caused by muscle movement and provide natural and symmetrical facial expressions.

**Keywords**

facial distortion, inclination of the lip and nose, lip protrusion, cleft lip, orbicularis oris muscle

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## Introduction

Cleft lip repair requires not only the lengthening of the philtrum and shaping of Cupid's bow but also focuses on various issues of interest, such as scar line arrangements, the nasal structure, and anatomical subunits. Reconstruction of the orbicularis oris muscle (OOM) is one of the crucial factors in surgical treatment[1-5)]. The OOM has a unique form in that its muscle fibers cross the midline of the upper lip from both modioli. The decussating structure and surrounding facial expression muscles enable complex movements of the lip[5-9)]. In patients with cleft lip, the OOM is separated by the cleft and abnormally attached to the septum, nose, maxilla, and piriform rim. Reconstruction of the OOM is therefore necessary for cleft lip repair; however, the complex structure of the OOM is probably impossible to construct. In addition, despite the fact that various facial expression muscles are interlaced with the OOM, reports of surgical procedures that emphasize the simultaneous reconstruction of these muscle abnormalities are very few[10,11)]. Furthermore, even those reports have only shown static reconstruction results as surgical outcome. That is, none of the changes associated with muscle movement have been evaluated thus far, although the results of cleft lip repair have been evaluated in many ways.

In this study, we examined the morphological changes in facial expressions that are unique to patients with cleft lip.

## Methods

Approval was granted by the ethics committee of our institution, and informed consent was obtained. An anthropometric analysis was performed using standardized frontal

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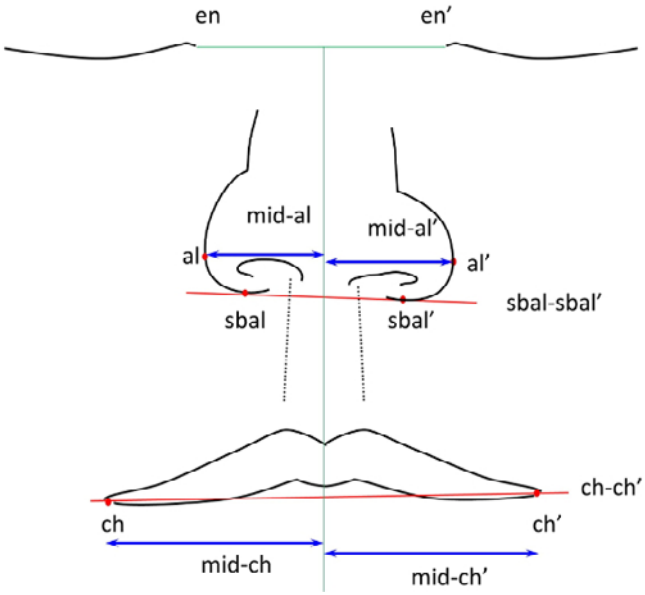


**Figure 1.** Anthropometric measurements. The horizontal distance from the midline to the labial commissures (mid-ch, mid-ch'), the horizontal distance from the midline to the alae (mid-al, mid-al'), the angle between the inter-cheilion line (ch-ch') and the horizontal axis (en-en), and the angle between the inter-subalare line (sbal-sbal') and the horizontal axis were measured.

photographs taken by digital single-lens reflex cameras obtained preoperatively and more than 3 years postoperatively. The preoperative photos were taken under general anesthesia at the time of primary cleft lip repair. The postoperative photos were taken at rest and with lip protrusion.

Based on the anthropometric values defined by Farkas, the endocanthion (en), subalare (sbal), alare (al), and cheilion (ch) were marked as soft tissue landmarks on the photos[11]. The reference horizontal axis (x) was positioned using the inter-endocanthion line (en-en'), and the vertical axis (y) was positioned through the midpoint of that line to define the facial midline. The following indices were calculated: [1] the lip width ratio, which is the ratio of the horizontal distance from the midline to the labial commissure on the affected side to the healthy side (mid-ch'/mid-ch); [2] the nasal width ratio, which is the ratio of the horizontal distance from the midline to the ala on the affected side to the healthy side (mid-al'/mid-al); [3] the inclination angle of the lip, which is the angle between the inter-cheilion line (ch-ch') and horizontal axis; [4] and the inclination angle of the nose, which is the angle between the inter-subalare line (sbal-sbal') and horizontal axis (**Figure 1**). For the inclination angles, the cranial deviation on the affected side was defined as the positive direction.

Measurements were performed using the ImageJ (v1.51g) software program. Measurements were taken three times on different days by two plastic surgeons, and the average of each measurement was applied.

