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Efficacy of modified anterior maxillary segmental distraction osteogenesis based on 3D visualisation for the treatment of maxillary hypoplasia among adolescents with cleft lip and palate

Yan Jiang[1,2†], Canyang Jiang[2,3†], Bin Shi[1,2,3], Jianping Huang[2,3], Yue Huang[2,3], Rihui Wang[2,3], Xiaohong Huang[1,2*†], Li Huang[1,2,3*†] and Lisong Lin[1,2,3*†]

Abstract

Background This study evaluates a three-dimensional (3D) visualisation design combined with customized surgical guides to assist anterior maxillary segmental distraction osteogenesis (AMSDO) in correcting maxillary hypoplasia in adolescents with cleft lip and palate (CLP), focusing on treatment outcomes, satisfaction and the validity of 3D planning.

Methods This retrospective cohort study was conducted at a single hospital in China. Between January 2020 and December 2023, 12 adolescents with CLP with maxillary hypoplasia were included. An advanced 3D simulation was used to convey the treatment strategy to the patients and their families. A customized surgical guide and distraction osteogenesis device were designed. Cephalometric analysis evaluated AMSDO changes and long-term stability. Patient satisfaction was assessed. The Chinese version of the Child Oral Health Impact Profile was used to evaluate the children's oral health-related quality of life before and after treatment. The postoperative outcomes were compared with the planned outcomes by superimposing the actual postoperative data onto the simulated soft tissue models and calculating the linear and angular differences between them.

Results One patient experienced postoperative gingivitis, yielding an 8.33% complication rate. Most patients (83.33%) were highly satisfied with the target position, with the rest content. Cephalometric analysis showed

†Yan Jiang and Canyang Jiang contributed equally as first authors.

†Xiaohong Huang, Li Huang, and Lisong Lin contributed equally to this manuscript as corresponding authors.

*Correspondence:
Xiaohong Huang
13905006768@139.com
Li Huang
lihuang@fjmu.edu.cn
Lisong Lin
dr_lls@163.com
Full list of author information is available at the end of the article

significant improvements in various indices post-traction. Quality-of-life scores significantly improved post-treatment. The discrepancies in facial soft tissue between the simulated and actual results were within clinically satisfactory ranges.

Conclusions Digitally designed surgical guides effectively treat maxillary hypoplasia in adolescents with CLP, ensuring stability, reducing complications, reducing dependency on operator experience, and enhancing satisfaction and health outcomes. Although the simulated results were clinically acceptable, it is important to inform patients of potential variations in the predicted soft tissue.

Keywords Anterior maxillary segmental distraction osteogenesis (AMSDO), Maxillary hypoplasia, Cleft lip and palate (CLP), Customized 3D-printed surgical splints, Child oral health impact profile (COHIP)

Background

Cleft lip and palate (CLP), the most prevalent congenital anomaly in humans, affects approximately 1 in every 700 newborns worldwide. The disease not only seriously affects facial aesthetics but also leads to functional disorders such as eating, speech, and hearing impairment. Therefore, CLP causes serious psychological trauma to both the patients and their families. Although patients with CLP can be treated through a comprehensive sequence of multidisciplinary collaborations at birth, most of them often develop severe skeletal Class III malocclusion, characterised by maxillary retrusion, upper arch constriction, and anterior and posterior dental crossbites, mainly because of congenital tissue defects and acquired scar contractures [1–4]. Currently, the treatment of maxillary hypoplasia in patients with CLP remains a complex problem [5]. In approximately 25–47% of patients with CLP, orthodontic treatment alone is not adequate for treating maxillary hypoplasia. They usually require surgical interventions, such as maxillary distraction osteogenesis and orthognathic surgical treatment in adulthood [6–9].

Anterior maxillary segmental distraction osteogenesis (AMSDO) has been considered the most appropriate surgery for patients with CLP and severe skeletal Class III malocclusion since its proposal at the beginning of the twenty-first century [10]. Compared with conventional adult maxillary Le Fort-I osteotomy, AMSDO can increase the maxillary anterior bone volume through distraction osteogenesis, anteriorly shift the maxilla, improve the lateral facial appearance, preserve the teeth, and avoid the aggravation of palatopharyngeal closure insufficiency [11–14]. More importantly, AMSDO can be performed during adolescence, which is also the best time for orthodontic treatment [15], providing a time window for improving the lateral appearance and occlusal function. Such procedures have been reported to improve children's physical and mental health and help them better integrate into society [16]. However, their special physiological characteristics, such as a lower maxillary sinus floor and underdeveloped tooth roots [17, 18], which result in higher surgical risks, and limited cognition [19] lead to poor postoperative cooperation, posing challenges to the overall treatment that are different from those of conventional adult treatment.

Previous research on AMSDO has mainly focused on the surgical stability, success rates, and enhancements of the distractor [14, 20–22]. Few studies have examined how to reduce surgical risks and postoperative complications from the physician's and the patient's point of views. Hence, this study proposed using three-dimensional (3D) visualisation to simulate treatment plans and communicate with adolescents and their families preoperatively, improving the digital surgical guide plate to assist in surgery. The effectiveness of the treatment was evaluated based on the treatment outcomes, impact on the quality of life (Child Oral Health Impact Profile [COHIP] [23]), patient satisfaction and the validity of 3D planning.

