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The Soft Tissue Angular Analysis of Facial Profile in Unoperated Adult Patients with Unilateral Cleft Palate

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Abstract

Objective The purpose of this study was to investigate the differences in facial profile development between unoperated adult cleft palate (UACP) patients and normal controls and to analyse the reasons for the differences.

Materials and Methods A total of 50 individuals with a unilateral cleft palate and 20 normal controls were selected to undergo angular measurement of their facial profiles. Data with significant differences between the two groups were analysed.

Results Seven angle measurements of the facial profile showed that the mid-facial protrusion of the UACP patients had no significant differences from the control group (p > 0.05). But their angle of the medium face (N'–Trg– Sn) was significantly lower than the non-cleft controls (p < 0.05), suggesting a worse vertical development of the middle face. A significantly larger nasal tip angle (Cm–Sn/ N'–Prn) for UACP patients suggested they had a rounder

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¹ College of Stomatology, Guangxi Medical University, 10 Shuangyong Rd, Nanning 530021, Guangxi, People's Republic of China and blunter nasal tip (p < 0.05). The soft tissue facial angle and chin–lip angle of UACP patients had significant differences from non-cleft controls (p < 0.05), but the head position angle (Sn–Sm–THP) had no significant difference between two groups (p > 0.05), which suggested a steep mandibular plane for UACP patients but without severe retraction of the chin.

Conclusion The development of facial protrusions in UACP patients is similar to that in normal adults, but the vertical development in the middle face is insufficient. Such hypoplasia may be related to the intrinsic deficiency of the maxilla. There is a tendency for flat nasal growth and insufficient development of the chin in UACP patients.

Level of Evidence IV This journal requires that authors assign a level of evidence to each article. For a full description of these Evidence-Based Medicine ratings, please refer to the Table of Contents or the online Instructions to Authors www.springer.com/00266.

Keywords Unoperated adult cleft palate patient \cdot Facial profile of soft tissue \cdot Angular analysis \cdot Mid-face development

Introduction

Cleft palate is the most common congenital deformity in the maxillofacial region, and it occurs in about one in 700 live births worldwide. Cleft palate affects maxillofacial growth, especially sagittal development of the middle face, and it leads to functional disorder, impaired aesthetics and psychosocial barriers. The optimal timing of palatoplasty remains controversial. The optimal timing for the procedure is generally suggested to be from an age before 12 months to 5 years of age. Supporters of early repair suggest performing the operation before the age of 12 months to facilitate speech development [1-4]. In contrast, proponents of delayed repair surgery believe that it is more important to perform the surgery at an age of 5 years to benefit proper maxillofacial growth [5-8]. However, in general, most of the patients undergo palatoplasty at an early age to obtain functional velopharyngeal closure. Many clinical studies have found that a substantial proportion of cleft palate patients who underwent the repair surgery in infancy have different degrees of maxillary hypoplasia and mid-face retraction deformities in adulthood, and other common symptoms include a narrow dental arch, crowded dentition, a mild crossbite in molar occlusion and a severe crossbite in anterior occlusion. Severe secondary dental-facial deformity after cleft palate surgery is one of the main therapeutic focuses in orthognathic surgery. An average of 25% of cleft palate patients with maxillary hypoplasia need surgical treatment to improve facial convexity through maxillary advancement [9, 10]. Do unoperated adult patients also have retraction deformity in their mid-face caused by maxillary hypoplasia? Since most patients undergo repair surgery at an early age in modern society, there are few opportunities to study the growth of the maxilla in adult cleft palate patients who did not receive repair surgical treatment. The purpose of this study was to investigate the growth potential of adult patients with non-surgical cleft palate and to explore the differences in facial profile development between UACP patients and normal controls.

Materials and Methods

Inclusion Criteria

A total of 50 cases were selected from adult patients with cleft palate who did not receive repair surgical treatment and who were admitted to the Oral and Maxillofacial Surgery Department of the College of Stomatology at Guangxi Medical University from January 2012 to November 2018. The inclusion criteria for patients were the following items: (1) Diagnosed with congenital cleft palate; (2) no syndromic cleft or maxillofacial systemic anomalies; (3) 16 years of age or older; (4) no cleft lip or received cleft lip repair before 1.5 years of age; (5) did not undergo other orthopaedic treatments, orthodontic treatment or plastic surgery; and (6) was a native resident of Guangxi Province, China. The subject ages ranged from 18 to 36 years. There were 20 females and 30 males. All patients signed informed consent forms. The study protocol was approved by the Internal Review Board of the Department of the College of Stomatology at Guangxi Medical University. The clinical information and cleft types for the patients were recorded. According to the classification of cleft palate, patients were divided into three groups, including cleft on the soft palate only (SCP); incomplete cleft of the hard and soft palate (ICP); and complete cleft of the hard and soft palate (CCP).