### *Statistical analysis*

All statistical analyses were performed using JMP® Pro version 16.2. The 95% confidence intervals were calculated for the ratios and angles, where if the confidence interval for the ratio does not include 1, the difference in width between the affected and healthy sides can be considered significant; if the confidence interval for the angle does not include 0, the difference in height between the affected and healthy sides can be considered significant. Statistical differences between resting and lip protrusion were analyzed using the paired *t* test. P-values of <0.01 were considered statistically significant.

### *Study population*

This prospective study included 52 consecutive Japanese patients who underwent unilateral cleft lip repair performed by a single surgeon (H.M.) between 2011 and 2015. Photos of 41 patients with non-syndromic unilateral cleft lip were obtained, including 29 cases with complete cleft lip (with cleft palate [cCLAP], n = 20; and with alveolus cleft [cCLA], n = 9), and 12 with incomplete cleft lip [iCL] (with cleft palate, n = 3; with alveolus cleft, n = 7; without alveolus cleft, n = 2). All patients had not undergone revisional surgery, including rhinoplasty or alveolar bone graft surgery, by the time of photography. Palatoplasty was performed on all patients with cleft palate. Four cases in the cCLAP group and one case in the cCLA group had Simonart's band. The mean age at the time of the initial cheiloplasty was 3.68 months (range: 2.8-5.4 months), and the mean age when photos were taken was 60.8 months (range: 41-77 months) (**Table 1**). In five cases, a photograph of the lip protrusion expression could not be obtained. Another six patients were lost to follow-up within 3 years after the operation.

Cleft lip repair was performed with the Scarless Peak technique or its prototype, which is a modified triangular flap technique[5]. The muscle fibers were released from their attachments to the bone or nasal structure and divided into the pars marginalis and pars peripheralis. Both sides of these muscles were sutured.

## Results

The intra-class correlation coefficients (ICCs), ICC (1, 1) (intra-rater reliability) and ICC (3, 1) (inter-rater reliability), of the two evaluators are shown in **Table 2**. The correlation coefficients for all items were greater than 0.7, with the lowest being that for the preoperative inclination angle of the nose.

The calculated data are shown in **Table 3 and 4**. The relationship between the preoperative and postoperative values and the relationship between resting and lip protrusion are shown as scatter diagrams for the lip width ratio, nasal width ratio, inclination of the lip, and inclination of the nose (**Figure 2, 3, 4 and 5**).

The results are summarized as follows: [1] The lip width ratio was approximately 1. There were no significant morphological changes among the three conditions, although the individual ratios were slightly emphasized with lip protrusion (**Figure 2**). [2] The nasal width ratio, which was low before surgery, improved to approximately 1 after surgery.

**Table 1.** Patient Demographics.

| Sex | | No. | |
|---|---|---|---|
| Male | | 25 | |
| Female | | 16 | |
| Cleft type | | No. | left:right |
| Complete CLAP | | 20 (4)* | 16 (2)*:4 (2)* |
| Complete CLA | | 9 (1)* | 8 (1)*:1 |
| Incomplete CLAP | | 3 | 2:1 |
| Incomplete CLA | | 7 | 5:2 |
| Incomplete CL | | 2 | 2:0 |
| | total | 41 | 33:8 |
| Age at operation (months) | | 3.68 ± 0.56** (range: 2.8–5.4) | |
| Age at follow-up evaluation (months) | | 60.8 ± 8.48** (range: 41–77) | |

*with Simonart's Band

** mean ± standard deviation

**Table 2.** Intra-Rater and Inter-Rater Reliability.

| | | rater A ICC (1, 1) (95% CI) | rater B ICC (1, 1) (95% CI) | Interrater reliability ICC (3, 1) (95% CI) |
|---|---|---|---|---|
| lip width ratio | preoperative | 0.913 (0.860–0.949) | 0.958 (0.931–0.976) | 0.927 (0.868–0.961) |
| | postoperative | 0.968 (0.948–0.981) | 0.920 (0.870–0.953) | 0.955 (0.918–0.976) |
| | lip protrusion | 0.954 (0.924–0.973) | 0.955 (0.927–0.974) | 0.962 (0.930–0.980) |
| nasal width ratio | preoperative | 0.978 (0.964–0.988) | 0.971 (0.952–0.983) | 0.967 (0.939–0.982) |
| | postoperative | 0.921 (0.873–0.954) | 0.978 (0.964–0.988) | 0.941 (0.893–0.968) |
| | lip protrusion | 0.894 (0.831–0.938) | 0.981 (0.968–0.989) | 0.933 (0.878–0.964) |
| Inclination angle of the lip | preoperative | 0.938 (0.900–0.964) | 0.933 (0.891–0.961) | 0.943 (0.895–0.969) |
| | postoperative | 0.944 (0.908–0.968) | 0.956 (0.928–0.975) | 0.958 (0.923–0.978) |
| | lip protrusion | 0.939 (0.900–0.965) | 0.861 (0.782–0.918) | 0.936 (0.826–0.965) |
| Inclination angle of the nose | preoperative | 0.724 (0.589–0.830) | 0.807 (0.703–0.884) | 0.848 (0.733–0.916) |
| | postoperative | 0.859 (0.779–0.917) | 0.874 (0.803–0.926) | 0.776 (0.617–0.874) |
| | lip protrusion | 0.924 (0.877–0.956) | 0.900 (0.840–0.942) | 0.911 (0.839–0.951) |

