Long-term Stability of Skeletal, Dental, and Facial Esthetic Outcomes Following Maxillary Distraction Osteogenesis with Rigid External Distractor in Subjects with Cleft Maxillary Hypoplasia

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Abstract

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Background: Maxillary hypoplasia, common in patients with cleft lip and palate, presents significant functional and esthetic challenges, including Class III malocclusion, negative overjet, and midface retrusion. While conventional Le Fort I osteotomy is effective for skeletally mature patients, growing patients are not eligible. Maxillary distraction osteogenesis using a rigid external distractor (MXDO-RED) is a surgical treatment for correcting severe maxillary hypoplasia and can be performed in growing individuals. Long-term data on stability following MXDO-RED, however, is limited by small sample sizes and short follow-up durations.

Objective: This retrospective study evaluated the long-term skeletal stability of MXDO-RED in patients with severe maxillary deficiency associated with cleft lip and palate. Additionally, the study aimed to assess the association between maxillary stability following MXDO-RED and factors including age at distraction, magnitude of advancement, mandibular growth, and mandibular plane angle.

Methods: A retrospective analysis was conducted on 60 subjects with severe cleft maxillary hypoplasia who underwent Le Fort I level MXDO-RED at a craniofacial center from 2008 to 2023. Records, including lateral cephalograms, extraoral and intraoral photos, and dental models, were analyzed at four time points: pre-operative (T0), immediate post-operative (T1), first follow-up (T2), and longest available follow-up (T3). Outcomes were categorized into short-term (total follow-up duration of 0.5 to 3 years post T1) and long-term (total follow-up duration of greater than 3 to 6.5 years post T1) post distraction follow-up groups. Generalized estimating equations (GEE) linear regression was used to evaluate longitudinal stability and the association between stability and secondary factors.

Results: Significant improvements in maxillary retrusion, facial profile, upper lip position, overjet, and crossbite were observed following MXDO-RED. The mean age at the time of distraction was 14.9 ± 1.7 years. The mean sagittal advancement at A-point was 12.7 ± 5.6 mm. Significant sagittal relapse was observed in both the short-term group (−3.5 ± 5.2 mm; 27.8% of advancement; $p < .001$) and the long-term group (−5.5 ± 6.0 mm; 42.6%; $p < .001$). Longitudinal regression confirmed a significant posterior relapse of −0.9 mm per year over the follow-up period for both groups (95% CI: -1.6 to -0.2; $p = .011$). No significant vertical changes were observed ($p = .850$). The magnitude of initial advancement was the strongest factor associated with maxillary sagittal relapse. Each additional millimeter of sagittal advancement was associated with 0.42 mm of subsequent horizontal relapse (95% CI: -0.62 to -0.23; $p < .001$) and 0.88 mm of vertical relapse (95% CI: -1.03 to -0.74; $p < .001$). Mandibular body growth (Gonion–Menton) was significantly associated with both sagittal ($p = .005$) and vertical ($p = .041$) maxillary displacement after adjusting for follow-up duration. Age at distraction and mandibular plane angle were not significantly associated with relapse. Upper lip position demonstrated significant progressive retrusion over the follow-up period ($p = .011$). Of the 57 subjects for whom status of further orthognathic surgery could be determined, secondary orthognathic surgery was completed or planned for 50.9%, and 49.1% of subjects did not require further surgery.

Conclusion: MXDO-RED achieves substantial correction of severe maxillary hypoplasia in growing patients with cleft lip and palate with significant improvements in facial profile and occlusion. Sagittal relapse approaching half of the initial advancement in the long term presents a clinical challenge. The strong association between magnitude of advancement and mandibular body growth and maxillary displacement underscores the multifactorial nature of post-distraction instability. The finding that half of subjects required secondary orthognathic surgery reinforces the importance of follow-up through skeletal maturity and preoperative counseling that MXDO-RED in growing patients may represent a single stage of a multi-stage treatment course.

INTRODUCTION

Maxillary hypoplasia is characterized by underdevelopment of the maxilla, producing a retruded or deficient midface. Its etiology can be congenital, as in craniosynostosis and syndromic conditions such as Crouzon and Apert syndromes, or acquired, most commonly secondary to cleft lip and/or palate repair or facial trauma. (1,2) The resulting functional complications may include class III malocclusion with anterior crossbite and large negative overjet, leading to difficulties in mastication and speech; nasal obstruction and impaired breathing; and velopharyngeal insufficiency, adversely affecting speech quality. (2–8) Esthetically, midface retrusion creates a sunken appearance with relative prominence of the lower lip and chin, which may contribute to psychological distress. (2,6,7) The combination of significant functional impairment, esthetic concerns, and psychosocial burden requires multidisciplinary management and may create an indication for early surgical intervention.

Conventional Le Fort I maxillary advancement orthognathic surgery with rigid internal fixation, with or without adjunctive bone grafting, has become the standard treatment option for skeletally mature patients with or without clefting who have moderate to severe maxillary hypoplasia. This procedure allows reliable three-dimensional repositioning of the maxilla and has demonstrated acceptably stable outcomes. (9,10) In non-cleft populations, longitudinal cephalometric studies report mild skeletal relapse of approximately 1.0 mm, or roughly 18% of surgical movement, at up to 3 years postoperatively, with 89% of postoperative change occurring within the first 6 months. (11) In cleft lip and palate (CLP) populations, systematic reviews report comparable absolute horizontal relapse values of approximately 1.2 mm, though proportional relapse rates tend to be higher (approximately 20–40% of sagittal advancement) attributed to additional instability factors including scar tissue contracture, deficient skeletal foundation, and potential presence of pharyngeal flaps. (3,12–14)