A control group was set up for comparison with the cleft palate group. Their facial profile data were measured to obtain the normal range of these data as a reference. The group consisted of 20 adult volunteers with normal profiles. There were 10 females and 10 males who were native residents of Guangxi Province, China. The inclusion criteria included the following items: normal craniofacial development, normal proportioned facial profile, and basic degrees of facial symmetry with class I occlusion without skeletal craniofacial deformities or pathological facial protrusions or retrusions. The upper and lower teeth were arranged neatly without, or only with minor amounts of, crowding or diastema, and the volunteers did not have a history of orthodontic or cosmetic treatment. The study was approved by the institutional ethical committee, and written informed consent was obtained from all patients and volunteers (Figs. 1, 2, 3, 4).



Fig. 1 Soft tissue landmarks of facial profile in one unoperated adult cleft palate patient



Fig. 2 Angle (1) G–N'–Prn, (2) Cm–Sn–Ls, (3) Cm–Sn/N–Prn, (4) Li–Sm–Pog', (5) N'–Trg–Sn, (6) Sn–Trg–Me', (7) N'Prn–TVL, (8) Sn–Sm–THP

Measurement of Facial Profile Data

According to strict shooting conditions, photographs of patients' standard facial profiles under a natural head position (NHP) were used as a posture for measurement. The true horizontal plane (THP) and true vertical line (TVL) were used as reference planes. Patients were asked to relax their facial muscles and lips naturally, look straight ahead, comb their hair behind their ears and expose their forehead and ears. The teeth were occluded at the maximum occlusion.

Using a Canon EOS700D SLR digital camera with 18 megapixels, a tripod and flash lamp, the camera was adjusted to the manual shooting function. The shutter speed was 1/60 s, and the aperture was f4.0. The subjects were standing at fixed landmarks on the ground. The distance between the camera and the subjects was fixed at 1.7 m. For excluding the facial distortion, the facial profile was shot by a right angle to the horizon of the objects' line of sight. A plumb line and measurement marks in front of the subjects were used to ensure that the photographs were recorded at a 1:1 ratio.

The landmarks and reference angles are shown in Table 1. The measurement items were as follows:



Fig. 3 Angle (9) Ul–N'–Pog', (10) G'–Sn–Pog', (11) G'–Prn–Pog'



Fig. 4 Angle (12) OP-N'Pog', (13) H line-N'Pog', (14) Ul-N'-Ll

Table 1	Landmarks	and reference	lines used	in the	facial	profile analysis
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Landmark/reference line	Description
Glabella point (G')	The intersection of the midpoint line of both eyebrows and the median sagittal plane
Nasion point of soft tissue (N')	The frontonasal suture on soft tissue
Pronasale point (Prn)	The most protruding point of the tip of the nose
Columella point (Cm)	Initial point of columella
Subnasale point (Sn)	The deepest point at the junction between the lower edge of nasal septum and the upper lip
Labrale superius point (Ul)	The most anterior point of the upper lip in sagittal plane
Labrale inferius point (Ll)	The most anterior point of the lower lip in sagittal plane
labial superior (Ls)	The point at the superior margin of the upper lip
labial inferior (Li)	The point at the inferius margin of the lower lip
Mentolabial sulcus point (Sm)	The deepest point in chin-lip sulcus
Pogonion point of soft tissue (Pog')	The most advanced point of chin soft tissue
Menton point of soft tissue (Me')	The intersection of the lowest point on chin soft tissue and the median sagittal plane under NHP
Tragus point (Trg)	The most anterior point in the supratragal notch of the ear
OP line	The line between the midpoint P of N'-Pog' and Trg point
H line	The tangent line between Pog' point and upper lip

- (1) Anterior nasal angle (G–N'–Prn).
- (2) Nasolabial angle (Cm–Sn–Ls).
- (3) Nasal tip angle (Cm-Sn/N'-Prn).
- (4) Chin–lip angle (Ll–Sm–Pog').
- (5) Convex angle of the upper and lower lip (Ul–N'–Ll).
- (6) Convex angle of the upper lip and chin (Ul–N'– Pog').
- (7) H angle (H line-N'Pog').
- (8) Mid-face angle (N'-Trg-Sn).
- (9) Subface angle (Sn-Trg-Me').
- (10) Vertical nasal angle (N'Prn–TVL).
- (11) Head posture angle (Sn–Sm–THP).
- (12) Soft tissue facial angle (OP–N'Pog').
- (13) Facial convexity angle (G'-Sn-Pog').
- (14) Full facial convexity angle (G'-Prn-Pog').

All data were measured three times and then averaged. The data were analysed by SPSS 19.0. The data difference of the facial profiles between unoperated adult patients with cleft palate and normal adults were analysed using an independent samples t test.