**Table 3.** Calculated Data.

| | | mean ± SD | 95%CI |
|---|---|---|---|
| Lip width ratio | preoperative | 1.0234 ± 0.1821 | 0.9660–1.0809 |
| | postoperative | 1.0040 ± 0.1249 | 0.9645–1.0434 |
| | lip protrusion | 0.9948 ± 0.2127 | 0.9276–1.0619 |
| Nasal width ratio | preoperative | 0.8380 ± 0.0959 | **0.8078–0.8683** |
| | postoperative | 0.9884 ± 0.0923 | 0.9592–1.0175 |
| | lip protrusion | 0.9907 ± 0.0973 | 0.9600–1.0214 |
| Inclination angle of the lip | preoperative | 2.2867 ± 1.7262 | **1.7418–2.8315** |
| | postoperative | 1.6747 ± 2.0009 | **1.0431–2.3062** |
| | lip protrusion | 2.9496 ± 2.1427 | **2.2733–3.6259** |
| Inclination angle of the nose | preoperative | –2.9499 ± 1.5575 | **–3.4415––2.4583** |
| | postoperative | –0.4018 ± 1.5794 | –0.9003–0.0967 |
| | lip protrusion | 1.4887 ± 2.4830 | **0.7049–2.2724** |

Highlighted in bold are variables considered to be significantly different between the affected and healthy sides.

No change was observed during lip protrusion (**Figure 3**). [3] The inclination angle of the lip was positive before surgery and did not change after surgery. In addition, the positive angles increased with lip protrusion (**Figure 4**, **Table 4**). [4] The inclination angle of the nose was negative before surgery and improved after surgery; however, the angle increased in the positive direction on lip protrusion (**Figure 5**, **Table 4**). A representative case is shown in **Figure 6**.

**Table 4.** Comparison between Resting and Lip Protrusion.

| | Resting | Lip Protrusion | *p* |
|---|---|---|---|
| Lip width ratio | 1.0040 ± 0.1249 | 0.9948 ± 0.2127 | 0.693 |
| Nasal width ratio | 0.9884 ± 0.0923 | 0.9907 ± 0.0973 | 0.826 |
| Inclination angle of the lip | 1.6747 ± 2.0009 | 2.9496 ± 2.1427 | <0.001 |
| Inclination angle of the nose | −0.4018 ± 1.5794 | 1.4887 ± 2.4830 | <0.001 |