Methods

Study participants and study design

Between 1 January 2020 and 31 December 2023, 12 consecutive patients with CLP who met the following inclusion criteria at the First Affiliated Hospital of Fujian Medical University were retrospectively enrolled and analysed: 1) aged between 10 and 18 years; 2) underwent AMSDO at our hospital during 1 January 2020 and 31 December 2023; 3) diagnosed with severe skeletal Class III malocclusion and severe midface hypoplasia because of CLP (maxillary anterior displacement of at least 6 mm; SNA less than 78°); 4) had normal mandibular development and without a family history of inherited mandibular excess; 5) had maxillary occlusion plane deviation of less than 2 mm and facial asymmetry less than 10%; 6) did not have palatal fistula and/or alveolar synostosis, or completed bone grafting or palatal fistula repair treatment at least 6 months before AMSDO; and 7) had complete clinical data. The exclusion criteria for this study were as follows: 1) inability to complete treatment because of the COVID-19 epidemic or personal reasons; 2) incomplete clinical data, 3) inability to speak and communicate; 4) mental disability, and 5) syndromic CLP. All children underwent a joint multidisciplinary discussion before treatment to develop a complete treatment plan consisting of oral and maxillofacial surgery, orthodontic, prosthodontic, endodontic, periodontal, and other oral subspecialty treatments.

Surgical design and procedure

Three-dimensional visualisation design, improved surgical guide plate and customised tooth-borne distractor design and fabrication

Patients underwent a thorough pretreatment examination, including medical history, clinical evaluation, photographic documentation, cephalometric lateral imaging, cone-beam computed tomography (CBCT), 3D computed tomography (CT), and an ultra-hard plaster model. The maxillofacial surgeon imported the patient's 3D CT in DICOM format into the Proplan CMF software (version 3.0; Materialise, Belgium) to simulate osteotomies avoiding critical anatomical structures, such as tooth roots, maxillary sinuses, and the nasal floor (Figure 1). Using these data, 3D-printed customized surgical guides were fabricated (Figure 2). We also integrated the osteotomy plan into the intraoperative navigation system (Brainlab, German). The distance of the DO was established based on the ideal position of the maxilla relative to the anterior cranial base, growth potential of the mandible, occlusal relationships, and the requirement for restoration. Three-dimensional visualisation was used to simulate the surgical plan, traction effects, and present the treatment results to patients and their families for better communication. Subsequently, we selected reasonably anchored teeth and designed a personalised tooth-supported distractor (Figure 3; Wilder Dental Equipment Co., Ltd., Fujian, China).

Surgical technique

Maxillary anterior distraction osteogenesis was performed under general anaesthesia using a meticulously designed incision strategy. The preoperative osteotomy plan as well as the guide design were imported into the surgical navigation system, registered, and rectified. The incision originated at the proximal midpoint of the left first premolar in the maxillary vestibular sulcus and extended across its counterpart on the right side. Successive layers, including the mucosa, submucosal tissue, and periosteum, were incised down to the bone surface, unveiling the anterior maxillary region and pyriform margin. The customized surgical guide was fixed in its designed position according to the position of the pyriform foramen and the root projection. If positioning was difficult, the exact position of the guide was collectively determined using 4–5 edge turning points on the guided palate by the navigation system. The bone cortex was incised along the edges of the guide using an ultrasonic

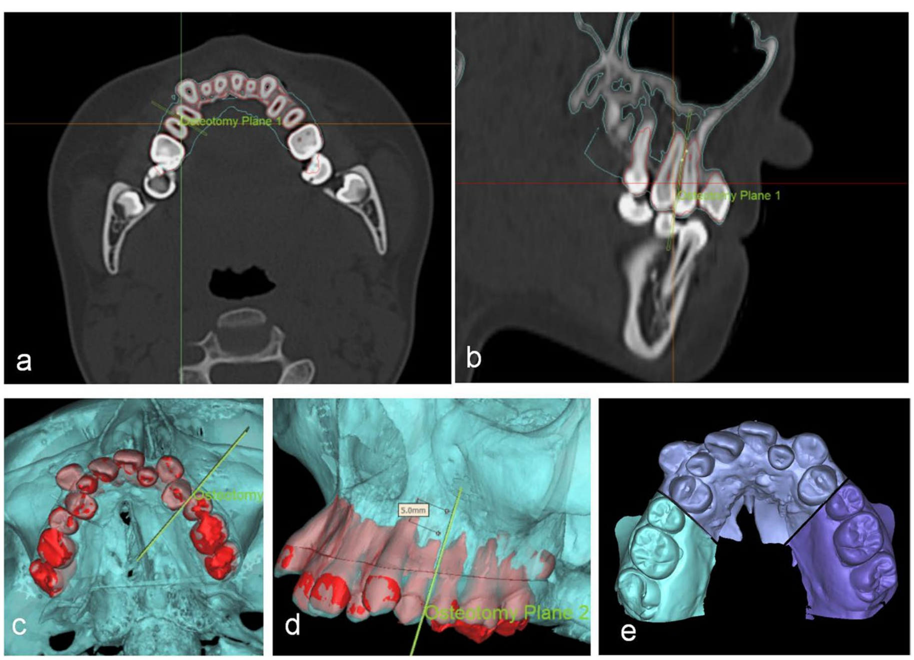

Fig. 1 Planning surgical pathways for different cross-sections **a** Relationship between the right maxillary osteotomy line and the teeth (cross-sectional view) **b** Relationship between the right maxillary osteotomy line and the teeth (sagittal view). **c** Relationship between the left maxillary osteotomy line and the teeth (3D elevation view). **d** Relationship between the left maxillary osteotomy line and the teeth (3D front view). **e** Tooth-bone segment after simulated osteotomy (3D elevation view)