Evidence regarding the impact of maxillary advancement magnitude on stability is inconsistent for both non-cleft and CLP populations. In one study of a non-cleft cohort (n=43), greater horizontal advancement and degree of inferior repositioning of the anterior maxilla were identified as significant risk factors for relapse. (11) Multiple studies of patients with CLP reported stable results across small (<5 mm), medium (5–10 mm), and large (≥10 mm) advancement groups, with no significant correlation between advancement distance and relapse. (3,15) In a mixed cleft/non-cleft cohort, both advancement magnitude and a history of cleft were positively associated with horizontal relapse, though the authors proposed that relapse may be reduced by bone grafting. (16) Despite its general stability, the utility of conventional Le Fort I advancement is limited in growing patients, and its predictability remains inconsistent for more severe maxillary deficiencies, particularly in the cleft population. (12,13)

Maxillary distraction osteogenesis (MXDO) utilizing a rigid external device (RED) was introduced in 1997 as an alternative to conventional Le Fort I osteotomy. (17) Distraction osteogenesis is the process of generating new bone within a gap that forms between two bony segments following a surgical cut traversed by a mechanical mechanism to provide gradual displacement. MXDO involves a Le Fort level maxillary osteotomy and placement of a distractor, followed by a short latency period to allow initial healing, and an active distraction of about 1 mm advancement per day until desired advancement is achieved. (18) The distractor device applies incremental force across the osteotomy gap and can be placed internally or externally. The external distractor, illustrated in Figure 1, is easier to place and allows multi-vector control during midface advancement, but is larger, visible, and less acceptable to patients. The internal distractor is smaller, less visible, and better accepted by patients but is more difficult to place and remove and has uni-vector control. (21) Turning the distraction screws of either system advances the maxilla in a controlled, gradual manner. A meta-analysis of 361 patients with CLP reported that external distractors achieved greater midface advancement but with higher relapse rates (up to 30%) compared to internal distractors (0 – 15% relapse), particularly in the vertical plane. (19)

Figure 1. Rigid external distractor (20)

MXDO can reliably achieve large advancements averaging 8mm to 15mm and the procedure can be performed in skeletally immature patients. (8,21) The incremental advancement allows gradual soft tissue adaptation, which has been proposed to minimize velopharyngeal disruption. (8,18,21,22) The evidence regarding the impact of MXDO on velopharyngeal function is conflicting, as there is evidence of increased risk of VPI in adolescents with cleft palate following MXDO relative to conventional orthognathic surgery. (23) MXDO is associated with greater patient morbidity and discomfort than conventional orthognathic surgery. MXDO requires longer hospital stays, more than twice as many post-operative visits, strict adherence to activation protocols, and patients need to wear the distractor device for several months, significantly impacting daily life. (24) The quality and long-term stability of functional and esthetic outcomes of MXDO may ultimately determine whether the significant investment of time, resources, and emotional burden is justified for the patient.

Evidence regarding the stability of MXDO comes from retrospective case series supplemented by a few systematic reviews and meta-analyses. Table 1 summarizes the design and findings from some of the key studies. Reported advancements achieved by MXDO from retrospective studies range from 10mm to over 22mm, with mean sagittal relapse ranging between 6-26%. (25–29) Systematic reviews similarly estimate average relapse of about 12-25%, although individual studies within these reviews report a wider range reflecting heterogeneity in patient characteristics, distraction protocol, and retention strategies. (10,13) Results from studies that stratify patients by growth status suggest greater and more variable relapse in growing patients, sometimes exceeding 50% relapse, compared to non-growing patients who show more mild to moderate relapse rates. (28,30) Most studies are limited by small sample sizes, heterogeneous samples, mixed osteotomy levels, and short or inconsistent follow-up. In addition, few studies have evaluated the impact of maxillary distraction on facial esthetics or occlusal outcomes. The available evidence suggests that MXDO produces meaningful improvements in facial esthetics, including nasal projection, improved upper lip prominence, correction of facial concavity, correction of Class III malocclusion to Class I relationships, and resolution of anterior crossbite. (31–33) Overall, current literature supports MXDO as a reliable method to correct maxillary hypoplasia but highlights the need for standardized protocols and stronger long-term, growth stratified studies to better understand patterns of relapse.

Table 1. Design and Key Outcomes of Peer-Reviewed Studies Evaluating MXDO for Cleft-Related Maxillary Hypoplasia

Study	Study Design	Population	Growth Status	N	Distraction Device	Osteotomy Level	Follow-up	Sagittal Relapse (%)
Nimbalkar et al. (2022)	SR and MA	CLP	Growing (7–15.9 y) and non-growing (13–40 y)	6–13 per study	RED (external)	Le Fort I	1–5 y	4–38% (NG); 23.5–54.4% (G)
Jiang et al. (2020)	SR and MA	CLP	Mixed	5–58 per study	Internal and external	Le Fort I	NR	12%
Rachmiel et al. (2020)	Retrospective case series	CLP	Non-growing (16–29 y)	42	Internal	Le Fort I	2 y	6%
Meazzini et al. (2015)	Retrospective cohort	CLP	Growing (7–12 y) and non-growing (17–44 y)	22	RED (external)	Le Fort I	5–13 y	16–26% (G); stable (NG)
Saltaji et al. (2012)	SR	CLP	Mixed (10.5–26 y)	6–22 per study	Internal and external	Le Fort I	1–6.5 y	8–25%
Kanno et al. (2008)	Retrospective case series	CLP and non-CLP	Non-growing (15.3–33.4 y)	19	Internal and external	Le Fort I	2–4.8 y	8%
Cho & Kyung (2006)	Retrospective case series	CLP	Growing/adolescent (13–19 y)	9	RED (external) + facemask	Le Fort I	1–6 y	23%
Gao et al. (2022)	Prospective case series	CLP	Growing (11–12.5 y)	9	RED (external)	Le Fort I	1 y	12.8%

Studies are listed in reverse chronological order. Relapse is reported as percentage of sagittal advancement lost at the latest follow-up.