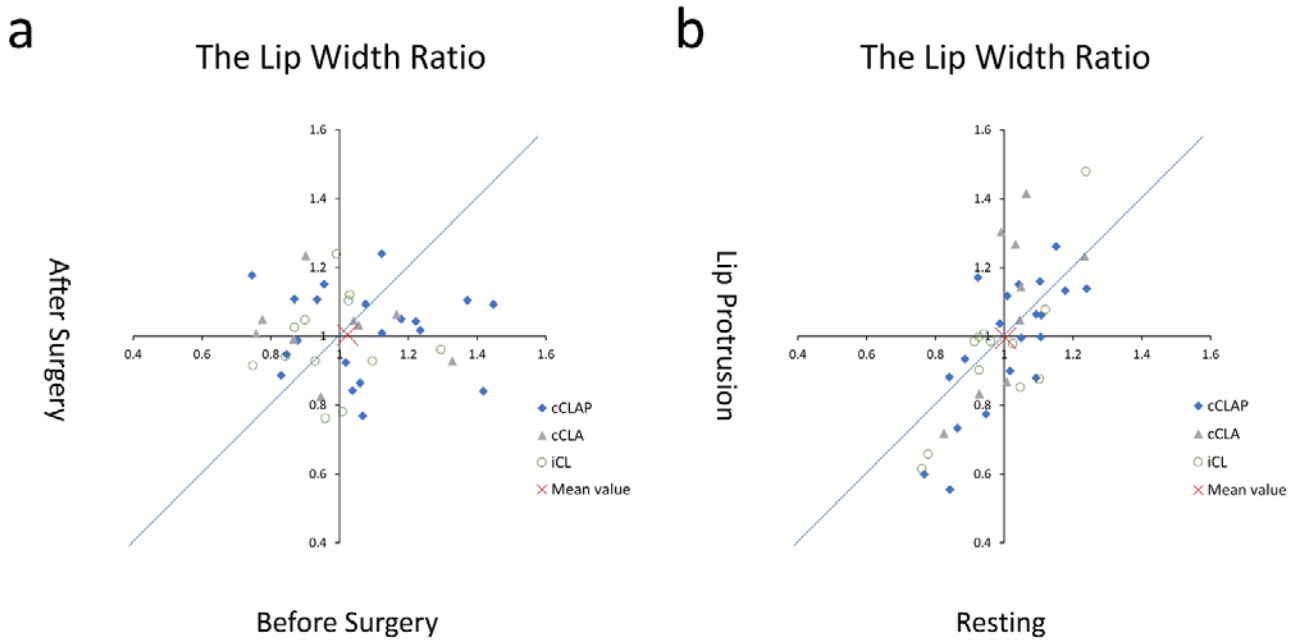


**Figure 2.** Scatter diagrams for the lip width ratio. The relationship between the preoperative and postoperative values (a) and the relationship between resting and lip protrusion (b) are shown. The blue diagonal auxiliary line in each graph represents y = x.

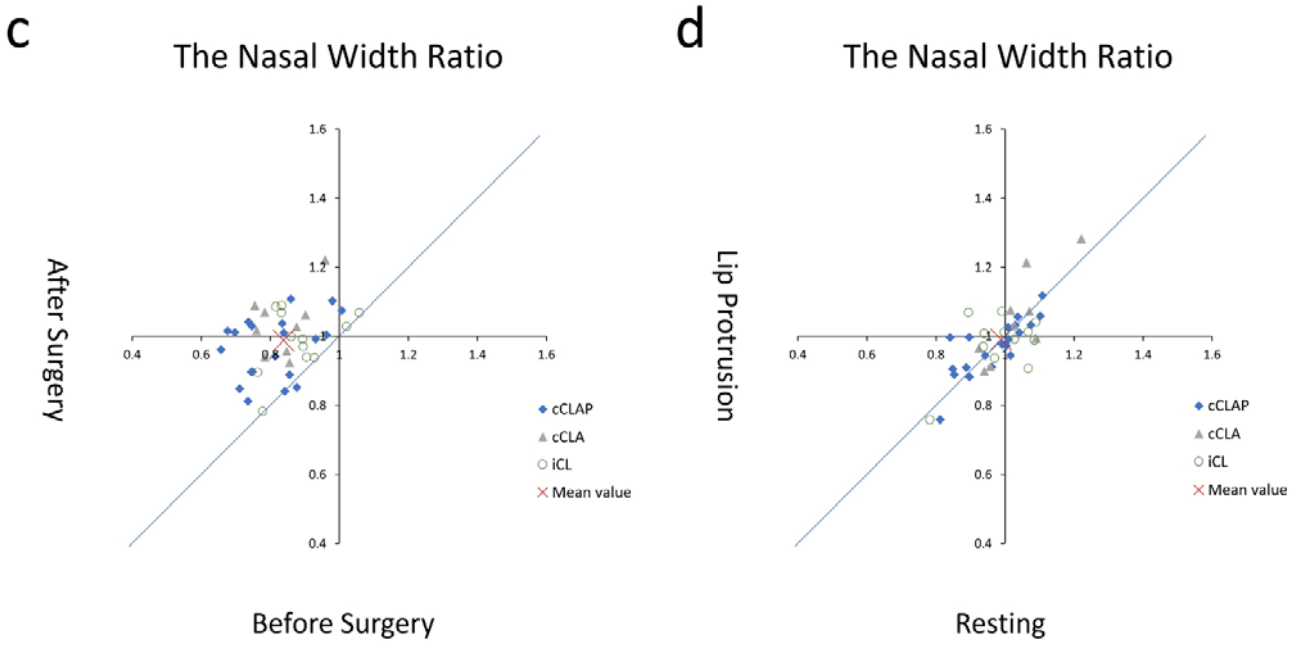


**Figure 3.** Scatter diagrams for the nasal width ratio. The relationship between the preoperative and postoperative values (c) and the relationship between resting and lip protrusion (d) are shown. The blue diagonal auxiliary line in each graph represents y = x.