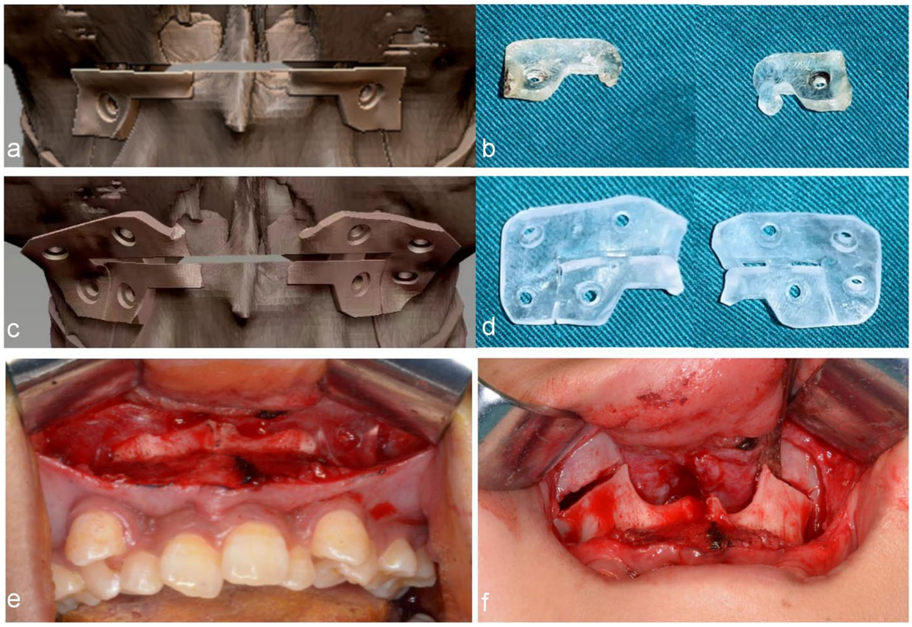

Fig. 2 Surgical guide plate. **a** Software simulation of customized modified guides. **b** Customized modified guide plates (small). **c** Software simulation of conventional guides. **d** Conventional guide plate (large). **e** Intraoperative use of guide plates. **f** Intraoperative osteotomy line

Fig. 3 Customised tooth-borne distractor (expander in the posterior region can be replaced by a transpalatal arch)

scalpel. Subsequent osteotomy procedures were performed using a scalpel and chiselling along a preestablished bone suture. The nasal septum was dissected using a septal osteotome, and the incision was closed tightly after confirming that the anterior portion of the maxillary bone was sufficiently loosened and free.

Postoperative procedure

Following initial intraoral wound healing at the 5-day postoperative mark, the tooth-supported distractor was cemented into the mouth and distracted 3–4 times daily at a rate of 1 mm/day. Upon reaching the pre-determined target position, lateral cephalometric radiographs were taken to confirm that the sagittal position of the maxilla was within the normal range. Patients were then questioned regarding their satisfaction with their lateral appearance, considering mild post-operative swelling, as indicated by changes in the upper lip thickness observed in pre-and post-operative lateral cephalometric radiographs. In cases of pronounced satisfaction, the traction process was halted, and Data B (target position) was recorded. If the patients were not sufficiently satisfied, traction was continued to the patients' ideal position, and Data C (ideal position) was recorded. After a 3-month fixation period, the distraction device was systematically removed, marking the commencement of postoperative orthodontic interventions. Data D (1-year post-operative position) were recorded after a 1-year follow-up period to obtain relevant information, and patient satisfaction with the post-treatment lateral appearance was assessed again.

Data collection

Measurement and analysis of lateral films for cephalic positioning

In this study, patients underwent lateral cephalometric radiography in the natural head position at a magnification ratio of 1:1.11. The patients with 'force bite' and a counterclockwise rotation of the mandible were instructed to relax the facial muscles with eyes closed and teeth in the resting position. For the missing upper incisors, we simulated the shape of the tooth upright in the alveolar bone on cephalometric lateral films for measurement. Using the Dolphin software, soft tissue lateral profile contour lines and anatomical landmarks were identified on the lateral cephalometric radiographs: sella [S] centre of the pituitary of fossa of the sphenoid bone, nasion [N] intersection of the internasal suture with the nasofrontal suture in the midsagittal plane, orbitale [Or] lowest point of the root of orbit, porion [Po] highest point of the ear canal, tip of the anterior nasal spine [ANS], tip of the posterior nasal spine [PNS], point A [A] deepest point of the curve of the maxilla between ANS and the dental alveolus, point B [B] most posterior point in the concavity along the anterior border of the symphysis, menton [Me] most inferior point of the bony chin, pogonion [Po] most anterior point of the bony chin, gnathion [Gn] midpoint between Po and Me, gonion [Go] mandibular angle point, pterygomaxillary fissure point [Ptm], pronasale [prn] tip of the nose, subnasale [sn] point where the nose connects to the centre of the upper lip, and vermilion borders labial margin point [24]. Cephalometric data were denoted as the initial position, fixed position, and 1-year post-operative position corresponding to the pre-AMSDO stage (initial position A), end of the fixation period when removing the distractor (target position B + ideal position C), and 1-year post-AMSDO (1-year post-operative position D), respectively. Key cephalometric measurements (SNA, SNB, ANB, ANS-PNS, Distance-A, Y-axis angle, anterior tooth overjet [OJ], upper Lip to E-plane, lower lip to E-plane, facial convexity, and nasolabial angle) were systematically compared at these three critical time points, encompassing the degree of postoperative stabilisation and the transformations resulting from the procedure. Comprehensive details of the cephalometric landmarks and reference lines are presented in Table 1. The same author performed all cephalometric measurements and reviewed them subsequently after a 6-week interval to ensure unwavering accuracy.

Questionnaire survey

The questionnaire used in the study contained three parts: 1) basic demographic information; 2) COHIP; and 3) a satisfaction survey. All patients completed the hard-copy survey according to the researcher's instructions.