Results

Detailed measurements of UACP patients are shown in Table 2, and the measurements from normal controls are shown in Table 3. There were 17 male patients and 4 female patients with CCP, 12 male patients and 14 female patients with ICP, as well as 1 male patient and 2 female patients with SCP. Male patients more often had CCP diseases, while female patients more often had ICP. Three

male patients had anterior crossbite occlusion, but their angles of convexity were normal, and there were no female patients with anterior crossbite occlusion. As shown in Table 4, there were no significant differences in G–N'–Prn, Cm-Sn-Ls, Ul-N'-Ll, H angle, Sn-Trg-Me', Sn-Sm-THP, G'-Sn-Pog' and G'-Prn-Pog' between adult cleft palate patients and the control group (p > 0.05). Four of the 14 measurements in the analysis showed significant differences in the control group for both males and females (p < 0.05), which were for Ll–Sm–Pog', N'–Trg–Sn, OP– N'Pog' and Cm-Sn/N'-Prn. The Ll-Sm-Pog' and Cm-Sn/ N'-Prn measurements were wider in adult cleft palate patients, especially for female patients. The angles for N'-Trg-Sn and OP-N'Pog' were smaller compared to noncleft controls. The difference in N'-Trg-Sn was similar to that in the ICP cases comparing with non-cleft controls (p < 0.05). There was no significant difference in the N'Prn-TVL between female cleft palate patients and the normal female group, but there was a significant difference in males. Female cleft palate patients had a significant difference for Ul-N'-Pog' compared to the control group. The Ul-N'-Pog' of female cleft palate patients was wider than non-cleft controls. Six male patients had a negative measurement for Ul–N'–Ll ranging from -0.1° to -1.8° , and 3 of these cases had anterior crossbite occlusion. Only one female patient had a negative measurement for Ul-N'-Ll with a measurement of -0.4° . There were three male cleft palate patients with negative facial convexity, which suggested they had a concave profile, but the angle was very small (0.5°, 2.8°, 3.6°), and this population did not coincide with the Ul-N'-Ll population with negative measurements. There were no female cleft palate patients

Table 2 Angle measurement of facial soft tissue profile in male and female patients

Patient	Gender	Cleft palate type	Age	Occlusion	G–N′– Prn	Cm–Sn– Ls	Cm–Sn/N– Prn	Li–Sm– Pog	Ul–N'– Ll	Ul–N'– Pog'	H– N'Pog'
1	М	ССР	40	Neutral	149.70	91.30	90.90	177.50	4.60	9.00	11.90
2	М	CCP	20	Crossbite	165.40	107.20	88.30	148.30	- 0.1	7.20	11.00
3	М	CCP	32	Neutral	134.20	95.40	91.60	117.60	4.50	12.20	18.40
4	М	CCP	35	Neutral	135.00	83.90	90.90	140.60	3.80	11.00	20.80
5	М	CCP	25	Neutral	147.80	100.20	94.40	135.00	- 1.5	6.00	9.70
6	М	ICP	31	Neutral	128.10	88.60	88.20	142.10	4.70	11.70	21.70
7	М	ICP	22	Neutral	120.20	81.60	85.20	152.90	2.90	9.50	13.30
8	М	ICP	26	Neutral	130.10	86.90	82.70	142.90	2.50	7.50	11.90
9	М	CCP	19	Neutral	135.40	86.60	86.30	145.60	2.60	9.80	16.40
10	М	ICP	27	Neutral	138.20	76.30	60.90	134.40	0.70	5.50	16.70
11	М	CCP	32	Neutral	132.40	88.40	78.40	127.80	4.10	9.30	14.90
12	М	CCP	23	Neutral	149.90	89.80	88.90	143.70	1.20	6.80	10.90
13	М	ICP	22	Neutral	137.90	98.60	88.60	143.70	3.40	9.90	15.80
14	М	ICP	18	Neutral	136.90	82.60	90.70	143.80	5.30	14.60	18.10
15	М	ICP	20	Neutral	129.10	96.10	89.70	147.00	6.60	12.00	19.80
16	Μ	CCP	42	Neutral	153.50	100.60	87.20	130.20	- 1.70	8.30	10.70
17	М	CCP	19	Neutral	143.40	77.40	75.10	144.30	3.40	12.40	21.60
18	М	CCP	18	Neutral	147.20	86.00	98.80	121.70	2.30	9.80	14.10
19	М	SCP	26	Neutral	122.30	96.20	88.50	140.90	4.50	10.50	15.40
20	Μ	CCP	19	Neutral	139.70	79.40	90.30	134.30	4.40	13.70	17.10
21	М	ICP	18	Neutral	138.90	79.10	95.40	161.90	2.70	16.00	19.60
22	М	ICP	30	Neutral	130.00	108.60	100.40	137.90	3.10	11.80	18.30
23	М	ICP	22	Crossbite	143.90	77.30	82.00	139.60	- 1.30	5.80	7.70
24	М	CCP	34	Neutral	132.20	84.50	83.40	145.20	0.30	5.70	8.30
25	М	CCP	19	Neutral	135.60	100.80	88.80	141.70	5.90	13.70	21.60
26	М	ICP	28	Neutral	140.40	82.80	84.60	155.70	1.40	11.00	15.70
27	М	ICP	23	Crossbite	143.80	88.70	96.30	173.60	- 1.80	4.70	16.80
28	М	CCP	36	Neutral	143.50	92.30	90.60	106.20	2.90	9.80	15.70
29	М	CCP	21	Neutral	148.30	104.30	87.40	146.60	- 0.40	9.30	14.10
30	М	CCP	20	Neutral	136.10	87.60	98.70	145.10	6.90	13.10	15.30
31	F	ICP	27	Neutral	142.70	83.20	101.20	145.20	7.00	13.50	20.30
32	F	CCP	17	Neutral	142.40	82.40	95.20	133.60	5.70	10.80	13.60
33	F	ICP	26	Neutral	125.90	104.40	98.50	157.20	3.20	10.50	15.40
34	F	ICP	17	Neutral	124.90	81.80	90.40	116.00	5.80	13.90	23.50
35	F	ICP	22	Neutral	135.70	77.10	92.40	148.60	3.40	9.00	14.00
36	F	ICP	18	Neutral	139.50	100.30	98.30	145.40	5.30	12.00	18.90
37	F	ICP	20	Neutral	140.00	104.70	100.80	131.00	4.30	9.10	14.30
38	F	CCP	42	Neutral	131.00	114.10	103.60	133.00	5.30	11.80	18.50
39	F	ICP	28	Neutral	131.40	95.80	103.20	129.60	1.10	11.00	15.50
40	F	ICP	21	Neutral	141.00	85.20	94.10	160.00	3.10	9.10	14.90
41	F	ICP	22	Neutral	134.20	89.80	88.30	146.80	4.10	9.60	17.50
42	F	ICP	20	Neutral	137.20	115.10	109.90	161.60	5.20	12.20	18.80
43	F	ICP	30	Neutral	127.30	104.10	90.50	150.40	3.90	8.80	13.90
44	F	CCP	28	Neutral	124.10	92.00	103.20	133.90	3.30	11.40	16.60
45	F	ICP	25	Neutral	133.10	104.50	100.40	139.20	6.80	12.20	19.50
46	F	SCP	18	Neutral	143.20	107.10	110.70	139.80	3.20	9.10	16.60
47	F	ICP	19	Neutral	140.40	124.20	104.40	152.00	3.10	8.40	14.90
48	F	CCP	31	Neutral	146.90	118.70	103.80	159.40	- 0.40	7.70	10.30