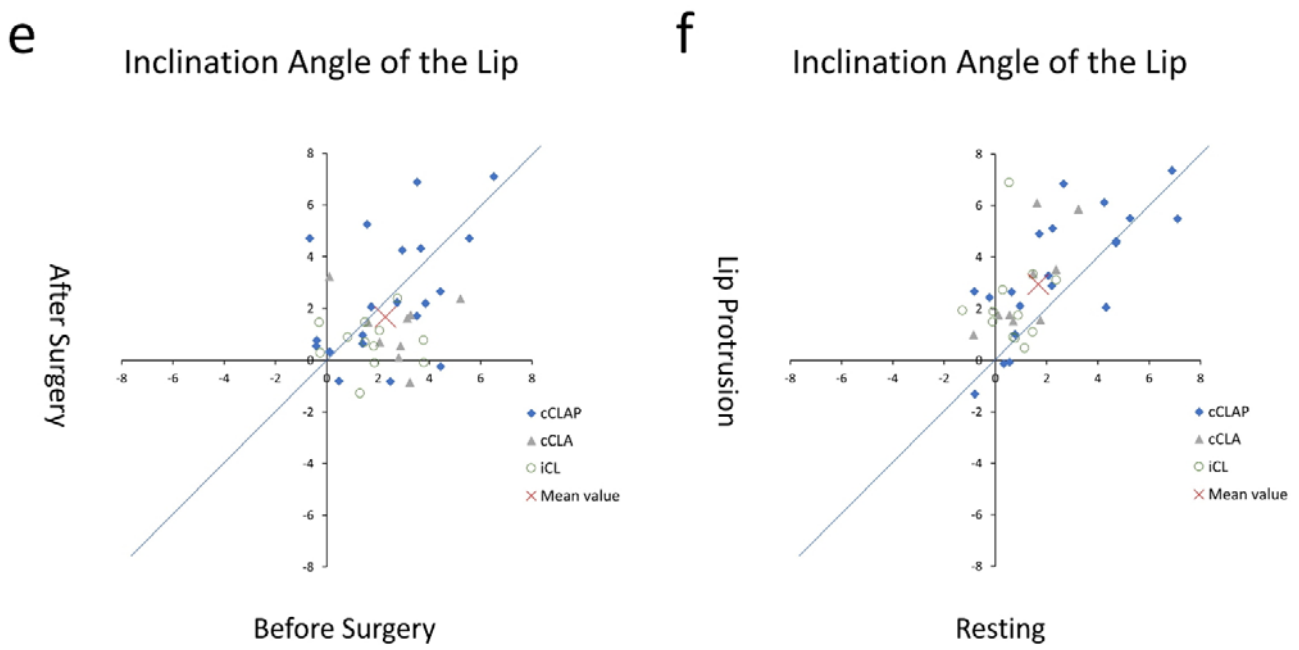


**Figure 4.** Scatter diagrams for the inclination of the lip. The relationship between the preoperative and postoperative values (e) and the relationship between resting and lip protrusion (f) are shown. The blue diagonal auxiliary line in each graph represents y = x.