Abbreviations: CLP, cleft lip and/or palate; G, growing; MA, meta-analysis; NG, non-growing; NR, not reported; RED, rigid external distractor; SR, systematic review; y, years.

PURPOSE OF THE STUDY

The purpose of the study was to evaluate the long-term stability of skeletal, dental, and facial esthetic outcomes of maxillary distraction osteogenesis with a rigid external distraction device (MXDO-RED) in a cohort of patients with cleft lip and palate.

LIST OF SPECIFIC AIMS

Aim #1: Evaluate the longitudinal stability of skeletal, dental, and facial esthetic outcomes in patients who underwent MXDO-RED

Aim #2: Evaluate the association between the stability of skeletal, dental, and facial esthetic outcomes of MXDO-RED and the following factors:

(1) Patient age at the time of MXDO-RED

(2) Magnitude of maxillary advancement achieved

(3) Magnitude of mandibular growth

(4) Direction of mandibular growth

METHODS

Subjects

Sixty-eight subjects with severe maxillary hypoplasia associated with cleft lip and palate (CLP) were identified for inclusion in this study. All were consecutively treated with MXDO-RED at Seattle Children's Hospital from 2008 to 2023.

Subjects were included in the study if they met the following criteria:

(1) Diagnosis of severe maxillary hypoplasia associated with CLP

(2) Treated with Le Fort I level MXDO-RED at Seattle Children's Hospital from 2008 to 2023,

(3) High-quality diagnostic records available at the time points of interest.

Subjects were excluded from the study for any of the following criteria:

(1) Treated with Le Fort II, Le Fort III, or monobloc level MXDO-RED,

(2) Use of an internal distractor device,

(3) Concurrent mandibular osteotomy,

(4) Insufficient records at the time points of interest.

Data Collection

Data were extracted from clinical records from the Seattle Children's Hospital Craniofacial Center. Records included the Electronic Health Record (EHR), extraoral/intraoral photographs, dental models and/or digital intraoral scans, and lateral cephalometric radiographs or those extracted from full-head computerized tomography (CT) or cone beam computerized tomography (CBCT) scans. Extraoral/intraoral photos, digital intraoral scans, and lateral cephalometric radiographs were imported into Dolphin Imaging Software for analysis. All data were anonymized and entered into REDCap, a secure, web-based database for management and analysis of electronic health record information. (34,35)

Demographic variables included date of birth, sex, race/ethnicity, and craniofacial diagnoses. Operative variables included age at MXDO-RED, the type of Le Fort osteotomy, the surgeon, and the dates and duration of active distraction and consolidation phases. The EHR were reviewed for any distraction-related complications and the requirement for any subsequent orthognathic surgeries during the follow-up period.

Lateral cephalograms were digitized for selected skeletal, dental, and soft tissue landmarks (Appendix I, II). A cartesian grid was established: the x-axis was constructed by drawing a line through the nasion 7 degrees downward from the Sella-nasion line, and the y-axis was drawn through Sella and perpendicular to the x-axis. Changes in the antero-posterior and vertical position of A-point were measured in millimeters. The effects of MXDO-RED on the position of the maxilla were assessed as changes in the location of A-point within the x-y coordinates. For subjects with CT or CBCT, a lateral cephalogram was reconstructed from CT or CBCT and traced.

Patient records, including lateral cephalograms, dental models, and clinical photographs, were evaluated at four time points: immediately pre-MXDO-RED (T0), immediately post-MXDO-RED (T1), first follow-up (T2), and further available follow-up (T3). T0 data were derived from records immediately preceding start of active distraction. T1 data were derived from the earliest post-osteotomy records, prioritizing lateral cephalograms taken at the end of active distraction. If a lateral cephalogram at the end of active distraction was not available, lateral cephalograms from the end of consolidation was used. In cases where extraoral photographs were not available, soft tissue data were extracted from lateral cephalograms. In cases without dental models or scans, overjet and overbite data were extracted from lateral cephalograms. All data points, summarized in Appendix III, were recorded into REDCap.

Data Analysis

Intra-examiner reliability was calculated based on the repeated measurements of 10 subjects at two time points (T0, T1). For the reliability at each time point, the measurements were combined (i.e., 20 observations for each measurement), and for the reliability of the change, the difference between the two time points was used (i.e., 10 observations for each measurement). To compare the two measurements for intra-examiner reliability, the mean and standard deviation (SD) were computed for each set of measurements. The mean (SD) of the differences, 95% confidence interval (CI) for the mean difference, the intraclass correlation coefficient (ICC), and 95% CI for the ICC were computed to describe the (relative) reliability. (36) An ICC > 0.75 generally indicates excellent agreement (i.e., the variation due to the different measurements is relatively small compared to the variation between cases), 0.40 to 0.75 is fair to good agreement, and below 0.40 is poor agreement. Dahlberg's error and the minimum and maximum for the absolute value of the difference between the two measurements were computed to describe the absolute measurement error. (37)

To evaluate Aim #1, horizontal and vertical stability of the maxilla was assessed via the average change over time in the position of A-point relative to the X and Y axes from immediately post-distraction (T1). To account for participants having two follow-ups, linear regression using generalized estimating equations (GEE) methodology was used to test if the average change was zero versus not zero. (38) For clinical interpretation, relapse was also expressed as a percentage based on the average surgical advancement (T1 – T0). Comparisons were performed separately for follow-up of 0.5 to 3 years after post-distraction (short-term follow-up) and follow-up of >3 to 6.5 years (long-term follow-up). The same methods were used to evaluate changes in overjet, overbite, profile angle, and lip positions. For crossbite, a categorical outcome, short-term and long-term outcomes were compared to T0 using McNemar's test or GEE logistic regression, depending on whether participants had one or two follow-ups.