For basic demographic information, data on sex, date of birth, place of residence, school attended, and grade were collected.

The COHIP was administered to patients before AMSDO and 1 year after AMSDO treatment [25]. It was developed by Broder et al. in 2007 and consists of 34 entries in five subscales: oral health, functional health, social and emotional health, school environment, and self-image. The first four subscales were negative: oral health (10 entries), functional health (six entries), social-emotional health (eight entries), and school environment (four entries). Responses were scored as follows: 4 = never, 3 = rarely, 2 = sometimes, 1 = often, and 0 = always. The fifth subscale (self-image, six entries) belongs to positive content and is rated opposite to the negative entries: 0 = never, 1 = almost never, 2 = sometimes, 3 = often, and 4 = always. The total COHIP score ranged from 0 to 136, with higher scores indicating a higher oral-related quality of life. This study used the Chinese version of the COHIP, translated and validated by Lin et al. [23]. Although the original version of the COHIP was applicable for ages 8–15 years [26], the present study relaxed the age of the COHIP to 8–18 years, taking into account the different national conditions in each country [27, 28].

For patient satisfaction survey, the patients were requested to complete a satisfaction survey upon

Table 1 Cephalometric landmarks and reference lines

Landmark	Definition
SNA (°)	The S–N-A angle
SNB (°)	The S–N-B angle
ANB (°)	The A-N-B angle
ANS-PNS (mm)	The distance from point ANS to point ANS
Distance-A (mm)	The distance between point A by superimposing the post-AMSDO onto the initial lateral cephalogram according to the SN plane
OJ (mm)	The overjet
Y-Axis (°)	The lower internal angle where the Y-axis intersects the FH plane
Upper Lip to E-Plane (mm)	Distance from the upper lip to the plane joining the tip of the nose and the soft tissue pogonion
Lower Lip to E-Plane (mm)	Distance from the lower lip to the plane joining the tip of the nose and the soft tissue pogonion
Nasolabial angle (°)	Angle between nasal columella and upper lip
Facial convexity (°)	The angle calculation of glabella, subnasal, and pogonion

reaching the targeted position as planned and 1 year after AMSDO treatment according to the improvement level of facial aesthetics (Table 2) [9, 29].

Comparison of simulated outcome and actual 1-year postoperative outcome that was performed using CT images

We compared the CT data of the patients 1 year after surgery with the simulated facial data after AMSDO, following the method of Wang et al. [30]. The patients' CT data were stored in the DICOM format and imported into Proplan. 3D models of soft and hard tissue were constructed and exported as STL files. The 3D simulated models and 1-year postoperative actual models were superimposed using the surface registration method on the basis of the cranial base and orbital area, which remained unchanged after AMSDO. The root mean square deviations between the registered surfaces were all smaller than 0.5 mm, which was considered acceptable to certify the precision. Thereafter, we measured the nasolabial angle, facial convexity, and distance between the upper and lower lips to the E-plane in two sets of 3D images for soft tissue profile comparison.

Statistical analysis

Continuous variables are presented as the mean and standard deviation (SD). The paired samples t-test or Wilcoxon test was used for pre- and post-treatment comparisons, depending on the normality of the data. All statistical analyses were performed using SPSS 25.0, with $P<0.05$ indicating significance. Graphs were drawn using GraphPad Prism software (version 7.0).

Results

Patient demographics and treatment effect

Between 1 January 2020 and 31 December 2023, 12 patients underwent AMSDO and were followed up for 1 year post-treatment. The age of the patients ranged from 12 to 16 years, with a mean age of 13.42 ± 1.44 years, of which four were males and four were females (Table 3). The diagnostic types of CLP were as follows: one case of simple cleft palate (without alveolar cleft), five cases of unilateral CLP with an alveolar cleft, and six cases of bilateral CLP with an alveolar cleft. The anterior traction used in treatment ranged from 6 to 10 mm, with a mean of 7.67 ± 1.07 mm. Five patients had clear anatomical features of the maxilla, allowing the guide plate to be positioned accurately, while the other required navigation-assisted positioning. During the entire treatment period, only one patient developed postoperative gingivitis. The patient's symptoms improved significantly after oral hygiene education and active treatment. None of the patients had treatment-related complications such as root damage, severe bleeding, mucosal tearing, infection, tooth and nerve damage, bone necrosis, pulpitis, gingival recession, or abnormal tooth movement. We did not detect

Table 2 Postoperative patient satisfaction survey

Degree of satisfaction	Extremely satisfied, n (%)	Satisfied, n (%)	Unsatisfied, n (%)
Aesthetical outcome	Significant correction of facial aesthetic	Minor imperfections of facial aesthetic	No obvious improvement of facial aesthetic

Table 3 Baseline patient demographics

Patient s	Sex	Age (years)	Diagnosis	Alveolar cleft grafted	Distance of distraction (mm)	Is there complication?
1	Female	15	UCLP	Yes	8	No
2	Male	12	UCLP	Yes	7	No
3	Male	13	BCLP	Yes	7	No
4	Male	12	UCLP	Yes	8	No
5	Female	15	ICP	No	7	No
6	Male	12	BCLP	Yes	8	No
7	Male	13	UCLP	Yes	6	Yes (gingivitis)
8	Male	13	BCLP	Yes	9	No
9	Female	12	UCLP	Yes	7	No
10	Female	15	BCLP	Yes	7	No
11	Male	16	BCLP	Yes	8	No
12	Male	13	BCLP	Yes	10	No

BCLP bilateral cleft lip and palate, *ICP* isolated cleft palate, *UCLP* unilateral cleft lip and palate, *G* gingivitis any differences between the outcomes of the two positioning methods. All patients in our study underwent orthodontic treatment after AMSDO and achieved satisfactory facial profiles.