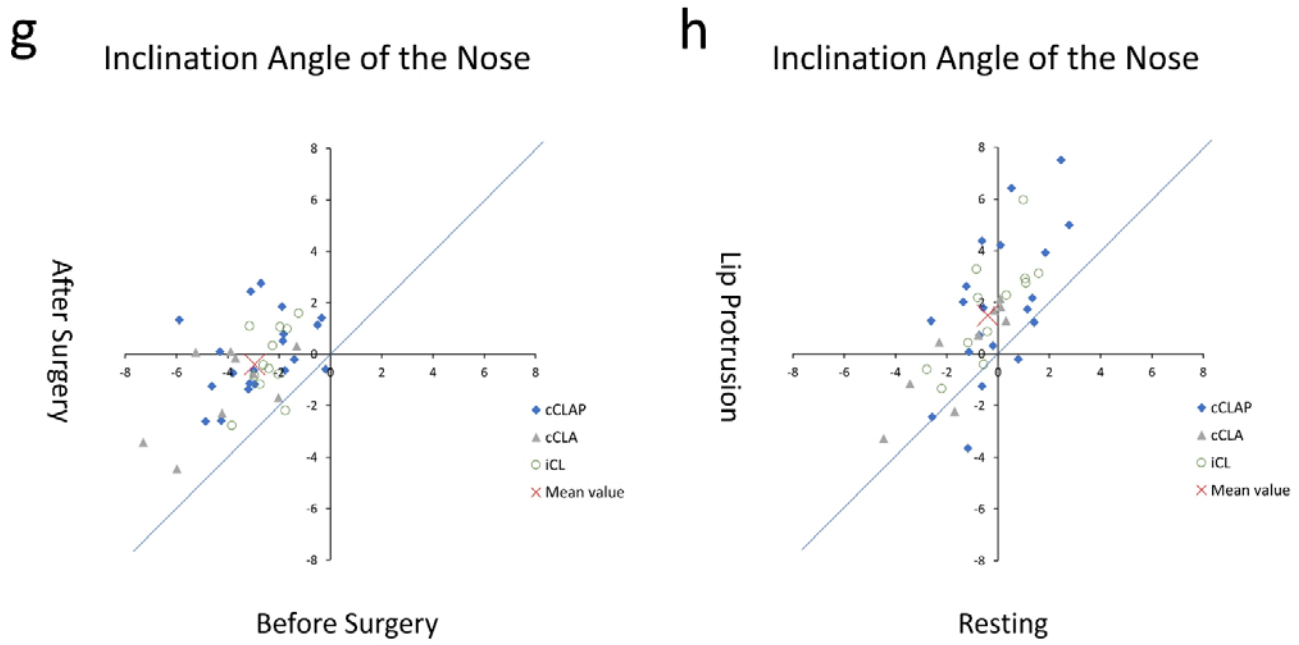


**Figure 5.** Scatter diagrams for the inclination of the nose. The relationship between the preoperative and postoperative values (g) and the relationship between resting and lip protrusion (h) are shown. The blue diagonal auxiliary line in each graph represents y = x.

## Discussion

We investigated the distortion of the lip and nose in patients with unilateral cleft lip. The results showed that the labial commissure on the affected side was cranially deviated before lip repair and did not improve even after surgery. In addition, lip protrusion aggravated the inclination of the lip. Furthermore, we found that the alar base on the affected side, which drooped before surgery, was improved after surgery; nevertheless, lip protrusion cranially deviated the alar base on the affected side. On the other hand, no horizontal displacement of the lips or nose was observed, with the exception that the nose was displaced to the healthy side before surgery.

The morphological evaluation of patients with cleft lip is usually performed using the standard anthropometric landmarks defined by Farkas[12]. The surgical results of lip repair are evaluated based on various items, including the length of

**Figure 6.** Representative case. Photographs of a patient with cCLA obtained preoperatively (*left*), 4 years postoperatively at rest (*center*), and 4 years postoperatively on lip protrusion (*right*). The inclination angles of the lip and nose were 1.60, 1.47, 3.37, and −5.25, 0.07, 2.15, respectively.

the philtrum, the shape of Cupid's bow, the inclination of the nasal column, and the difference in nostril width. Direct measurement is considered reliable for assessing the results of cleft lip repair[13-15]; however, the situations in which it can be measured are limited. Furthermore, differences in measured values do not necessarily reflect the degree of facial distortion because the angles of each measured item may be asymmetric. Photographs are frequently used to assess facial symmetry because they are relatively easy to obtain, which is an advantage. It is possible to measure the ratio or inclination from standardized photographs; however, the items that can be measured are limited, and it is not possible to obtain an actual measurement value. In order to measure the inclination or deviation, a line through the left and right medial canthi, lateral canthi, or centers of the pupil is used as a horizontal reference line that is not affected by medical conditions, such as cleft lip[16-19]. Measurements using three-dimensional images, which have been reported in recent studies, allow for a more accurate analysis[20-25]; however, these instruments have not yet been widely distributed to general facilities because they require expensive camera equipment and software programs to perform image analysis. Unfortunately, since numerous items can be measured and measurements are complicated, many studies that have used three-dimensional imaging have only described the numerical evaluation of the same measurement items as direct measurement, without setting a reference point or a reference line. Actually, only a few studies have evaluated facial components three-dimensionally[25,26]. Prior to this study, we were not able to find reports that referred to the height differences in the labial commissure or the inclination of the lip using direct measurement, standard photographs, or three-dimensional photographs. In other words, the previous reports are highly likely to have evaluated the lip and nose morphology in patients with cleft lip based on the premise that the labial commissures are symmetrical.

The difference in the height of the peak of Cupid's bow between the affected and healthy sides is generally discussed as an indicator of the outcome of cleft lip repair. Similarly, the difference in the lateral lip width between the affected and healthy sides is often pointed out[13-17,21,24]. These differences are due not only to the congenital difference in length, but also to the design; that is, the point of the Cupid's bow peak, as determined by the operator. Moreover, the methods for dividing or suturing the muscle may affect the symmetry of the lip. However, whether these factors influence the height of the labial commissures has not been reported. The results of this study showed that the alar base on the affected side, which had drooped congenitally, was corrected to the same level as that on the healthy side after the operation. As the symmetry of the nasal alae can be regarded as one of the indicators of whether surgical correction was performed properly, we believe that the surgical procedure we performed was one of the appropriate methods—at least in terms of the static evaluation.