Table 2 continued

Patient	Gender	Cleft palate type	Age	Occlusion	G–N′– Prn	Cm–Sn– Ls	Cm–Sn/N– Prn	Li–Sm– Pog	Ul–N'– Ll	Ul–N'– Pog'	H– N'Pog'
49 50	F F	ICP SCP	23 38	Neutral Neutral	142.50 145.70	110.10 79.50	84.90 81.50	150.90 162.40	5.00 5.90	13.70 6.00	22.40 5.50
Patient	Gender	Cleft palate type	Age	Occlusion	G-N'- Prn	Cm–Sn– Ls	Cm–Sn/N– Prn	Li–Sm– Pog	Ul–N'– Ll	Ul–N'– Pog'	H– N'Pog'
1	М	ССР	40	Neutral	22.80	23.50	29.20	90.30	103.60	173.70	154.70
2	М	CCP	20	Crossbite	29.50	34.90	22.70	82.20	100.30	170.70	147.00
3	М	CCP	32	Neutral	26.50	35.40	26.50	86.50	102.80	175.70	156.00
4	М	CCP	35	Neutral	23.30	29.90	24.90	79.80	103.60	169.00	152.60
5	М	CCP	25	Neutral	25.00	27.50	16.60	81.20	106.30	178.80	157.90
6	М	ICP	31	Neutral	23.20	20.90	25.10	80.30	110.30	173.90	153.00
7	М	ICP	22	Neutral	27.40	35.20	28.40	94.70	113.10	- 3.60	143.50
8	М	ICP	26	Neutral	24.30	28.60	26.30	90.80	104.70	173.60	140.80
9	М	CCP	19	Neutral	26.60	33.20	22.10	80.20	97.90	173.80	150.50
10	М	ICP	27	Neutral	28.20	30.00	25.50	86.80	109.70	176.80	147.70
11	М	CCP	32	Neutral	24.90	32.90	28.20	77.30	97.60	172.10	144.50
12	М	CCP	23	Neutral	26.40	27.40	24.90	91.50	103.30	173.90	154.80
13	М	ICP	22	Neutral	26.10	31.30	29.60	87.10	102.30	172.20	148.20
14	М	ICP	18	Neutral	28.40	32.50	26.80	77.90	104.70	166.60	142.00
15	М	ICP	20	Neutral	28.10	29.70	27.10	77.60	105.10	168.70	145.30
16	М	CCP	42	Neutral	23.50	31.30	29.80	85.90	100.30	171.50	151.20
17	М	CCP	19	Neutral	25.60	31.90	22.70	89.30	102.80	170.90	151.00
18	М	CCP	18	Neutral	24.50	29.50	21.10	88.90	100.10	177.60	158.00
19	М	SCP	26	Neutral	25.20	34.20	27.10	79.60	105.60	176.20	150.00
20	М	CCP	19	Neutral	25.20	32.60	28.50	86.30	98.90	175.30	149.80
21	М	ICP	18	Neutral	28.20	38.20	27.80	90.00	103.30	- 0.50	149.80
22	М	ICP	30	Neutral	24.20	35.90	30.80	79.00	97.30	164.90	143.50
23	М	ICP	22	Crossbite	25.10	28.20	13.30	83.90	103.90	179.80	157.50
24	М	CCP	34	Neutral	24.30	26.10	19.20	88.50	105.80	- 2.80	158.50
25	М	CCP	19	Neutral	25.90	32.10	31.90	76.40	102.10	161.10	144.50
26	М	ICP	28	Neutral	24.50	33.80	23.30	86.20	101.10	175.40	149.90
27	М	ICP	23	Crossbite	26.80	41.30	24.50	92.90	99.60	167.60	148.90
28	М	ССР	36	Neutral	26.90	32.20	15.50	69.20	102.20	174.70	157.30
29	М	ССР	21	Neutral	22.40	34.00	28.00	87.80	100.90	171.50	151.80
30	М	CCP	20	Neutral	25.70	38.20	29.70	80.70	103.20	169.00	152.70
31	F	ICP	27	Neutral	24.40	30.70	28.30	71.50	101.20	165.40	138.10
32	F	CCP	17	Neutral	24.20	31.50	22.60	80.30	101.90	177.20	154.20
33	F	ICP	26	Neutral	25.30	36.40	36.90	89.70	104.80	174.50	154.60
34	F	ICP	17	Neutral	24.80	28.00	28.80	78.50	105.00	162.60	134.10
35	F	ICP	22	Neutral	23.90	32.00	30.30	91.40	106.20	179.10	150.70
36	F	ICP	18	Neutral	24.90	32.00	26.30	81.70	102.60	161.60	148.70
37	F	ICP	20	Neutral	24.20	29.10	27.90	79.30	101.30	170.70	147.10
38	F	CCP	42	Neutral	27.00	30.90	30.70	86.10	104.00	176.10	160.50
39	F	ICP	28	Neutral	22.80	28.10	28.00	84.00	102.90	176.20	153.60
40	F	ICP	21	Neutral	24.40	30.00	36.20	97.60	107.10	176.60	152.10
41	F	ICP	22	Neutral	25.00	32.00	3.30	86.00	102.40	172.60	147.30
42	F	ICP	20	Neutral	27.30	31.60	27.30	79.90	105.10	164.40	147.10
43	F	ICP	30	Neutral	22.60	28.00	29.00	81.00	108.30	172.00	151.30

