

Does Initial Cleft Lip Width Predict Final Aesthetic Outcome?

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Background: A natural aesthetic appearance of the lip and a favorable scar are essential goals of cleft lip repair. Wider clefts intuitively pose a greater technical challenge; however, the relationship between initial width and aesthetic outcome remains controversial. The current study aimed to determine whether lip adhesion can help wider clefts achieve safe, consistent aesthetic outcomes.

Methods: A retrospective cohort study was conducted on unilateral cleft lip patients who underwent lip repair within a 2-year period by the senior author. Subjects were divided into three groups based on cleft severity: (1) wide complete clefts that required lip adhesion before definitive repair, (2) narrower complete clefts that did not require lip adhesion, and (3) incomplete clefts. Aesthetic outcomes related to the vermillion and upper lip scar were rated by 48 blinded observers. Statistical analysis was performed using Kruskal-Wallis and Mann-Whitney tests. Nasal outcomes were not assessed.

Results: Seventeen patients were included in the study: five in group 1, six in group 2, and six in group 3. Patients with the widest clefts did not have inferior results compared with the other groups. In fact, they had statistically significantly higher aesthetic scores in all scar-related outcomes compared with those in groups 2 and 3 (P<0.0001). **Conclusions:** This study suggests that a wide cleft does not necessarily foreshadow a poor surgical outcome. In fact, wide clefts may have pleasing results, and the use of a staged lip adhesion approach can be useful for achieving the desired outcome in wider clefts. (*Plast Reconstr Surg Glob Open 2021;9:e3966; doi: 10.1097/GOX.00000000003966; Published online 6 December 2021.*)

INTRODUCTION

Cleft lip with or without cleft palate is the most common congenital craniofacial abnormality, affecting approximately one in 1000 newborns.¹ Inherent in the cleft lip defect, the adjacent nasolabial structures exhibit variable amounts of displacement and distortion. These include a protruding and outwardly rotated premaxilla, hypoplastic orbicularis oris, and shortened philtrum with adhesion between the lip and gingiva.² These factors contribute to the severity of the cleft, and must be taken into account when considering treatment.

The most salient goal in the treatment of cleft lip is achieving a natural, aesthetic nasolabial appearance with minimal scar burden. It seems intuitive that wider clefts pose a greater technical challenge, consequently leading to poorer results. This belief, however, remains

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Received for publication January 21, 2021; accepted July 28, 2021. Copyright © 2021 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.00000000003966 controversial, with only a few studies available in the literature with conflicting findings. $^{\rm 3-5}$

Lip adhesion is a technique designed to convert a wide cleft into an incomplete cleft to facilitate definitive closure. Indeed, Vander Woude and Mulliken have found improved labial height after infants undergo lip adhesion compared with before the procedure;⁶ however, there is still a lack of evidence regarding the final aesthetic endpoint after lip adhesion and definitive repair. These long-term aesthetic data are necessary for counseling parents on postoperative expectations, and for offering the most beneficial procedures to patients while minimizing surgical and anesthetic risk.

The present study aims to determine whether lip adhesion can help achieve a comparable aesthetic endpoint in patients with severe clefts compared with those with narrower or incomplete clefts. We hypothesize that pleasing results can be achieved regardless of initial severity, and lip adhesion can be safely used in selected cases to help achieve consistent pleasing results.

METHODS

Study Design

This was a retrospective cohort study (IRB #1101007932) of infants who underwent primary cleft

Disclosure: The authors have no financial interest in relation to the content of this article. lip repair by the senior author (DS) between November 2015 and November 2017 at Yale-New Haven Children's Hospital. All subjects underwent repair using the modified inferior triangle technique^{7,8} (Fig. 1) and lip adhesion when indicated by severity of the cleft. Patients also underwent botox injection and fat grafting intraoperatively.

Inclusion criteria included (1) nonsyndromic, unilateral cleft lip; (2) no other lip defects or surgeries; (3) no presurgical orthodontic intervention (ie, naso-alveolar molding); (4) definitive repair before 1 year of age; and (5) reliable and reproducible initial width measurements based on nasolabial molds taken at the time of surgery (Fig. 2).

Predictor Variables

Subjects were divided into three groups based on initial cleft width (Table 1). The initial cleft widths were measured using nasolabial molds that were taken at the time of surgery, when the patients were under general anesthesia. The width of the defect at the level of the white roll was recorded in millimeters. Patients in the widest cleft group underwent lip adhesion before definitive repair, and their cleft widths were measured at the time of lip adhesion. Demographic information and notable peri- and postoperative events were recorded.