In patients with cleft lip, the labial commissure on the affected side tends to show cranial displacement congenitally. Moreover, we found that the displacement remained after surgery despite the transfer of the OOM by releasing the muscle attachment, such as the piriform rim or nasal structure. We also found that the displacement in patients with cCLAP was greater than that in patients with iCL. It is obvious that the inclination of the lip affects the visual impression of the height of the peak of Cupid's bow. In other words, evaluations of the operative results for cleft lip differ according to whether the line connecting the labial commissures or the line connecting the medial canthi is used as the reference axis. Although it is unclear whether different surgical procedures have different consequences for the inclination of the lip, there are no reports of a surgical procedure for cleft lip repair that aims to correct the inclination of the lip. It would be preferable to add the inclination angle of the lip as a postoperative evaluation item.

In the dynamic evaluation, we found that both the labial commissure and the alar base of the affected side were elevated by lip protrusion. Especially in the cases of iCL, the inclination of the lip and nose, which was almost absent at rest, occurred on lip protrusion (**Figure 7**). The degree of

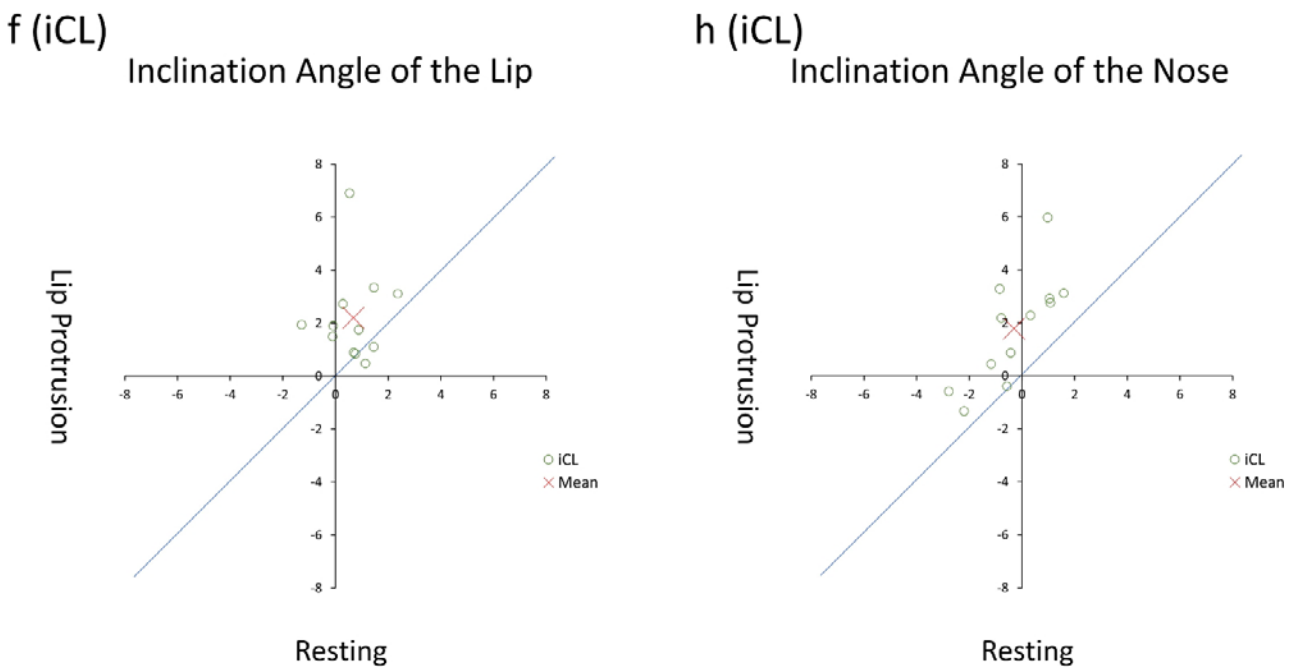


**Figure 7.** Scatter diagrams for the inclination of the lip and nose in patients with incomplete cleft lip. The relationship between resting and lip protrusion are shown. The blue diagonal auxiliary line in each graph represents y = x.