Skeletal changes, soft tissue changes, and stability after AMSDO

Table 4 shows the changes at the initial position (denoted as A, before AMSDO), fixed position (denoted as B + C, 3 months after a distraction), and 1-year post-operative position (denoted as D, 1 year after AMSDO). Post-treatment cephalometric analysis (fixed and 1-year post-operative positions) showed a clear improvement in the maxilla and no statistically significant change in the mandible compared with the initial position. Comparing the 1-year post-operative and fixed positions, we found a tendency for regression. However, the change was not statistically significant ($P>0.05$), indicating that the maxilla was significantly lengthened with AMSDO to correct the maxillary hypoplastic deformity. The treatment effect was stable for the following year.

Quality of life of patients

Comparison of COHIP scale factors and total scores before and after treatment

Among the five factors on the COHIP scale, score for three factors, including oral health, social-emotional health, and self-image, as well as the total score, were significantly higher after treatment ($P<0.05$) (Table 5 and Figure 4). Functional health was improved after treatment but was not statistically significant.

Patient satisfaction

Figure 5 shows the software-simulated target bitmap and the real target bitmap. When traction reached the target position, 10 of 12 patients (83.33%) were very satisfied with the target position, and two patients (16.67%) were satisfied with the target position, of which one requested to continue forward traction from the target position for 1 day and then back off to the original target position. The other patient continued forward traction from the target position for 0.5 days and stopped traction to reach the ideal position (Table 6). One year after treatment, 11 of the 12 patients (91.67%) were very satisfied with their lateral facial appearance, and one (8.33%) was satisfied.

Table 4 Changes of skull structure on the lateral cephalograms at initial position (A), fixed position (B + C), and 1-year post-operative position (D)

Landmark	Initial Position (A)		Fixed position (B + C)		1-year post-op position (D)		(B + C) *vs.*A	D *vs.* A	D *vs.* (B + C)
	Mean	SD	Mean	SD	Mean	SD	*P-value*	*P-value*	*P-value*
SNA (°)	76.71	3.04	82	3.36	81.43	2.88	0.000	0.000	0.438
SNB (°)	76.57	3.91	77.14	3.29	77.02	3.36	0.470	0.569	0.873
ANB (°)	0.14	3.76	4.86	4.08	4.41	3.2	0.000	0.000	0.569
Y-Axis (°)	64.43	3.1	65.57	2.76	64.71	3.35	0.117	0.706	0.241
ANS-PNS (mm)	49.00	2.45	53.29	1.98	52.86	2.41	0.000	0.000	0.485
Distance-A (mm)	0	0	5.95	1.54	5.68	1.23	0.000	0.000	0.673
OJ (mm)	-1.20	2.32	3.57	2.60	2.92	2.56	0.000	0.000	0.332
Upper Lip to E-Plane (mm)	-5.47	2.23	-1.25	1.88	-1.21	1.65	0.000	0.000	0.942
Lower Lip to E-Plane (mm)	1.64	1.54	0.56	1.28	0.48	1.24	0.036	0.025	0.863
Facial convexity (°)	178.85	7.87	172.34	5.96	173.45	5.87	0.000	0.000	0.276
Nasolabial angle (°)	68.6	5.66	87.53	2.88	86.88	2.78	0.000	0.000	0.354

Table 5 Comparison of COHIP scores before and after treatment (n = 12)

Factor	Pre-treatment	Post-treatment	t/Z	*P* value
Oral health well-being	25.89 ± 3.33	30.00 ± 5.10	-3.406	0.009
Functional well-being	17.33 ± 5.70	20.89 ± 1.36	-1.923	0.091
Social-emotional well-being	19.78 ± 6.57	26.89 ± 1.17	-3.240	0.012
School environment	16.00 (13.50, 16.00)	16.00 (14.00, 16.00)	-1.000	0.317
Self-image	9.78 ± 3.03	16.78 ± 2.77	-9.635	0.000
Total scores	87.78 ± 10.44	109.78 ± 5.61	-11.077	0.000

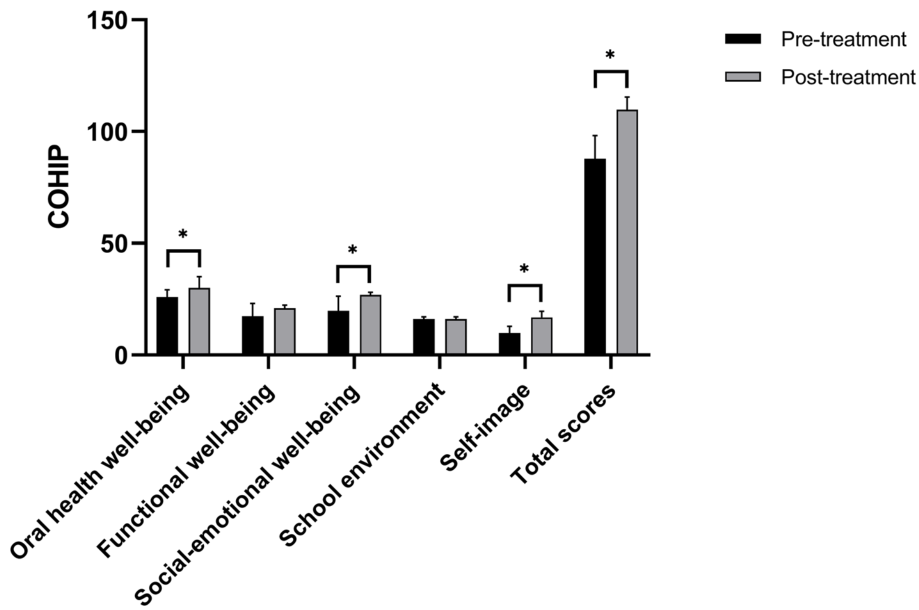

Fig. 4 Comparison of various factors and total scores of the COHIP scale before and after treatment. COHIP, Child Oral Health Impact Profile

