

The Influence of Four Different Treatment Protocols on Maxillofacial Growth in Patients with Unilateral Complete Cleft Lip, Palate, and Alveolus

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Background: The purpose of this study was to evaluate the effect of four different treatment protocols on maxillofacial growth in patients aged 7 to 8 years with unilateral complete cleft lip, palate, and alveolus.

Methods: Sixty-one patients with nonsyndromic unilateral complete cleft lip, palate, and alveolus were entered into this study and grouped as follows: group 1 patients had a repaired lip and an unrepaired palate; group 2 patients underwent one-stage palatoplasty; group 3 patients underwent two-stage palatoplasty; and group 4 patients underwent lip adhesion and two-stage palatoplasty. The control group was composed of 16 patients with unilateral incomplete cleft lip. The Kolmogorov-Smirnov test was used to test the nature of data distribution. The Bonferroni test and the Kruskal-Wallis H test were used for multiple comparisons.

Results: Group 5 showed a more protruding maxilla (basion-nasion-A point, basion-nasion-anterior nasal spine, sella-nasion-anterior nasal spine; $p < 0.05$), longer maxillary sagittal length (anterior nasal spine-posterior maxillary point; $p < 0.05$) and maxillary basal sagittal length (A point-posterior maxillary point; $p < 0.05$), and a better jaw relationship (A point-nasion-B point angle; $p < 0.05$) than groups 2, 3, and 4. Group 2 had higher anterior facial height (anterior nasal spine-nasion, anterior nasal spine-menton, nasion-menton; $p < 0.05$) and posterior facial height (registration point-posterior maxillary point; $p < 0.05$) than groups 3 and 4. Groups 2 and 3 had better maxillary position (sella-pterygomaxillary fissure; $p < 0.05$) and deeper bony pharynx (basion-posterior maxillary point; $p < 0.05$) than group 4.

Conclusions: In patients aged 7 to 8 years with unilateral complete cleft lip, palate, and alveolus, both one- and two-stage palatoplasty inhibited maxillary sagittal growth. Vomer flap repair with denuded bone inhibited maxillary vertical growth. Lip adhesion did adversely affect maxilla position. (*Plast. Reconstr. Surg.* 144: 180, 2019.)

CLINICAL QUESTION/LEVEL OF EVIDENCE: Therapeutic, III.

There is still great debate over the best treatment protocol for patients with clefts. Identifying a treatment protocol that leads to the fewest adverse effects on maxillary growth is a vital aim of surgeons. Maxillary growth depends on treatment protocol.¹

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Lip adhesion was first proposed by Simon in the nineteenth century to mold the premaxilla, and was used by Johanson and Ohlsson to repair primary alveolar bone grafts in the twentieth century.²⁻⁴ This method was modified and popularized by Millard and others⁴⁻⁶ to decrease the gap in alveolar segments. Then, it was used in bilateral clefts by Spina⁷ and Millard and Latham,⁸ and was used in all patients with complete clefts to facilitate closure by Randall.⁶ Lip adhesion can narrow

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the alveolar gap, increase orbicularis oris tissue, and increase the vertical height of the lateral and medial lip elements. Lip adhesion can convert a complete cleft lip into an incomplete cleft lip^{9,10} and make the definitive closure simpler and easier. Extensive soft-tissue undermining has deleterious effects on maxillary growth.^{11,12} Although there are benefits of the above, some scholars expressed that it might cause inestimable damage to the growing maxilla. As an additional operation, lip adhesion can create scar tissue, which increases the difficulty of secondary interventions and extra expense.^{6,13} It is more frequently used in patients with a complete unilateral cleft lip than for those with bilateral clefts. Of the surgeons in America and Canada, 11 percent used lip adhesion in complete bilateral cleft lip and palate,¹⁴ and 39 percent of the surgeons used it in 25 percent or more with complete unilateral cleft lip and palate.^{14,15}

Treatment protocols that include lip adhesion for primary repair of unilateral and bilateral complete cleft lip seem to depend primarily on expert opinion and experience, with only a few comparative studies published.^{16–19} Previous studies involving lip adhesion focused mostly on maxillary arch morphology and dimension, but seldom reported on its effect on maxillofacial morphology.^{18,19} We discussed the influence of two different palate repair protocols on maxillofacial growth in patients with unilateral complete cleft lip, palate, and alveolus.²⁰ Now, we aim to evaluate the effect of four different treatment protocols on maxillofacial growth in patients aged 7 to 8 years with unilateral complete cleft lip, palate, and alveolus, and will discuss the effect of lip adhesion.