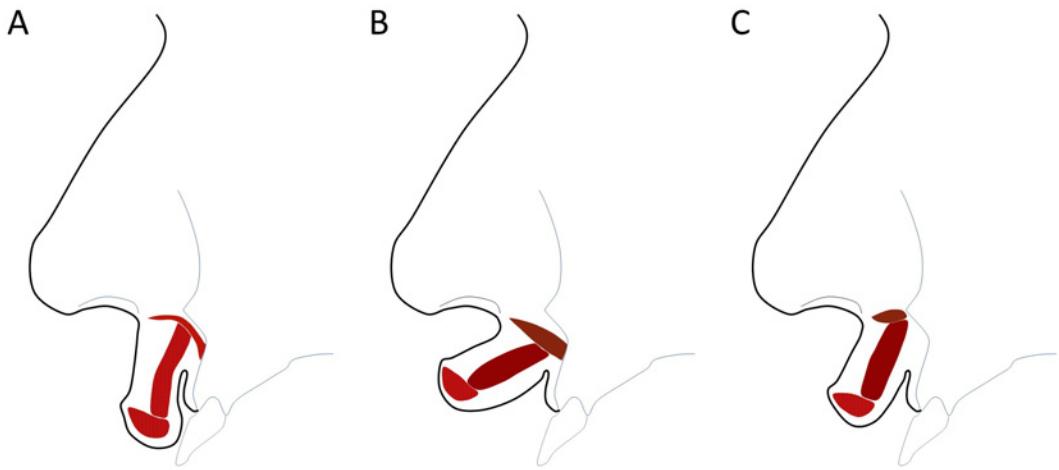


**Figure 8.** Schematic illustration of the hypothetical mechanism of the lip protrusion. (A): Resting position of the OOM and DSN. (B): Lip protrusion with the OOM and DSN function. (C): Lip protrusion without effective DSN function.

change in inclination was similar to that of complete cleft. Scar rigidity may have affected the inclination of the lip; however, simultaneous cranial elevation of the nasal ala by lip protrusion cannot be explained as an effect of the scar. In other words, this motion reflects muscle dysfunction and is not the result of static morphological reconstruction. We therefore assume that some of the facial muscles, including the OOM, have insufficient movability or abnormal movement directions in patients with cleft lip.

The movement of the lips is complicated, and various facial muscles around the OOM interact to enable facial expression. In our previous report, we found that the peripheral portion of the OOM and its crossing fibers pull the lateral lip inward and increase the thickness of the philtrum on lip protrusion[8]. From this point of view, we inferred that the functions of the peripheral and marginal portions were lip protrusion and lip closure, respectively. Therefore, at the time of surgery, we divided the OOM into the peripheral and marginal portions and sutured each portion separately. We also found that the depressor septi nasi muscle (DSN) anchors the cranial rim of the OOM to the maxilla in addition to pulling down the nostril floor[9]. If the OOM contracts without the DSN function, the cranial rim of the OOM slides up over the anterior surface of the maxilla (**Figure 8**). Thus, the unreconstructed attachment of the affected DSN to the maxilla is responsible for the labial commissure and alar base of the affected side moving to the cranial side in patients with cleft lip. It is expected that other facilities will

report on this aspect in the future, as other methods of muscle layer suturing or surgical design may give different results.

Taken together, we found that some of the facial distortion that exists from birth not only remains after cleft lip repair but is also emphasized when making facial expressions. The cranial displacement of the labial commissure and nasal ala by lip protrusion was consistent with our previous study's theory of the DSN function that anchors the cranial end of the OOM to the maxilla, considering the lack of DSN attachment to the maxilla in patients with cleft lip[9]. Although we could not evaluate the degree of lip protrusion because measurements were performed using standard two-dimensional photographs, we consider that the absence or dysfunction of the DSN probably causes insufficient warping and protrusion when the lips are protruded.

## Conclusion

We confirmed the characteristic facial distortion in patients with cleft lip. Patients with cleft lip had distortions in the labial commissure as well as the nose. These distortions were increased by muscle movement in the direction of the labial commissure and the alar base of the affected side toward the cranial side. We therefore consider that one of the important future challenges in cleft lip treatment is to focus on the changes caused by muscle movement and to provide natural and symmetrical facial expressions.

**Author Contributions:** H.M. designed the study; H.M. and M.F. performed the experiments and analyzed the data; N.K. and K.K. supervised the examination; H.M. wrote the manuscript.

**Conflicts of Interest:** There are no conflicts of interest.

**Ethical Approval:** This study was reviewed and approved by the ethics committee of Kansai Medical University (approval no.: 2022224).

**Consent to Participate:** The patients provided their written informed consent to participate in this study.

**Consent for Publication:** The patients provided their written informed consent for the publication of this study.

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