Outcomes Variables

Postoperative photographs taken at an average of 17 months after definitive repair were analyzed for seven aesthetic outcomes variables: (1) overall scar appearance, (2) scar pigment, (3) scar width, (4) scar contour, (5) lip symmetry, (6) lip fullness, and (7) lip contour. Photographs were de-identified, blinded, cropped, and enlarged to show the lip and philtral region and limit the impact of other facial features on aesthetic perception.

Each postoperative photograph was evaluated by 48 blinded evaluators, 17 of which had plastic surgery (PRS) training, and 31 laypeople. Evaluators ranked each outcome



Fig. 1. Surgical plan using the modified inferior triangle technique marked in methylene blue.



Fig. 2. Representative photograph of a lip impression and 3D plaster model.

variable on a seven-point Likert scale (7 being most aesthetically pleasing). Photographs and rating forms were presented electronically via secure online survey administered by Yale University's licensed Qualtrics software.

Data Analysis

At the end of the study period, data were exported from Qualtrics to Excel (Microsoft Corp, Redmond, Wash.) for tabulation and statistical analysis. We carried out statistical analyses separately for PRS-trained evaluators and layperson evaluators.

Data were analyzed as nonparametric ordinal data. As such, Kruskal-Wallis tests were used to determine whether the three groups were statistically different in each of the seven outcomes variables. Where indicated, post-hoc Mann-Whitney tests were used for pairwise comparisons.

RESULTS

Subject Characteristics

Seventeen patients were included in the study: five in group 1, six in group 2, and six in group 3. Demographics are summarized in Table 1. Patients in group 1 had the widest clefts despite being the youngest at the time of measurement.

Aesthetic Outcomes

Figure 3 shows representative pre- and postoperative photographs.

Table 1. Patient Characteristics

	Wide Cleft	Narrow Cleft	Incomplete Cleft
No. subjects (men) African American White Uiseanic	5(3) 1 3		$\begin{array}{c} 6 (3) \\ 2 \\ 4 \\ 0 \end{array}$
Average width at white roll (mm)	15.9 ± 2.5	11.3 ± 4.2	0 N/A
Average age at the time of measurement (mo)	1.7 ± 0.1	4.4 ± 2.4	4.9 ± 1.9



Fig. 3. Representative pre- and postoperative photographs of patients from the wide cleft group (A, B), and the incomplete cleft group (C, D).

The three groups were statistically different in terms of overall scar appearance, pigment, width, and shape (Table 2). There was no significant difference in lip symmetry, lip fullness, or lip contour. The mean aesthetic scores and comparisons across the patient groups are summarized in Table 3.

Patients with the widest initial clefts did not have inferior scores compared with the narrow and incomplete cleft groups. In fact, they had significantly higher scores in all scar-related outcomes (P < 0.0001). There was no significant difference between narrow and incomplete groups. Findings were similar among PRS-trained and layperson evaluators.

None of the patients in our cohort experienced immediate or delayed operative complications such as anesthetic complication, infection, or wound dehiscence.

DISCUSSION

The results of this study confirm our hypothesis that a wide cleft does not necessarily foreshadow a poor surgical outcome, and that a pleasing outcome can be achieved in patients with wide initial clefts with use of lip adhesion as an adjunct to our standard modified inferior triangle technique. In fact, the patients in our study with the widest initial clefts received the best aesthetic ratings, and no significant difference was found between the narrow and the incomplete cleft groups. Our results suggest that there is no inherent correlation between width of cleft and final aesthetic potential when staged repair is used as indicated.

There is a surprisingly small body of literature pertaining to aesthetic outcomes following cleft lip repair,^{3–5,9–11} with an exceedingly small portion discussing the influence

Table 2. Kruskal-Wallis Test Results for Outcomes Variables

	Overall Scar	Scar Pigment	Scar Width	Scar Shape	Lip Contour	Lip Fullness	Lip Symmetry
PRS-trained	P < 0.0001	P < 0.0001	P < 0.0001	P < 0.0001	P = 0.51	P = 0.58	P = 0.62
Layperson	P < 0.0001	P < 0.0001	P < 0.0001	P < 0.0001	P = 0.09	P = 0.65	P = 0.075

	Overall Scar Appearance			Scar Pigment		Scar Width			Scar Contour			
	Mean Score	Comparison Group	Р	Mean Score	Comparison Group	Р	Mean Score	Comparison Group	Р	Mean Score	Comparison Group	Р
Wide	5.54 ± 0.10	Narrow Incomplete	<0.001	5.48 ± 0.10	Narrow Incomplete	<0.001	5.33 ± 0.11	Narrow Incomplete	<0.001	5.16 ± 0.11	Narrow Incomplete	<0.001
Narrow	4.29 ± 0.12	Wide Incomplete	<0.001 <0.001 0.60	4.50 ± 0.12	Wide Incomplete	<0.001 <0.001 0.47	4.22 ± 0.12	Wide Incomplete	<0.001 <0.001 0.73	4.10 ± 0.12	Wide	<0.001 <0.001 0.74
Incomplete	4.20 ± 0.11	Wide Narrow	<0.001 0.60	4.38 ± 0.12	Wide Narrow	<0.001 0.47	4.16 ± 0.12	Wide Narrow	<0.001 0.73	4.15 ± 0.11	Wide Narrow	<0.001 0.74