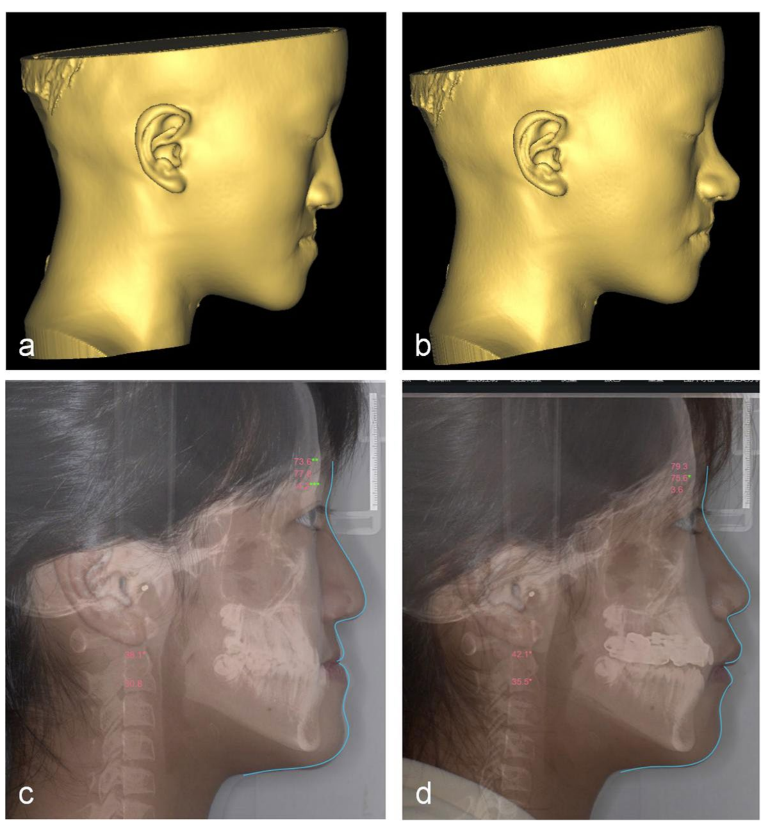

Fig. 5 Comparison of surgical simulation and actual face profile. **a** Facial simulation before treatment. **b** Facial simulation at the target position. **c** Actual facial profile before treatment. **d** Actual facial profile at the target position

Table 6 Patient satisfaction with treatment (n = 12)

	Extremely satisfied, n (%)	Satisfied, n (%)	Unsatisfied, n (%)
At the target position	10 (83.3%)	2 (16.7%)	0
1 year post-treatment	11 (91.7%)	1 (8.3%)	0

Figure 6 shows the patient's facial profile and intraoral occlusion before treatment, when the target position was achieved as planned, and 1 year after AMSDO treatment.

Validity of 3D planning in predicting the soft tissue outcome after AMSDO

We compared the simulated facial profile with the real profile at 1 year postoperatively and found that the simulated soft tissue profile exhibited greater facial convexity and a larger nasolabial angle. The upper lip was positioned more posteriorly to the E-plane, and the lower lip was positioned more anteriorly to the E-plane. These differences were statistically significant ($P < 0.05$) (Table 7). The overlap of the two STL files indicated discrepancies in the positions of the lips and the tip of the nose, whereas the paranasal and base of the nose fit well (Fig. 7).

Table 7 Comparison of simulated and actual postoperative outcomes was using CT images before and 1 year after surgery

Variable	Simulated		1-year post-operation		Simulated vs.1-year post-operation
	Mean	SD	Mean	SD	*P-value*
Upper Lip to E-Plane (mm)	-2.23	0.88	-1.09	1.05	0.009
Lower Lip to E-Plane (mm)	1.58	0.74	0.56	1.15	0.017
Facial convexity (°)	174.64	4.96	171.35	5.45	0.002
Nasolabial angle (°)	90.45	3.88	87.21	2.55	0.000

Fig. 6 Profile and intraoral occlusion of the patient before AMSDO, reaching the target position as planned and 1 year after AMSDO treatment. **a** Photographs, intraoral view and lateral cephalometric radiograph before AMSDO. **b** Photographs, intraoral view and lateral cephalometric radiograph when the target position was reached as planned. **c** Photographs, intraoral view and lateral cephalometric radiograph 1 year after AMSDO treatment