To evaluate Aim #2, GEE linear regression was used to test the association between horizontal and vertical A-point change and the following variables:

(1) Patient age: Evaluated as age at the start of distraction.
(2) Magnitude of advancement: Analyzed by correlating the surgical change (T1-T0) with the follow-up change (Follow-up – T1).
(3) Mandibular growth: Measured via changes in Gonion to Menton distances (Follow-up – T1).
(4) Mandibular plane angle: Evaluated using the SN-MP angle at T0

All regression models were adjusted for follow-up time to ensure the results accounted for the variable duration between T1 and follow-up. Statistical significance for all tests was set at $P < 0.05$ and all statistical analyses were performed using R software version 4.5.2.

RESULTS

Sample

A total of 68 potential subjects with cleft lip and palate consecutively treated with MXDO-RED at Seattle Children's Hospital from 2008 to 2023 were screened. Eight subjects were excluded for the following reasons: three had Le Fort III distraction, two utilized internal distractors, two lacked follow-up records, and one had diagnosis of craniosynostosis in addition to CLP. Sixty subjects were included in the final analysis.

Subject Characteristics and Follow-up Duration

Most subjects were male (71.7%). Race was most frequently White or Caucasian (40.0%) or Asian (36.7%). (Table 2) All participants had a diagnosis of unilateral or bilateral cleft lip and/or palate.

The short-term follow-up (>0.5 to 3 years post T1) consisted of 43 subjects. The mean age of subjects in the short-term group was 14.9±1.7 years at T0, 15.2±1.7 at T1, and 17±1.9 at the latest follow-up. (Table 3) The average duration of the follow-up period for the short-term group was 1.6± 0.7 years. (Table 3)

The long-term follow-up (>3 to 6.5 years post T1) consisted of 22 subjects. The mean age of subjects in the long-term group was 14.8+ 1.6 years at T0, 15.1± 1.6 years at T1, and 19.6±1.8 years at the latest follow-up. (Table 4) The average duration of the follow-up period for the long-term group was 4.5±1.0 years post distraction. (Table 4) The longest follow-up time at T3, 11.5 years, was omitted for analysis to maintain consistency.

Table 2. Demographic Characteristics of Subjects Receiving MXDO-RED (N = 60)

Characteristic	N = 60
Sex, n (%)	
Male	43 (71.7%)
Female	17 (28.3%)
Race/Ethnicity, n (%)	
White or Caucasian	24 (40.0%)
Asian	22 (36.7%)
Hispanic or Latino	8 (13.3%)
Other	3 (5.0%)
African American	2 (3.3%)
American Indian or Alaska Native	1 (1.7%)
Native Hawaiian or Other Pacific Islander	0 (0.0%)

Table 3. Age and Follow-up Duration of Short-term Follow-up Group Subjects with Cleft Maxillary Hypoplasia Receiving MXDO-RED at Study Time Points

Characteristic	Value
Age	
Age at Pre-Distraction (T0), years (N = 43)	
Mean (SD)	14.9 (1.7)
Median (IQR)	14.7 (14.0, 16.0)
Min to Max	10.5 to 18.6
Age at Post-MDO (T1), years (N = 43)	
Mean (SD)	15.2 (1.7)
Median (IQR)	15.0 (14.3, 16.2)
Min to Max	11.0 to 18.8
Age at Follow-Up, years (N = 55)	
Mean (SD)	17.0 (1.9)
Median (IQR)	17.1 (15.8, 18.1)
Min to Max	13.0 to 20.8
Treatment and Follow-Up Time	
Time from T0 to T1, years (N = 43)	
Mean (SD)	0.3 (0.4)
Median (IQR)	0.2 (0.1, 0.4)
Min to Max	0.1 to 2.5
Time from T0 to T1, months (N = 43)	
Mean (SD)	4.0 (4.6)
Median (IQR)	2.4 (1.2, 4.8)
Min to Max	1.2 to 30.0
Time from T1 to Follow-Up, years (N = 55)	
Mean (SD)	1.6 (0.7)
Median (IQR)	1.4 (1.1, 2.1)
Min to Max	0.5 to 3.0

Abbreviations: *SD = standard deviation; IQR = interquartile range (25th, 75th percentiles); MDO = Maxillary distraction osteogenesis; T0 = pre-distraction; T1 = post-MDO.*

Table 4. Age and Follow-up Duration of Long-term Follow-up Group Subjects with Cleft Maxillary Hypoplasia Receiving MXDO-RED at Study Time Points