PATIENTS AND METHODS

Patients were selected according to the following inclusion criteria: (1) patients with nonsyndromic complete unilateral cleft lip, palate, and alveolus, without Simonart band; (2) patients of Han nationality from the southwest of China; (3) patients who underwent cheiloplasty and palatoplasty at West China Hospital of Stomatology, People’s Republic of China, from 2005 to 2009;

(4) patients who underwent cheiloplasty at 3 to 6 months and palatoplasty at 12 to 18 months; (5) patients who had lateral cephalometric radiographs available at the age of 7 to 8 years; (6) patients who had not undergone any other operations besides cheiloplasty and palatoplasty; (7) patients who had no preoperative or postoperative orthodontic treatments; and (8) patients who had no family history or trauma history in the craniofacial area. The study protocol was appraised and approved by the Research Subject Review Board and Ethical Scientific Board of Sichuan University. Informed consent was obtained from all patients or their parents.

For all patients, lip adhesion was performed at 1 month of age, cheiloplasty at 3 to 6 months of age, and palatoplasty at 12 to 18 months of age. Cleft palate was closed using the Sommerlad surgical method. Selected subjects were assigned to one of four groups according to the treatment protocol (Table 1). Patients with unilateral complete cleft lip, palate, and alveolus who had a repaired lip and an unrepaired palate were placed in group 1 (lip group). Patients with unilateral complete cleft lip, palate, and alveolus who underwent one-stage palatoplasty were placed in group 2 (one-stage group). Patients with unilateral complete cleft lip, palate, and alveolus who underwent two-stage palatoplasty were placed in group 3, whose hard cleft palate was closed using vomer flap repair at the time of lip repair (vomer flap group). Patients with unilateral complete cleft lip, palate, and alveolus who underwent lip adhesion and two-stage palatoplasty were placed in group 4 (lip adhesion group). The control group (group 5) was composed of 16 patients with unilateral incomplete cleft lip, were of Han nationality, and were age- and sex-matched with the case groups. None of the subjects in the control group had a family or trauma history in the craniofacial area. Treatment protocols for each group are shown in Table 2.

All of the radiographs were taken by the same professional radiologist using the same equipment. All of the cephalometric radiographs were obtained in centric occlusion, with the patients

Table 1. Sample Distribution

Characteristic	Group 1	Group 2	Group 3	Group 4	Group 5
Sex					
Male	4	10	12	6	6
Female	5	7	11	6	10
Total	9	17	23	12	16
Mean age, yr	7.77 ± 0.34	8.12 ± 0.60	7.85 ± 0.69	7.60 ± 0.79	7.84 ± 0.64

Table 2. Treatment Protocols for Each Group

Group	1 Mo	3–6 Mo	12–18 Mo
1 (lip)	—	Cheiloplasty	—
2 (one-stage)	—	Cheiloplasty	Hard- and soft-palate repair
3 (vomer flap)	—	Cheiloplasty plus vomer flap repair	Soft-palate repair
4 (lip adhesion)	Lip adhesion	Cheiloplasty plus vomer flap repair	Soft-palate repair
5 (control group)	—	Cheiloplasty	—

positioned in a standardized upright posture with the transporionic axis and Frankfort horizontal plane parallel to the floor.^{21,22} Patients were posed with centric occlusion in a standardized upright posture, with the transporionic axis and Frankfort horizontal plane parallel to the floor when the radiographs were taken. All measurements were traced and measured by the same researcher using WinCeph7.0 cephalometric software (Rise Corp., Sendai, Japan). Tracing was performed twice, with a 2-week interval between tracings. Intrainvestigator reliability was assessed within 15 randomly selected subjects, and intraclass correlation coefficients were above 0.9 for all measurements, suggesting a satisfactory level of agreement. Mean values were used for the analyses.

The constructed landmarks used were traced according to Ross.²³ Anatomical landmarks are shown in Figure 1 and measurements are listed in Table 3. The posterior nasal spine was not included in the measurements because it was not clear enough in the cleft palate patients to be included for evaluation.²⁴

Statistical Analysis

Statistical analyses were performed using the IBM SPSS Version 19.0 software package (IBM Corp., Armonk, N.Y.). The nature of data distributions was tested with the Kolmogorov-Smirnov test. Multiple comparisons were performed with the Bonferroni test and the Kruskal-Wallis H test. A significant difference was defined at the 95 percent level.