Table 2 continued

Patient	Gender	Cleft palate type	Age	Occlusion	G–N′– Prn	Cm–Sn– Ls	Cm–Sn/N– Prn	Li–Sm– Pog	Ul–N'– Ll	Ul–N'– Pog'	H– N'Pog'
44	F	ССР	28	Neutral	22.40	25.90	30.40	84.00	103.70	178.90	153.90
45	F	ICP	25	Neutral	23.60	27.70	26.50	78.80	103.50	168.90	147.60
46	F	SCP	18	Neutral	23.10	26.70	18.00	81.30	99.60	174.30	157.70
47	F	ICP	19	Neutral	22.30	32.00	37.60	91.10	103.70	172.00	148.20
48	F	CCP	31	Neutral	25.80	32.30	24.90	83.60	108.50	170.70	152.60
49	F	ICP	23	Neutral	29.10	34.80	35.30	79.20	99.20	156.60	134.70
50	F	SCP	38	Neutral	23.60	32.20	29.80	90.30	105.40	179.50	149.90

with negative facial convexity and Ul–N'–Ll. CCP patients were more likely to have negative Ul–N'–Ll than ICP patients (4 vs 2, all male). The patient with the most serious negative Ul–N'–Ll (-1.8°) was an ICP patient.

Discussion

Current studies of maxillary development in adult patients with cleft palate are mostly based on the analysis of lateral cephalometric radiographs. However, because the thickness of soft tissue covering the surface of hard tissue is not uniform, and the soft-to-hard tissue ratios varied greatly [11], the profile of the hard tissue cannot fully reflect the profile of the soft tissue. In clinical practice, these soft tissue morphologies sometimes provide us with a more intuitive impression. Patients with secondary maxillary hypoplasia caused by a repaired palate mainly complain about the non-aesthetic mid-face retraction rather than disordered occlusion when they ask for treatment at a hospital. The aesthetic information for the face, which plastic or maxillofacial surgeons pay attention to, also often comes directly from the patient's own photographs or standard facial profile photographs rather than X-ray findings [12]. Therefore, the authors of the present study used the patient's profile photographs to study whether the soft tissue profile development of adult cleft palate patients is similar to the development of normal adults through multiangle analysis.