Characteristic	Value
Age	
Age at Pre-Distraction (T0), years (N = 22)	
Mean (SD)	14.8 (1.6)
Median (IQR)	14.5 (14.0, 15.4)
Min to Max	12.0 to 18.6
Age at Post-MDO (T1), years (N = 22)	
Mean (SD)	15.1 (1.6)
Median (IQR)	14.8 (14.3, 16.0)
Min to Max	12.1 to 18.8
Age at Follow-Up, years (N = 23)	
Mean (SD)	19.6 (1.8)
Median (IQR)	19.3 (18.4, 21.2)
Min to Max	15.5 to 23.1
Treatment and Follow-Up Time	
Time from T0 to T1, years (N = 22)	
Mean (SD)	0.3 (0.2)
Median (IQR)	0.3 (0.1, 0.3)
Min to Max	0.1 to 0.7
Time from T0 to T1, months (N = 22)	
Mean (SD)	3.2 (2.0)
Median (IQR)	3.0 (1.2, 3.6)
Min to Max	1.2 to 8.4
Time from T1 to Follow-Up, years (N = 23)	
Mean (SD)	4.5 (1.0)
Median (IQR)	4.6 (3.6, 5.7)
Min to Max	3.1 to 6.5

Abbreviations: *SD = standard deviation; IQR = interquartile range (25th, 75th percentiles); MDO = Maxillary distraction osteogenesis; T0 = pre-distraction; T1 = post-MDO.*

Surgery Data

Every subject underwent Le Fort I level MXDO-RED: 40 by surgeon A (66.7%), 15 by surgeon B (25.0%), and five by surgeon C (8.3%). The active distraction phase averaged 24.2 days, with most subjects distracting at a rate of 1 mm/day. (Appendix IV) For 69.8% of subjects, there were no significant complications. In cases where issues arose, complications were predominantly mechanical in nature, involving adjustments to the external distractor to optimize the distraction vector. (Appendix V)

During the study period, further orthognathic surgery to address residual skeletal malocclusion was performed for 11 subjects and was planned for an additional 18 subjects. Surgeries were: five Le Fort I advancement with bilateral sagittal split osteotomy (BSSO), four Le Fort I advancement, one BSSO set-back, and one repeat MXDO-RED. Twenty-eight subjects did not need further surgery, and the need for further surgery was unknown for three subjects.

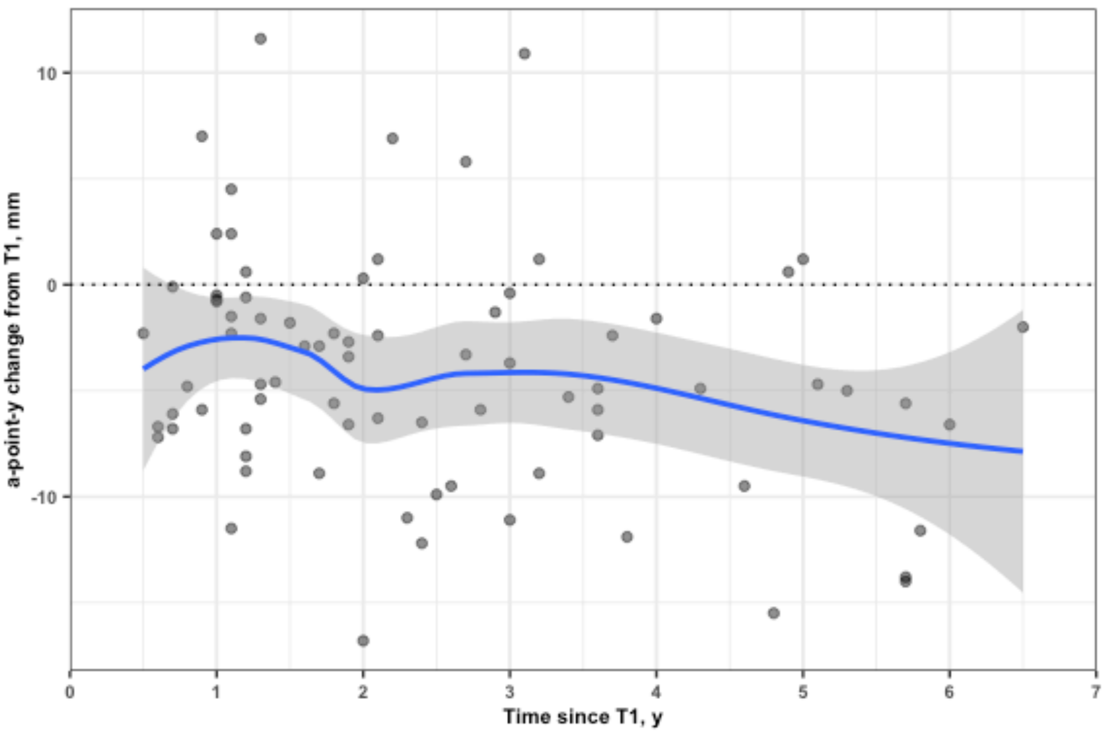
Intra-examiner Reliability

Based on the ICC, the intra-examiner reliability for T0 and T1 were excellent (ICC = 0.88 to 1.00). (Appendix VI) Dahlberg's error (DE) was small for all variables. Intra-examiner reliability for T0 and T1 change was also excellent (ICC = 1.00). (Appendix VII) Dahlberg's error was small for most variables, except possible measurement errors for profile angle are moderate (DE = 5.4). ICC and Dahlberg's error calculations for T0, T1, and T0 to T1 change are shown in Appendix VI and VII. Percent agreement and Kappa statistic for categorical variable were 100% and 1, respectively.