RESULTS

No significant difference was shown in the sex ratio among groups. None of the measurements showed significant differences between men and women within each group. There should be no significant difference in growth between boys and girls between 6 and 10 years of age.^{25,26} Therefore, data from boys and girls in each group were combined in the analyses.

The results showed that no significant differences existed between groups 1 and 5. Group 5 showed a more protruding maxilla

(basion-nasion-A point, basion-nasion-anterior nasal spine, sella-nasion-anterior nasal spine; $p < 0.05$), a longer maxillary sagittal length

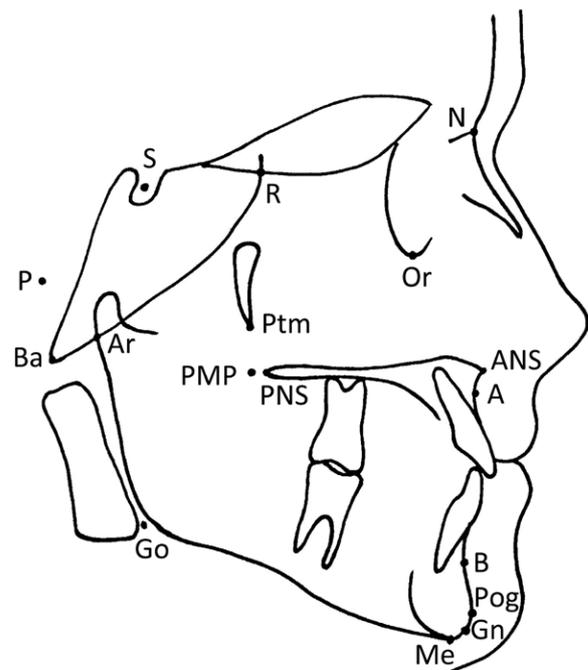


Fig. 1. Landmarks traced on lateral cephalometric radiographs. Sella (S), midpoint of sella turcica determined by inspection; nasion (N), most anterior part of nasofrontal suture; orbitale (Or), most inferior point on infraorbital margin; porion (Po), superior border of external auditory meatus; anterior nasal spine (ANS), most anterior point on nasal spine; (ANB), posterior nasal spine (PNS) most posterior point on nasal plane; A point (A), point of the greatest concavity of the alveolar process of the maxilla; B point (B), point of the greatest concavity of the alveolar process of the mandible; basion (Ba), median point of anterior margin of foramen magnum; gonion (Go), most inferior and posterior point at angle formed by ramus and body of mandible; articular (Ar), point of intersection between the shadow of zygomatic arch and posterior border of mandibular ramus; pogonion (pog) most anterior point on bony chin; gnathion (Gn), point on symphysis between pogonion and menton farthest from condyle; menton (Me), most inferior point on midsagittal plane of symphysis of mandible; registration point (R), point of crossing of greater wing of sphenoid and planum sphenoidale; posterior maxillary point (PMP), construction created by dropping perpendicular line to maxillary plane from pterygomaxillary fissure; pterygomaxillary fissure (Ptm), inferior point in fissure.