Maxillary hypoplasia secondary to cleft palate surgery has long been noticed. Some studies suggested that maxillary hypoplasia was not necessarily caused by cleft palate itself; in contrast, without the restriction of scars, patients with a bilateral cleft lip and palate often have excessive premaxillary protrusion [13, 14]. Such dislocation of the premaxillary in the cleft lip and palate complicates the treatment severely, and some scholars even suggested using special devices to limit the over protrusion of the premaxillary to facilitate cleft lip and palate surgery later on [15]. Therefore, scholars believe that cleft lip and palate surgery itself may be the cause of maxillary hypoplasia. After using 62 beagle puppies as animal models in a 1990 study, Bardach et al. [16] indicated that the size of the area of the palatal bone exposed following palatoplasty does subsequently affect craniofacial growth, and the contraction of the scar on the palate may also disturb maxillary development. Such local soft tissue scars around the maxilla not only limit the maxilla development but also restrict maxilla advancement in orthognathic surgery [17] and maintain a high relapse rate (25-40%) [18]. However, compared to patients after palatoplasty, adult patients with a unilateral cleft palate may develop a maxillary protrusion. Capelozza et al. [19] conducted a cephalometric analysis of 26 white adult patients with complete unilateral cleft lip and palate and found their maxilla was smaller and more protruded than normal individuals. Liao and Mars [20] suggested the growth inhibition caused by cleft lip and palate (CLP) is mainly influenced primarily by the height of maxillary development and, second, by the length. They believed that reduced posterior height of the maxilla could be attributed to intrinsic effects while anterior height could be attributed to functional effects [20]. Shetye and Evans [21] reported a study of cephalometric analysis of 30 adult patients with complete unilateral cleft lip and palate, and they found the sizes of patients' maxillae were normal and the maxillae protruded forward. They indicated that the potential for normal maxillary development still exists in patients, and disturbances of maxillary development may be mainly related to the surgical intervention of such acquired factors. Cao et al. [22] conducted a study of cephalometric analysis in 20 unoperated overt cleft palate patients and found patients had smaller mandibles and shorter maxillae but had normal maxillary protrusions.

Our results showed that the protrusion of the mid-facial profile of the unoperated cleft palate patients had no significant differences from the normal control group, which also meant that there was no tendency for abnormal maxillary protrusion. The nasolabial angle (Cm–Sn–Ls) is one of the most routine indices for assessing the beauty of the mid-facial profile in orthognathic surgical treatment. The

 Table 3
 Angle measurement of facial soft tissue profile in non-cleft controls

Volunteer	Gender	Age	G–N′–Prn	Cm–Sn–Ls	Cm-Sn/N-Prn	Li–Sm–Pog'	Ul–N′–Ll	Ul-N'-Pog'	H–N'Pog'
1	М	19	135.40	84.10	86.70	106.20	6.00	10.50	16.60
2	М	22	136.60	99.20	85.50	139.00	3.40	9.80	18.60
3	М	20	128.40	108.40	82.40	135.30	2.60	9.00	15.30
4	М	23	129.20	90.50	88.50	136.90	3.50	11.00	16.00
5	М	21	128.10	100.00	90.80	127.90	3.40	10.10	16.70
6	М	25	133.60	97.10	89.80	126.40	3.40	9.60	16.90
7	М	19	132.30	80.00	72.10	115.60	3.60	4.70	8.10
8	М	24	146.50	94.30	88.30	140.00	4.20	11.60	19.80
9	М	21	136.00	77.40	83.20	106.50	3.60	11.40	18.60
10	М	22	139.70	81.70	86.60	115.80	4.00	10.50	18.70
11	F	19	140.30	96.40	83.20	142.70	4.70	9.70	16.00
12	F	22	137.90	71.30	77.20	140.00	3.50	8.50	15.10
13	F	18	142.30	114.00	90.70	134.60	3.40	9.30	16.50
14	F	23	132.70	95.50	77.60	138.70	3.80	8.50	16.10
15	F	21	128.60	122.10	106.80	120.00	0.70	7.70	13.00
16	F	22	141.00	109.00	88.80	137.80	2.50	5.40	10.40
17	F	23	137.70	88.60	86.40	128.30	2.50	5.90	12.40
18	F	20	135.60	101.90	83.40	114.10	4.80	10.80	20.40
19	F	18	133.00	101.60	96.00	121.90	3.10	8.00	15.60
20	F	21	130.60	83.10	83.10	116.60	4.70	9.10	13.70
Volunteer	Gender	Age	G-N'-Prn	Cm–Sn–Ls	Cm-Sn/N-Prn	Li-Sm-Pog'	Ul-N'-Ll	Ul-N'-Pog'	H–N'Pog'
1	М	19	26.50	35.20	33.40	87.70	108.30	174.30	156.60
2	М	22	27.50	33.60	32.10	79.70	110.20	173.90	146.80
3	М	20	28.40	35.40	34.90	80.00	109.20	169.10	148.10
4	М	23	28.30	30.90	29.60	83.40	105.80	173.00	152.90
5	М	21	28.00	29.10	39.00	91.60	110.20	177.40	156.20
6	Μ	25	28.70	30.30	36.60	91.90	104.60	174.10	156.50
7	Μ	19	25.70	31.80	24.50	79.50	109.80	175.80	155.90
8	Μ	24	29.50	33.40	34.00	82.70	112.60	161.70	160.20
9	Μ	21	29.00	31.20	35.20	84.70	106.00	172.10	142.20
10	Μ	22	29.50	30.20	19.20	68.40	105.60	167.80	142.50
11	F	19	29.00	37.70	34.30	77.60	102.50	165.80	138.90
12	F	22	22.70	28.20	34.60	89.80	116.90	174.50	152.90
13	F	18	27.60	31.20	26.30	78.00	103.40	168.70	149.30
14	F	23	27.60	30.00	27.80	76.80	101.70	166.70	140.50
15	F	21	27.00	36.80	34.80	81.30	104.20	169.60	151.50
16	F	22	25.00	29.50	30.90	85.50	106.90	175.30	154.10
17	F	23	25.10	26.30	28.70	82.30	107.60	173.80	148.30
18	F	20	26.20	29.90	25.20	66.60	110.30	162.60	142.50
19	F	18	27.00	32.20	35.90	87.40	106.30	169.40	152.00
20	F	21	28.40	31.70	30.40	79.20	106.00	175.20	150.30