Skeletal Outcomes

Longitudinal analysis using generalized estimating equations (GEE) demonstrated a statistically significant posterior repositioning (relapse) of A-point relative to the Y-axis over the post-distraction follow-up period, with a slope of −0.9 mm per year (95% CI: −1.6 to −0.2; p = .011). (Figure 2)

Figure 2. Sagittal Stability of Advanced Maxilla Following MXDO-RED in Subjects with Cleft Maxillary Hypoplasia, Measured as Change in A-point Distance from the Y-axis over the Post-Distraction Follow-up Period



Each point represents one follow-up observation. The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model, which accounts for within-subject correlation due to repeated observations. The slope of −0.9 mm per year (95% CI: −1.6 to −0.2; p = .011; r = −0.26) indicates a statistically significant posterior repositioning (relapse) of A-point relative to its immediate post-distraction position (T1). Negative values on the Y-axis denote posterior movement (relapse); positive values denote continued anterior positioning relative to T1.

In the short-term follow-up group (N = 55; 91.7%), the mean sagittal advancement from T0 to T1 was +12.6±5.4 mm. A-point relapsed posteriorly by a mean of 3.5 mm±5.2 (95% CI: −4.9 to −2.1; $p < .001$), representing 27.8% of the initial advancement. (Table 5) Approximately 42% of subjects experienced sagittal relapse exceeding 5 mm, and an additional 22% experienced relapse of 2 to 5 mm. (Table 6)

In the long-term follow-up group (N = 23; 38.3%), the mean initial advancement was 12.9 ±6.2mm, which decreased by 5.5±6.0mm (95% CI: −7.9 to −3.1; $p < .001$) over the follow-up period, corresponding to 42.6% of the surgical advancement. (Table 5) At the longest follow-up, 52% of subjects showed relapse exceeding 5 mm, and 26% showed relapse of 2 to 5 mm. (Table 6)

GEE linear regression revealed no significant change in the vertical position of A-point (distance from the X-axis) over the post-distraction follow-up period ($p = .850$). (Appendix VIII)

Table 5. Sagittal and Vertical Changes in A-point Position Following MXDO-RED in Subjects with Cleft Maxillary Hypoplasia: Surgical Advancement (T0–T1) and Post-distraction Relapse for Short-term (0.5–3 Years, N = 55) and Long-term (>3–6.5 Years, N = 23) Follow-up Groups

Characteristic	Surgical Advancement (T0–T1)	Follow-up Change from T1	95% CI	p-value
Short-term Follow-up (0.5–3 years post-distraction; N = 55)				
Sagittal: A-point from Y-axis (mm)				
Mean (SD)	12.6 (5.4)	−3.5 (5.2)	-4.9, -2.1	<0.001
Median (IQR)	10.8 (9.1, 16.9)	−3.3 (−6.7, −0.6)		
Min to Max	3.1, 27.3	−16.8, 11.6		
Vertical: A-point from X-axis (mm)				
Mean (SD)	1.7 (4.5)	−0.7 (5.2)	−2.1, 0.6	0.280
Median (IQR)	0.7 (−1.9, 4.3)	1.0 (-3.8, 2.9)		
Min to Max	−7.9, 14.3	-12.7, 7.9		
Long-term Follow-up (>3–6.5 years post-distraction; N = 23)				
Sagittal: A-point from Y-axis (mm)				
Mean (SD)	12.9 (6.2)	−5.5 (6.0)	−7.9, −3.1	<0.001
Median (IQR)	12.9 (6.5, 17.6)	−5.3 (−9.5, −2.0)		
Min to Max	3.1, 27.3	−15.5, 10.9		
Vertical: A-point from X-axis (mm)				
Mean (SD)	3.7 (4.9)	−1.4 (5.1)	−3.4, 0.7	0.198
Median (IQR)	2.4 (0.2, 7.2)	−0.5 (−5.5, 2.4)		
Min to Max	−2.5, 14.3	−10.2, 7.5		

Surgical advancement = T0–T1 change. Follow-up change = change from T1 to follow-up time point. Negative values indicate relapse (posterior or superior movement). p-values from generalized estimating equations (GEE) linear regression testing whether mean change from T1 differs significantly from zero. Abbreviations: SD, standard deviation; IQR, interquartile range; CI, confidence interval; T0, pre-distraction; T1, post-MXDO; T2, first follow-up; T3, last follow-up.

Table 6. Distribution of Sagittal and Vertical A-point Relapse Categories Following MXDO-RED in Subjects with Cleft Maxillary Hypoplasia, by Short-term (0.5–3 Years, N = 55) and Long-term (>3–6.5 Years, N = 23) Follow-up Periods

	Short-term Follow-up (0.5–3 y, N = 55)		Long-term Follow-up (>3–6.5 y, N = 23)	
Change Category (mm)	Sagittal (Y-axis)	Vertical (X-axis)	Sagittal (Y-axis)	Vertical (X-axis)
< −5 mm	23 (41.8%)	11 (20.0%)	12 (52.2%)	6 (26.1%)
−5 to −2 mm	12 (21.8%)	8 (14.5%)	6 (26.1%)	3 (13.0%)
> −2 to 0 mm	10 (18.2%)	16 (29.1%)	1 (4.3%)	7 (30.4%)
> 0 to < 2 mm	3 (5.5%)	0 (0.0%)	3 (13.0%)	0 (0.0%)
2 to 5 mm	3 (5.5%)	17 (30.9%)	0 (0.0%)	4 (17.4%)
> 5 mm	4 (7.3%)	3 (5.5%)	1 (4.3%)	3 (13.0%)

Values are n (%). Negative values indicate relapse (posterior movement in the sagittal plane; superior movement in the vertical plane). Sagittal change measured as A-point distance from the Y-axis; vertical change measured as A-point distance from the X-axis. Change is calculated from immediately post-distraction (T1) to the respective follow-up time point.

Abbreviations: MXDO-RED, maxillary distraction osteogenesis with rigid external distractor; T1, post-MXDO.