Table 3. Statistical Descriptions of All Measurements of Each Group

Variables	Group 1	Group 2	Group 3	Group 4	Group 5
A-B	6.300 ± 3.749	1.821 ± 4.685	2.413 ± 4.928	-0.167 ± 4.357	6.631 ± 3.656
ANB	4.600 ± 2.787	0.932 ± 3.172	1.865 ± 3.674	-0.450 ± 3.552	4.719 ± 2.471
ANS-Me	59.256 ± 5.306	63.090 ± 5.454	57.143 ± 10.128	50.400 ± 11.938	59.456 ± 5.214
ANS-N	46.189 ± 5.581	50.258 ± 4.908	44.526 ± 6.973	39.533 ± 9.402	46.963 ± 4.275
ANS-PMP	44.244 ± 2.928	42.716 ± 4.509	40.070 ± 6.735	33.783 ± 8.327	45.925 ± 4.455
A-PMP	42.667 ± 4.105	40.447 ± 4.314	37.813 ± 6.466	32.033 ± 7.462	43.294 ± 4.300
Ar-Go	35.667 ± 2.089	39.484 ± 4.989	36.813 ± 6.499	30.617 ± 9.974	39.031 ± 3.867
Ar-Go-Me	125.022 ± 5.026	125.342 ± 4.617	126.961 ± 6.880	127.667 ± 6.901	122.812 ± 5.416
Ba-N-ANS	61.944 ± 3.326	56.916 ± 5.280	60.496 ± 3.451	56.750 ± 3.632	64.774 ± 4.599
Ba-N-Pog	55.956 ± 2.625	55.073 ± 2.985	56.626 ± 2.597	56.450 ± 2.862	57.413 ± 3.662
Ba-PMP	37.556 ± 3.462	37.984 ± 3.517	36.239 ± 6.305	38.938 ± 6.428	38.938 ± 3.232
Go-Gn-SN	35.433 ± 5.170	35.253 ± 4.463	35.800 ± 5.566	34.383 ± 5.925	33.838 ± 5.025
Go-Po	64.622 ± 5.082	66.516 ± 5.034	60.417 ± 8.984	55.433 ± 13.111	65.919 ± 4.576
NA-Po	8.967 ± 6.147	0.600 ± 6.557	3.617 ± 7.923	-1.033 ± 7.362	9.906 ± 5.733
N-Ba	93.211 ± 6.354	97.021 ± 7.705	88.183 ± 13.599	77.433 ± 17.802	93.838 ± 7.205
N-S-Ba	128.800 ± 4.382	128.932 ± 4.323	130.491 ± 4.632	126.417 ± 2.190	130.131 ± 6.157
S-Ptm	14.867 ± 2.686	13.668 ± 3.312	13.665 ± 3.669	9.533 ± 1.884	15.656 ± 2.849
R-PMP	39.789 ± 2.690	43.632 ± 4.234	38.700 ± 6.758	35.200 ± 10.348	41.313 ± 4.691
S-Ba	40.511 ± 3.968	42.384 ± 4.433	38.635 ± 5.685	34.833 ± 8.319	39.531 ± 4.244
S-N	62.344 ± 3.450	64.690 ± 4.644	58.152 ± 9.624	51.600 ± 11.885	63.513 ± 4.657
S-N-ANS	81.656 ± 3.188	76.737 ± 5.293	79.983 ± 3.746	77.917 ± 3.043	83.494 ± 4.504
SNA	80.089 ± 3.702	75.210 ± 4.480	77.926 ± 4.208	77.000 ± 2.532	80.838 ± 4.191
Ba-N-A	60.400 ± 3.681	55.395 ± 4.422	58.439 ± 3.503	55.817 ± 3.099	62.138 ± 3.813
N-Me	105.411 ± 9.352	113.332 ± 9.250	101.648 ± 16.118	89.917 ± 20.916	106.400 ± 8.338
SNB	75.511 ± 2.279	74.284 ± 3.248	76.052 ± 3.426	77.467 ± 2.319	76.137 ± 3.809

A, A point; B, B point; ANB, A point-nasion-B point; S, sella; N, nasion; Po, porion; ANS, anterior nasal spine; Ba, basion; Go, gonion; Ar, articular; Pog, pogonion; Gn, gnathion; Me, menton; R, registration point; PMP, posterior maxillary point; Ptm, pterygomaxillary fissure.

(anterior nasal spine-posterior maxillary point; $p < 0.05$) and maxillary basal sagittal length (A point-posterior maxillary point; $p < 0.05$), and a better jaw relationship (A point-nasion-B point; $p < 0.05$) than groups 2, 3, and 4. Groups 1, 2, 3, and 5 had a better maxillary position (sella-ptyergomaxillary fissure; $p < 0.05$) and deeper bony pharynx (basion-posterior maxillary point; $p < 0.05$) than group 4.

Group 2 had higher anterior facial height (anterior nasal spine-nasion, anterior nasal spine-menton, nasion-menton; $p < 0.05$) and posterior facial height (registration point-posterior maxillary point; $p < 0.05$) than groups 3 and 4. Statistical results are shown in Tables 3 and 4.