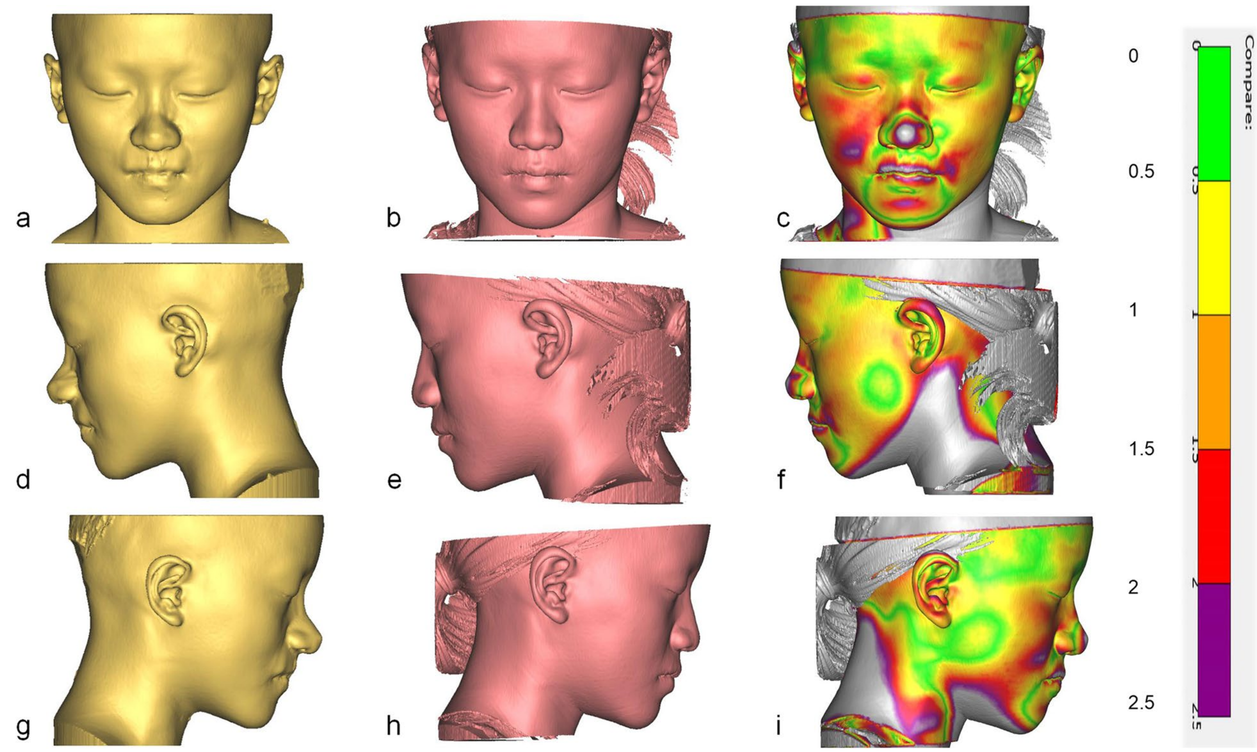

Fig. 7 Overlap of the 'simulated' and '1-year post-operative' 3D models. The bar on the left side indicates the differences between the two models. The areas with purple colouration exceeded the 2-mm threshold (areas of inaccuracies). **a**, **d**, **g** Facial simulation at the target position. **b**, **e**, **h** Actual outcome 1 year post-operation. **c**, **f**, **i** Overlap of the 'stimulated' and '1-year post-operative' 3D models

Discussion

In this study, aiming to improve AMSDO for adolescents with CLP, we used 3D visualisation to simulate the treatment plan, communicate with children and their families, and improve guide plates to assist surgery. Twelve adolescent patients underwent AMSDO using the customized guide plate. After treatment, the patients' maxilla significantly lengthened and showed stable outcomes over the following year. Only one patient developed postoperative gingivitis. The discrepancies in facial soft tissue between the simulated and actual results were within clinically satisfactory ranges. Most patients were very satisfied with the treatment results, and their quality of life significantly improved.

Although AMSDO has numerous advantages, it is inevitably associated with postoperative complications. According to Sunil et al. [31], the probability of complications associated with AMSDO, such as bleeding, haematoma, mucosal tear, root damage, traction device deformation, dislodgement, postoperative infections, and the need for secondary surgery, is approximately 21.17%. These complications are broadly related to the surgeon's experience, surgical approach, outcomes, and devices. In this study, only 1 of the 12 patients developed postoperative gingivitis. After hygiene education and standardised oral hygiene care, no significant inflammation of soft tissues or wound infections were observed. Importantly, none of the patients had any complications such as root injury, mucosal tear, fracture of the anterior osteotomy segment, or postoperative haematoma. The low complication rate may be related to several factors. First, integrating advanced digital tools and a customized surgical guide plate is an advanced approach to surgical planning and execution, which can result in a lower risk of damage, shorter surgery time, and lower caregiver burden among adult patients with maxillary hypoplasia [32–34]. As the anatomical structures of adolescents are more susceptible to irreversible intraoperative damage in AMSDO that may affect growth and development, our study modified guided plate by reducing its size and performing navigation-assisted guide positioning when necessary during operation to compensate for positioning deviations due to the reduced size. A Smaller 3D guide combined with ultrasonic bone cutters that reduce intraoperative stripping range is a notable feature that minimises the dependence on the operator's experience and surgical trauma and subsequently lowers the incidence of postoperative complications, such as soft-tissue oedema, haemorrhage, and infection. Moreover, the reduction in tissue swelling facilitates a more accurate judgement by the physician and patient with regards to satisfaction with the lateral facial appearance in conjunction with the lateral film when traction reaches the target position. Meanwhile, in this study, we reduced the risk of root resorption, which may result from the process of aligning the tooth for root control prior to osteotomy [35], and shortened the overall treatment time by avoiding root damage through precise guide positioning. Second, we focused on the psychological condition of children with CLP. Adolescents have a high potential for growth and development, and their tissues are active in response to external stimuli, which is the best period for orthodontic treatment. Notably, during this time, children are in the 'formal operations' period. They may have the experience of an imaginary audience for special orthodontic appliances. However, as they cannot correctly guess the audience's reaction, they need guidance to help them accurately assess their insights to better cooperate with the treatment to get the best therapeutic effect [19]. In children with CLP, psychological counselling of patients, parents, and the general public is needed alongside the treatment of CLP to achieve positive outcomes and improve patient well-being [36]. Although the planning software available in the market are not completely reliable for soft tissue prediction, and there may be some bias in patient satisfaction results when the patients were asked regarding their satisfaction at the second and third postoperative weeks as they still have a small degree oedema, adolescents with CLP experience various mental health benefits from this communication process. We observed significant improvement in postoperative cooperation during this time compared to that when the patients did not have a good understanding of the therapy. This preoperative demonstration may help enhance the confidence of children and their families, reduce psychological fear, and strengthen cooperation, helping them overcome various postoperative difficulties and finally a high satisfaction in our study.