convex angle between the upper and lower lip (Ul–N'–Ll) refers the position relationship between the upper and lower lip. The smaller the angle is, the more forward the lower lip is relative to the upper lip. The angle of facial

convexity (G'–Sn–Pog') and the angle of total facial convexity (G'–Prn–Pog') reflect the protrusion of the middle face relative to the whole profile. In our study, there were no significant differences in the Cm–Sn–Ls, Ul–N'–Ll, H

Reference angle	Male patients	Normal male controls	<i>p</i> value of male comparison	Female patients	Normal female controls	<i>p</i> value of female comparison
G–N′–Prn	138.97 ± 9.54	134.58 ± 5.68	p = 0.179	136.46 ± 7.12	135.97 ± 4.64	p = 0.845
Cm–Sn–Ls	89.97 ± 9.09	91.27 ± 10.21	p = 0.706	98.71 ± 14.20	98.35 ± 14.97	p = 0.949
Cm–Sn/N–Prn	88.167 ± 7.67	85.39 ± 5.38	*p = 0.000	97.77 ± 7.92	87.32 ± 8.92	*p = 0.003
Ll-Sm-Pog'	142.26 ± 14.28	124.96 ± 13.11	*p = 0.001	144.8 ± 12.61	129.47 ± 10.63	*p = 0.002
Ul-N'-Ll	2.60 ± 2.46	3.77 ± 0.89	p = 0.150	4.22 ± 1.82	3.37 ± 1.27	p = 0.197
Ul–N'–Pog'	9.92 ± 2.92	9.82 ± 1.97	p = 0.902	10.49 ± 2.13	8.29 ± 1.65	*p = 0.007
H line–N'Pog'	15.44 ± 3.95	16.53 ± 3.28	p = 0.473	16.25 ± 4.07	14.92 ± 2.74	p = 0.360
N'-Trg-Sn	25.62 ± 1.81	28.11 ± 1.24	*p = 0.000	24.54 ± 1.75	26.56 ± 1.87	*p = 0.006
N'-Trg-Sn (ICP only)	26.21 ± 1.88	28.11 ± 1.24	*p = 0.013	24.61 ± 1.81	26.56 ± 1.87	*p = 0.018
Sn-Trg-Me'	31.75 ± 4.32	32.11 ± 2.18	p = 0.802	30.60 ± 2.66	31.35 ± 3.55	p = 0.521
N'Prn-TVL	25.24 ± 4.55	31.85 ± 5.97	*p = 0.000	27.91 ± 7.50	30.89 ± 3.856	p = 0.250
Sn-Sm-THP	84.29 ± 5.89	82.96 ± 6.86	p = 0.619	83.77 ± 5.98	80.45 ± 6.55	p = 0.176
OP–N'Pog'	103.08 ± 3.65	108.23 ± 2.61	*p = 0.000	103.82 ± 2.57	106.58 ± 4.45	*p = 0.04
G'-Sn-Pog'	154.94 ± 53.47	171.92 ± 4.59	p = 0.326	171.50 ± 6.46	170.16 ± 4.41	p = 0.561
G'-Prn-Pog'	150.43 ± 5.08	151.79 ± 6.42	p = 0.497	149.20 ± 6.88	148.03 ± 5.43	p = 0.643

Table 4 Average and statistical analysis of angle measurement of facial soft tissue profile in male and female patients and control group

*p < 0.05

angle, G'–Sn–Pog' and G'–Prn–Pog' between adult cleft palate patients and the control group (p > 0.05).

Although the main tendency of the mid-facial profile protrusion is normal in our cases, mild mid-facial dysplasia still exists in patients. The angle of the medium face (N' -Trg-Sn) of a cleft patient had a significant difference from the non-cleft controls. The lower angle measurements of cleft palate patients suggested that their vertical development of the middle face was worse than that of the control group. This outcome is consistent with the results of Ye's scholarly research [23]. Their study found that both the unilateral cleft lip-palate adults and adults with lip repairs had vertical and sagittal hypoplasia of the maxilla. They believed that palatoplasty had no adverse effects on vertical dysplasia of the maxilla, and they suspected the main cause was intrinsic deficiency in maxillary growth. In 1996, Capelozza et al. [24] conducted a cephalogram study of 93 male adult patients who had undergone palatoplasty or cheiloplasty, and the results suggested the facial morphology of the patients was mainly affected by surgical repair of the lip, and palatal repair seems to have had little impact. However, most scholars had negative opinions on whether cheiloplasty is the main cause that worsened the maxillary development [25-27]. Therefore, we tended to exclude consideration of the influence of cleft lip repair on maxillary development. In this study, there were no cleft lips in patients with ICP, but a significant difference was also observed in the mid-facial angle between these patients and non-cleft controls. Therefore, we support the hypothesis that the abnormal vertical height development of the maxilla in patients with cleft palate is related to intrinsic deficiency.