Table 3. Mean Aesthetic Scores of Each Cleft Lip Severity Group and Comparison of Means across the Groups

of initial cleft width.^{3–5} These existing studies reported conflicting results, and many were limited by a small sample size, use of different repair techniques, different operating surgeons, and inconsistent use of outcomes rating scales. A recently published retrospective study on this topic containing the largest sample size to date consisted of multi-national unilateral cleft lip patients of all ages who underwent primary repair through Operation Smile.⁵ Three layperson evaluators rated each subject's pre-op and immediate postoperative photographs, and demonstrated that increased initial cleft severity was correlated with poor aesthetic result.

Our study design was fundamentally different and aimed at evaluating this question in a different healthcare setting. Due to inherent limitations of mission trips, all patients in the above-mentioned study received singlestage repairs by a variety of techniques, and only immediate "on table" results were available for evaluation. In contrast, the patients in our study all underwent repair using a consistent technique, with the widest initial clefts undergoing an additional lip adhesion. Our patients were also more homogenous in age at the time of surgery. In our opinion, this scenario is more representative of most permanent cleft surgery practices in developed countries, and our findings offer more relevant insights for this setting.

An unexpected but interesting finding is that subjects with the widest clefts requiring lip adhesion received the best scar aesthetic ratings. However, the magnitude of difference was only about 1 to 1.5 points on a seven-point Likert scale, which likely does not result in a large clinical difference. Nevertheless, this result suggests that the outcomes in these severe cleft cases are not inferior to those achieved in less severe cases.

Lip adhesion can be a controversial procedure. Compared with common pre-surgical orthopedic appliances (ie, NAM and lip taping), lip adhesion has the advantage of requiring low burden of care by the family, and no need for frequent orthodontic visits.^{12,13} Lip adhesion has been shown to effectively improve the nasal contour, approximate and align the alveolar segments, decrease the tension of definitive repair, thicken the orbicularis oris muscle, and lengthen the lateral and medial lip elements.^{6,14} Nevertheless, there are notable risks to lip adhesion, including additional general anesthesia, infection, and discarding/scarring tissue that could be otherwise used during definitive repair.¹⁶ In addition, reported rates of dehiscence can be as high as 24% in bilateral cases and 8% in unilateral cases. 17,18

Fortunately, no patients in our study experienced any of these complications; however, the complication risk profile is not inconsequential, and should be carefully weighed against the potential benefit on a case-bycase basis. We believe that the procedure should only be offered to patients with very wide clefts to limit the amount of soft tissue undermining, as undermining may potentially negatively impact maxillary growth.¹³

A final finding in our study is that the ratings were incredibly similar among PRS-trained and laypeople. This agrees with prior studies on the subject, and re-emphasizes that even a repaired cleft lip is easy to spot by others regardless of their experience level. This finding potentially speaks to the psychosocial impact that cleft patients experience. A contributing factor to this finding may be that the photographs were cropped and zoomed in to show a clear resolution of the lip and scar. This was done to aid the detection of even minute details that could better parse out aesthetic differences. However, perhaps if the entire face was shown from a normal distance, we may elucidate differences in the two groups' abilities to pick up on smaller nuances.

Our study had several limitations. First, this was a single-institution, single-surgeon investigation. Although this strengthens the internal validity, it limits the external validity. Second, we had a small sample size who retrospectively met the inclusion criteria of having plaster molds available for measurement. We attempted to offset this by recruiting a large number of evaluators. We ended up with 48 evaluators for each subject, which is much higher than prior studies on this topic. Third, we did not evaluate postoperative nasal aesthetics. The cleft nose plays an important role in facial balance, and a secondary rhinoplasty is often indicated in adolescence for further refinement. Future studies focusing on nasal outcomes in relation to initial nasal deformity would be interesting. Lastly, the indication for lip adhesion was subjectively determined by the senior surgeon based on clinical experience. The retroactively measured cleft "width" reflects horizontal displacement as well as protrusion of the premaxillary segment. Future studies should quantify and refine the objective indications for lip adhesion. Furthermore, subsequent studies should evaluate aesthetic results several years out from repair to determine the effects of growth.

CONCLUSIONS

A wide cleft does not necessarily predict poor aesthetic outcome. Using lip adhesion as an adjunct to our modified inferior triangle technique, pleasing results can be achieved even in the most severe clefts.

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PATIENT CONSENT

Parents or guardians provided written consent for the use of the patients' images.

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