DISCUSSION

In this study, no significant difference was found in any of the measurements between the lip group and the control group. Both groups had almost the same craniofacial morphology. Therefore, cheiloplasty carried out at 3 months had no detrimental effect on craniofacial morphology. Shao et al.²⁷ found that patients with unilateral cleft lip and palate had an almost normal maxillary sagittal position and a short maxillary sagittal length after lip repair. However, some scholars concluded that lip repair could negatively influence maxillary growth, although it is not the main reason.¹²

All case groups with repaired palates had a less protruding maxilla, short maxillary sagittal length, and unmatched jaw relationship. Thus, palatoplasty—both one-stage and two-stage palatoplasty—adversely affected maxillary sagittal length and position. The effect of palatoplasty on maxillary sagittal growth was in accordance with almost all of the other studies.²⁸ Only a few studies confirmed the excellent anteroposterior maxillary morphology.^{29,30}

When compared with the vomer flap group and the lip adhesion group, the one-stage group had a larger anterior and posterior facial height, and longer cranial length. These measurements were not significantly different between the vomer flap group and the lip adhesion group. Vomer flap repair inhibited maxillary vertical growth, although it reduced the difficulties with and need for lateral releasing incisions at palate repair. Denuded bone existed in the vomer and midline of the plate after vomer flap repair and in the lateral part of the plate after one-stage repair. The resulting scar covered the palate firmly and attached to the palatal bone, with Sharpey fiber connecting the maxilla, palatine bone, and pterygoid plates of the sphenoid together, leading to maxillary growth retardation,^{23,31} which has been proved.^{32,33} Maxillary growths occurs in both the sutures and the periosteal lining.³⁴ Tanino et al.^{35,36} compared two groups of patients: one group with

Table 4. Statistical Results between Every Two Groups

Variables	G1 vs. G2	G1 vs. G3	G1 vs. G4	G1 vs. G5	G2 vs. G3	G2 vs. G4	G2 vs. G5	G3 vs. G4	G3 vs. G5	G4 vs. G5
A-B	0.025*	0.020*	0.013*	0.887	0.781	0.294	0.006*	0.178	0.005*	0.006*
ANB	0.011*	0.051	0.009*	0.932	0.318	0.340	0.002†	0.162	0.015†	0.004†
ANS-Me	0.085	0.722	0.099	0.910	0.015*	0.005*	0.066	0.132	0.549	0.055
ANS-N	0.076	0.785	0.175	0.552	0.003*	0.003*	0.031*	0.118	0.339	0.032†
ANS-PMP	0.290	0.026*	0.005*	0.396	0.153	0.011*	0.034†	0.080	0.002†	0.001†
A-PMP	0.110	0.018*	0.005*	0.650	0.098	0.002*	0.049†	0.059	0.002†	0.001†
Ar-Go	0.036†	0.208	0.045*	0.019†	0.300*	0.013*	0.778	0.036*	0.184	0.022†
Ar-Go-Me	0.892	0.400	0.392	0.366	0.373	0.397	0.205	0.792	0.032*	0.086
Ba-N-ANS	0.009*	0.167	0.013*	0.183	0.028†	0.899	0.000†	0.059	0.002†	0.001†
Ba-N-Pog	0.468	0.570	0.754	0.246	0.098	0.328	0.024†	0.898	0.421	0.503
Ba-PMP	0.712	0.900	0.009*	0.350	0.456	0.002*	0.389	0.011*	0.110	§0.001
GoGn-SN	0.931	0.857	0.700	0.460	0.733	0.720	0.421	0.551	0.246	0.826
Go-Po	0.403	0.167	0.077	0.552	0.004*	0.004*	0.643	0.225	0.012†	0.012†
NA-Po	0.007*	0.062	0.021*	0.821	0.161	0.702	0.000†	0.178	0.011†	0.007†
N-Ba	0.219	0.267	0.013*	0.755	0.006*	0.002*	0.202	0.049*	0.204	0.010†
N-S-Ba	0.946	0.371	0.348	0.506	0.296	0.265	0.462	0.067	0.818	0.109
S-Ptm	0.588	0.529	0.001*	0.462	0.970	0.009*	0.128	0.0016*	0.107	0.001†
R-PMP	0.024†	0.644	0.238	0.692	0.005*	0.030*	0.128	0.389	0.424	0.121
S- Ba	0.372	0.358	0.040*	0.650	0.022*	0.003*	0.108	0.112	0.595	0.061
S-N	0.168	0.090	0.009*	0.610	0.004*	0.002*	0.518	0.118	0.014†	0.006†
S-N-ANS	0.012*	0.173	0.059	0.651	0.053	0.703	0.001†	0.346	0.012†	0.009†
SNA	0.008*	0.148	0.124	0.713	0.081	0.356	0.002†	0.435	0.058	0.050
Ba-N-A	0.008*	0.121	0.009*	0.755	§0.026	0.726	0.000†	0.118	0.002†	0.002†
N-Me	0.052	0.785	0.051	0.610	0.003*	0.002*	0.022*	0.067	0.408	§0.018
SNB	0.359	0.677	0.263	0.649	0.087	0.042†	0.101	0.351	0.937	0.401