There were three male patients with a slight concave facial profile (negative facial convexity: 0.5° , 2.8° , 3.6°), which indicated mild maxillary hypoplasia, and other patients had straight or convex facial types. However, all three patients had a neutral occlusal relationship. The normal Ul–N'–Ll angle values of those three patients also indicated that their labrale superius point was in front of the labrale inferius point, which meant the patients did not show the lip shape that is characteristic of maxillary retraction and anterior crossbite occlusion. The slight concave facial angle and normal relative relationship between the upper and lower lips led these patients to have relatively normal facial profiles.

The nose shapes of patients in this study had unique characteristics. The wider the anterior nose angle (G-N'-Prn) was, the more the position of N' was retracted from the other two points, and the deeper the position of the nose root was. A larger nasal tip angle (Cm-Sn/N'-Prn) refers to the smoother curve from the nasal dorsum to nasal base, which led to a rounder and blunt nasal tip. Vertical nose angle (N'Prn-TVL) refers to the protrusion of the nasal dorsum in the natural head position. The larger the angle was, the more the nasal dorsum protruded. In our study, the N'Prn-TVL of the male patients showed flatter nasal development than non-cleft male controls (p < 0.05), and N'Prn-TVL indicated that the nasal development of female patients was straighter than that of male patients. Smahel et al. [28] conducted cephalometric studies of 114 adult males with cleft lips with or without cleft palate, and they

suggested that flattening of the nose was an important feature of cleft patients, especially in cases with complete cleft palate. This finding was consistent with our findings. In their study, the large horizontal slope of the columella led to a reduction of the nasolabial angle (Cm–Sn–Ls), which was observed in cleft patients. However, in our study, the mean of the nasolabial angle of cleft patients had no statistically significant difference from that of non-cleft controls.

Moreover, the chin-lip angle of both male and female cleft patients was significantly different from the non-cleft controls. The chin-lip angle of the patients was wider compared to the control group. An excessive chin-lip angle usually means underdevelopment and retraction of the chin. Angle OP-N'Pog', which is also called the soft tissue facial angle, had a significant difference between UACP patients and non-cleft controls in our study. The OP plane is considered to be approximately parallel to the SN plane of hard tissue, and therefore, it is used as a base plane for photogrammetric analysis. The OP-N'Pog' represents the protrusion degree of the chin. The smaller facial angle in UACP patients supports the fact that the chin development of the patients was insufficient. Malek et al. [29] found lack of prenatal folic acid supplementation made it easier for microgenia to occur in a male cleft palate rat model. Cao et al. [22] found adult submucous cleft palate patients who did not receive a repair operation developed a smaller mandible, a steeper mandibular plane and a maxillary with normal protrusion. They believed that the possible cause of the underdevelopment of the mandible was recurrent respiratory infection and mouth breathing, which occurs in over 70% of the cleft population. Ye et al. [23] also discovered this phenomenon and suggested that the clockwise rotation of the mandible after palatoplasty in cleft palate patients was caused by compensatory retraction of the mandibular alveolar process. To some extent, these hypotheses explain the same results for the chin that we have observed. Despite two indicators suggesting that the chins of UACP patients were hypoplastic and retracted, the H angle (H line–N'Pog') was still not significantly different from that in the control group, which meant there was supplementary evidence that the UACP patients had sufficient protrusions in the mid-face. However, it is worth mentioning that there was no significant difference between the cleft group and non-cleft controls for the angle of the head position (Sn-Sm-THP). Sn-Sm-THP was used to analyse the lower profile orientation. Wider angles show a tendency to be prognathic, while lower angles suggest retrognathic profiles [30]. Combining the data from these two angles, we can see that the mandibular planes of cleft patients were steep and the development of the chins was indeed insufficient; however, severe retraction did not occur. This lower profile is aesthetically tolerable.

Currently, there is no evidence about the perfect surgical procedure and timing for cleft palate surgery, and there is no gold standard management method [31, 32]. There are many different opinions about the prognosis of different surgical methods, such as early cleft palate surgery, delayed cleft palate surgery and two-stage cleft palate surgery. Considering the commonly poor pronunciation of cleft palate patients, as well as the mild maxilla vertical hypoplasia, nasal hypoplasia, and the compensatory chin hypoplasia of UACP patients found in our study, despite the facial profiles developing normally in general, these results were not sufficient to prove the necessity of delayed palatoplasty. In contrast, proper early cleft palate surgery may help remove this dysplasia.

Conclusion

The development of facial protrusions in adult cleft palate patients who did not receive a repair operation is similar to that of normal adults, but vertical development in the middle face is insufficient. Such hypoplasia may be related to intrinsic deficiencies of the maxilla. There is a tendency for flat nasal growth and insufficient development of the chin in this cleft population.

Compliance with Ethical Standards

Conflict of interest We declare that we have no conflict of interest.

Ethical Approval The study protocol was approved by the Internal Review Board of the Department of the College of Stomatology at Guangxi Medical University.

Informed Consent All patients signed informed consent forms.

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