Effective scale surveys can clarify the patient's condition and provide better guidance for treating the disease. Several researchers have studied the oral health, quality of life, and psychological status of children with CLP using various scales [37–39]. The COHIP is one of the most frequently used tools to evaluate children's oral health. It was developed and released by Broder et al. [26, 40, 41] in 2007 and has been used in many countries, such as the Netherlands, South Korea, the UK, and China [23, 42, 43]. The validity and reliability of COHIP have been confirmed in various countries, including China [23, 28, 44–46]. Our results indicate that oral health, socioemotional health, self-image, and total scores on the COHIP scale significantly improved after treatment ($P<0.05$), which is consistent with the findings of previous studies [25, 47]. Notably, improved indicators were largely associated with the patients' increased self-identity, which may come from improved appearance and increased participation throughout the treatment.

Tabchi et al. pointed out that when using various software to predict the postoperative results of orthognathic surgery, hard tissue prediction is highly accurate and reliable, but soft tissue prediction remains challenging. The software Proplan CMF performs relatively well compared to others [48]. Therefore, our study employed Proplan to simulate the treatment process. Regarding the accuracy of our modified method for simulation in hard tissue, a 3D visualisation simulation of the surgical path in conjunction with intraoperative navigation was employed to achieve precise osteotomy with no damage to important anatomical structures. Post-treatment cephalometric analysis indicated that the maxilla was significantly lengthened with AMSDO, and maxillary hypoplastic deformity was corrected, consistent with previous findings [32–34]. However, on comparing the overlap between the simulated post-AMSDO soft tissue and the actual soft tissue reconstructed from CT scans 1 year post-operation, we found that while the nasal base and paranasal regions fit well, there were significant discrepancies in the positions of the nasal tip and lips, similar to the finding of Lee et al. [49]. In most studies, the success criteria for simulation are set at a 2-mm difference in linear measurements and a 4° difference in angular measurements [48]. Although the discrepancies in facial soft tissue between the virtual and actual results in our study were within these ranges, the difference between them was statistically significant ($P<0.05$). This may be due to the fact that the patients included in this study were adolescents who still have growth potential, especially in the lower part of the face. Additionally, the inclination of the teeth also affects the position of the lips to a certain extent. Therefore, patients must be informed of potential variations in the predicted soft tissue profile to avoid inappropriate expectations.

A limitation of the study was that this improved treatment method increased the time spent on design and communication, as well as the cost of producing the guides. Moreover, its restricted sample size, featuring only pre- and post-treatment comparisons within the same patient group, presents a noteworthy constraint, whereas the retrospective and non-randomised design limits the generalisability of the results. Future research should aim to encompass a larger pool of participants and conduct randomised controlled trials to substantiate the efficacy of this technique further. Additionally, we hope to utilise artificial intelligence to learn and simulate facial appearance alterations according to different distances of DO to enhance the precision of soft tissue prediction.

Conclusions

In summary, using a 3D visualisation design alongside customized guide plate-assisted AMSDO demonstrated superior effectiveness in treating maxillary hypoplasia in adolescents with CLP by reducing postoperative complications and dependence on the operator's experience. Although the simulated results were clinically acceptable, it is important to inform patients of potential variations in the predicted soft tissue. More attention should be paid to the psychological state of adolescents with CLP to improve overall satisfaction with treatment. It must be emphasised that less-experienced surgeons and patients who are fearful and resistant to treatment particularly benefit from this approach, despite the substantial preoperative workload.

Abbreviations

CLP	Cleft lip and palate
AMSDO	Anterior maxillary segmental distraction osteogenesis
3D	Three-dimensional
COHIP	Child Oral Health Impact Profile
CBCT	Cone-beam computed tomography
CT	Computed tomography
S	Sella
N	Nasion
Or	Orbitale
ANS	Anterior nasal spine
PNS	Posterior nasal spine
A	Point A
B	Point B
Me	Menton
Po	Pogonion
Gn	Gnathion
Go	Gonion
Ptm	Pterygomaxillary fissure point
prn	Pronasale
sn	Subnasale

Supplementary Information

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Supplementary Material 1.

Supplementary Material 2.

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Authors' contributions

LLS, HL, SB, HXH and JY contributed to the study conception and design. JY, JCY and HJP performed data analysis. JY and JCY wrote the first draft of the manuscript, and all authors commented on the previous versions. All the authors have read and approved the final version of the manuscript.

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Availability of data and materials

The datasets used and/or analysed during the current study available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

This study was approved by the Medical Ethics Committee of the First Affiliated Hospital of Fujian Medical University (approval no. MRCTA, ECFAH of FMU [2020] 212) and performed in accordance with the 1964 Helsinki Declaration and its later amendments. All patients and their relatives provided written informed consent before inclusion in this study.

Consent for publication

Written informed consent was obtained from the parents for the publication of clinical details.

Competing interests

The authors declare no competing interests.

Author details

[1]Department of Stomatology, The First Affiliated Hospital of Fujian Medical University, Tai-Jiang District, No.20 Cha-Ting-Zhong Road, Fuzhou 350005, China. [2]Department of Stomatology, National Regional Medical Center, Binhai Campus of the First Affiliated Hospital, Fujian Medical University, Fuzhou 350212, China. [3]Department of Oral and Maxillofacial Surgery, the First Affiliated Hospital of Fujian Medical University, Fuzhou 350005, China.

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