G, group; A, A point; B, B point; ANB, A point-nasion-B point; S, sella; N, nasion; Po, porion; ANS, anterior nasal spine; Ba, basion; Go, gonion; Ar, articular; Pog, pogonion; Gn, gnathion; Me, menton; R, registration point; PMP, posterior maxillary point; Ptm, pterygomaxillary fissure; SNA, sella-nasion-A point; SNB, sella-nasion-B point.

*The mean difference is significant at the 0.05 level. The former is larger than the latter.

†The mean difference is significant at the 0.05 level. The former is smaller than the latter.

a repaired hard palate with a vomer flap covered by a full-thickness skin graft, and the other group with a repair by push-back of the mucoperiosteal flaps. The first group showed satisfactory maxillary growth, as no denuded bone was left and less palatal scar formed.^{35,36} Thus, vomer flap repair with a denuded vomer, rather than lip adhesion, inhibited maxillary vertical growth. Swennen et al.^{29,30} also confirmed the reduction in maxillary vertical height. Holland³⁷ and Liao et al.^{38,39} came to almost the same conclusion. Ganesh et al.⁴⁰ carried out a randomized trial, and reported marginally better maxillary growth in the vomer flap repair in terms of dental arch relationships, but with poorer speech outcomes when patients were aged 7 to 9 years. Hay et al.⁴¹ compared patients with and without vomer flap closure of the hard palate at the time of lip repair, and suggested that vomer flap repair has no detrimental effects on maxillary growth. Silva Filho et al.⁴² and Johnston et al.⁴³ came to the same conclusion.

Lip adhesion is used as a solitary preliminary step or in combination with presurgical orthopedics.^{10,44,45} It takes advantage of natural forces to mold the maxillary segments; then, after lip repair, it can be performed under less tension and minimal undermining dissection. The use of lip

adhesion as a single preliminary step for definitive lip closure can cause a collapse of the maxillary segments toward the midline in the unilateral and bilateral cleft lip and palate.^{17,19} As a result, the alveolar and palatal cleft width was reduced. For unilateral cleft lip and palate, the alveolar cleft width decreased from 6.2 to 9.6 mm and the palatal cleft width decreased from 3.8 to 6.7 mm.⁴⁶ This could negatively affect the development of the maxillary arch dimension and nasolabial aesthetics.⁴⁶ In addition, crossbites were also detected after lip adhesion.

In this study, the lip adhesion group had decreased facial height, a shallow bony pharynx, and a retrusive maxilla compared with the control group, but these morphologic differences did not exist in any of the other case groups. The lip adhesion group also had a shallow bony pharynx and a retrusive maxilla compared with the vomer flap group. Thus, lip adhesion inhibited the maxilla from moving forward, and led to a shallow bony pharynx and retrusive maxilla. Lip repair performed at 3 months showed no adverse effects on maxillofacial morphology; whether the morphologic changes listed above resulted from lip adhesion or operation frequency still needs further verification. However, the long-term results

of presurgical orthopedics followed by periosteoplasty and lip adhesion showed adverse effects on maxillary growth.¹⁰

Limitations of our study include unknown original cleft size and small and unequal sample size. Different original cleft size may lead to a different stitch tension and then lead to a different effect on maxillofacial growth. Strict inclusion criteria directly caused small sample size; thus, original cleft size was not considered. Besides, patients aged 7 to 8 years (whose craniofacial growth was not finished) were entered into this study. A final evaluation should be delayed until growth of the craniofacial skeleton is complete.

CONCLUSIONS

In patients aged 7 to 8 years with unilateral complete cleft lip, palate, and alveolus, cheiloplasty had no detrimental effect on craniofacial morphology. Both one- and two-stage palatoplasty inhibited maxillary sagittal growth. Vomer flap repair with denuded bone inhibited maxillary vertical growth. Lip adhesion did adversely affect maxilla position.

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