Dental Outcomes – Overjet and Overbite

Paired comparisons at discrete time points demonstrated significant relapse from the immediate post-distraction position. In the short-term group, overjet increased from a mean of -11.5±4.9 at T0 to a mean of 2.9±3.9 at T1 but subsequently decreased by 4.2±5.4 mm, resulting in a final mean overjet of -1.2±5.5mm ($p < .001$). In the long-term group, overjet increased from a mean of -10.4±4.4mm at T0 to a mean of 1.9±3.7mm at T1, then decreased by 4.3±8.2 mm over the follow-up period, resulting in a final mean overjet of −2.3±6.1mm ($p = .010$). (Table 7) The overall GEE longitudinal regression revealed no significant continuous change in overjet over the post-distraction follow-up period ($p = .373$). (Appendix IX)

Overbite data from lateral cephalograms were available for 18 short-term and 10 long-term subjects. In the short-term group, overbite increased by a mean of 1.7±4.4 mm between T1 and T2 ($p=0.085$). In the long-term group, an increase of 3.8±4.1 mm was observed ($p = .002$), with mean overbite shifting from -1.3 ± 4.7mm at T1 to $+2.4 \pm 2.2$mm. (Table 7) GEE longitudinal regression showed no significant continuous change in overbite over the follow-up period ($p = .373$). (Appendix X)

Table 7. Overjet and Overbite at Each Study Time Point and Post-distraction Change Following MXDO-RED in Subjects with Cleft Maxillary Hypoplasia, by Short-term (0.5–3 Years) and Long-term (>3–6.5 Years) Follow-up Periods

Characteristic	Pre-distraction (T0)	Post-MXDO (T1)	Follow-up	Change (T1 to Follow-up)	95% CI	p-value
Short-term Follow-up (0.5–3 years post-distraction)						
Overjet, mm (N = 52)						
Mean (SD)	−11.5 (4.9)	2.9 (3.9)	−1.2 (5.5)	−4.2 (5.4)	−5.6, −2.7	<0.001
Median (IQR)	−11.0 (−14.7, −7.5)	3.0 (0.9, 5.4)	2.0 (−5.2, 2.7)	−3.6 (−7.8, 0.3)		
Min to Max	−21.8, −2.9	−10.0, 11.2	−17.6, 5.3	−26.0, 3.5		
Overbite, mm (N = 18)						
Mean (SD)	−0.4 (5.8)	−0.3 (2.2)	1.5 (3.2)	1.7 (4.4)	−0.2, 3.7	0.085
Median (IQR)	1.0 (−6.6, 4.9)	0.0 (−2.0, 0.0)	0.8 (−1.0, 1.3)	0.2 (−1.1, 3.5)		
Min to Max	−8.2, 9.0	−4.0, 4.4	−2.0, 8.0	−3.8, 11.7		
Long-term Follow-up (>3–6.5 years post-distraction)						
Overjet, mm (N = 23)						
Mean (SD)	−10.4 (4.4)	1.9 (3.7)	−2.3 (6.1)	−4.3 (8.2)	−7.5, −1.0	0.010
Median (IQR)	−10.0 (−12.4, −6.8)	2.4 (0.5, 5.0)	−3.1 (−6.2, 2.0)	−4.2 (−10.0, −1.8)		
Min to Max	−21.2, −2.9	−10.0, 6.0	−14.0, 14.7	−13.8, 24.7		
Overbite, mm (N = 10)						
Mean (SD)	1.2 (5.0)	−1.3 (4.7)	2.4 (2.2)	3.8 (4.1)	1.4, 6.2	0.002
Median (IQR)	0.1 (−0.4, 6.4)	0.0 (−1.4, 0.6)	2.8 (0.1, 3.8)	2.2 (1.7, 4.6)		
Min to Max	−7.4, 8.1	−14.0, 2.1	−0.3, 6.8	0.0, 14.1		

Change = follow-up value minus T1 value. Sample sizes differ between overjet and overbite due to data availability. p-values from generalized estimating equations (GEE) linear regression testing whether mean change from T1 differs significantly from zero. Abbreviations: MXDO-RED, maxillary distraction osteogenesis with rigid external distractor; SD, standard deviation; IQR, interquartile range; CI, confidence interval; T0, pre-distraction; T1, post-MXDO;

Dental Outcome - Crossbite

Crossbite correction was the most significant dental improvement observed following MXDO-RED. At T0, 100% of subjects presented with some form of crossbite, with complete crossbite being the most prevalent (70.9% short-term group, 73.9% long-term group). T1 crossbite data were available for 17 of 55 subjects (30.9%) in the short-term group and 7 of 23 (30.4%) in the long-term group; among those with available data, 70.6% and 71.4% had no crossbite immediately post-distraction, respectively. In the paired short-term analysis ($N = 47$), the proportion of subjects free of any crossbite increased from 0.0% at T0 to 36.2% at follow-up ($p < .001$, GEE logistic regression), while complete crossbite decreased from 72.3% to 27.7%. Although partial relapse was observed, improvement remained statistically significant in the long-term paired analysis ($N = 21$), with 23.8% of subjects maintaining no crossbite and complete crossbite decreasing from 76.2% to 47.6% ($p < .001$). (Table 8)

Table 8. Dental Crossbite Status at Each Study Time Point and Paired Comparisons Following MXDO-RED in Subjects with Cleft Maxillary Hypoplasia, by Short-term (0.5–3 Years) and Long-term (>3–6.5 Years) Follow-up Periods

Panel A. Crossbite Prevalence by Type at Each Time Point (All Available Data)

Crossbite Type	Pre-distraction (T0)	Post-MXDO (T1)	Follow-up
Short-term Group (0.5–3 years post-distraction)			
Available n / Total N	**55 / 55**	**17 / 55**	**47 / 55**
None	0 (0.0%)	12 (70.6%)	17 (36.2%)
Anterior	7 (12.7%)	1 (5.9%)	6 (12.8%)
Unilateral posterior	2 (3.6%)	3 (17.6%)	4 (8.5%)
Bilateral posterior	1 (1.8%)	0 (0.0%)	2 (4.3%)
Anterior & unilateral posterior	4 (7.3%)	0 (0.0%)	5 (10.6%)
Anterior & bilateral posterior	2 (3.6%)	0 (0.0%)	0 (0.0%)
Complete	39 (70.9%)	1 (5.9%)	13 (27.7%)
Long-term Group (>3–6.5 years post-distraction)			
Available n / Total N	**23 / 23**	**7 / 23**	**21 / 23**
None	0 (0.0%)	5 (71.4%)	5 (23.8%)
Anterior	4 (17.4%)	2 (28.6%)	3 (14.3%)
Unilateral posterior	0 (0.0%)	0 (0.0%)	0 (0.0%)
Bilateral posterior	1 (4.3%)	0 (0.0%)	1 (4.8%)
Anterior & unilateral posterior	1 (4.3%)	0 (0.0%)	2 (9.5%)
Anterior & bilateral posterior	0 (0.0%)	0 (0.0%)	0 (0.0%)
Complete	17 (73.9%)	0 (0.0%)	10 (47.6%)

Panel B. Paired Comparison of Crossbite Presence (Restricted to Subjects with Data at Both T0 and Follow-up; None vs. Any Crossbite)

		No Crossbite, n (%)		Any Crossbite, n (%)		
Follow-up Group	**Paired N**	**T0**	**Follow-up**	**T0**	**Follow-up**	**p-value**
Short-term	47	0 (0.0%)	17 (36.2%)	47 (100%)	30 (63.8%)	<.001
Long-term	21	0 (0.0%)	5 (23.8%)	21 (100%)	16 (76.2%)	<.001

Panel A reports all subjects with available crossbite data at each time point; percentages based on available (non-missing) observations. T1 data were available for only 17 of 55 subjects (30.9%) in the short-term group and 7 of 23 (30.4%) in the long-term group. Panel B dichotomizes crossbite as none versus any type and restricts to subjects with data at both T0 and follow-up. p-values from GEE logistic regression.

Abbreviations: MXDO-RED, maxillary distraction osteogenesis with rigid external distractor; GEE, generalized estimating equations; Ant, anterior; Uni, unilateral; Bi, bilateral; Post, posterior; T0, pre-distraction; T1, post-MXDO.

Facial Esthetic Outcomes

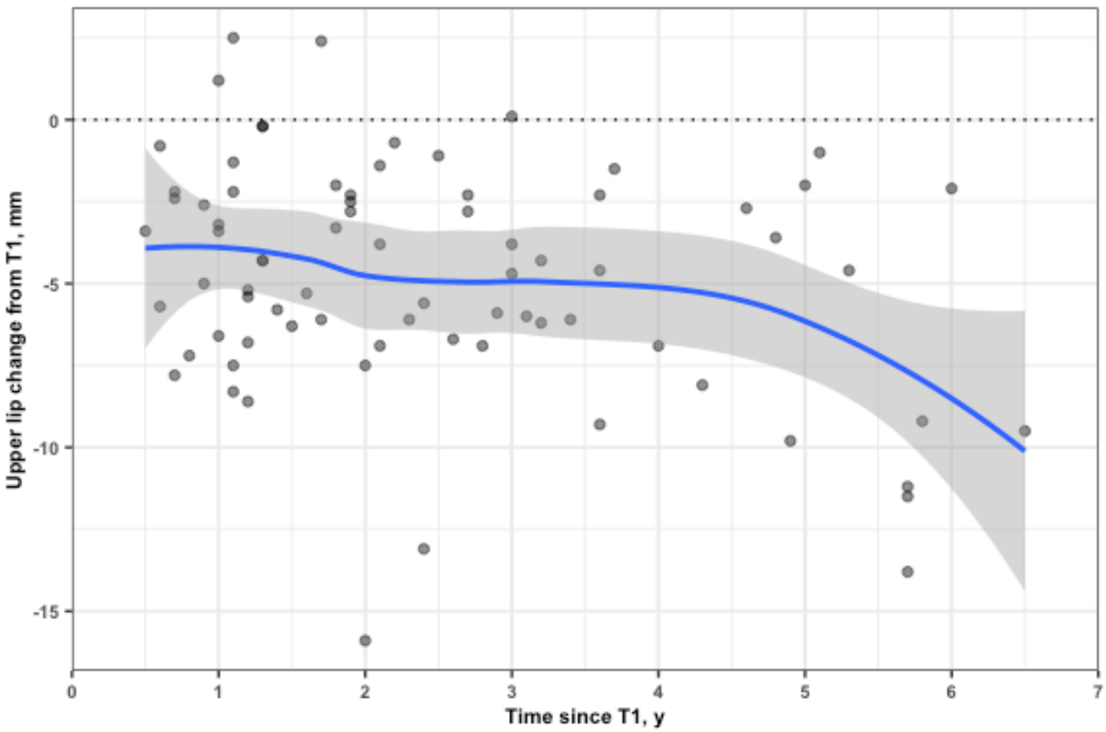
From T0 to T1, all facial esthetic measurements showed significant improvement. Profile angle decreased by a mean of 21.4±10.2 degrees in the short-term group ($N = 52$) and 20.0±9.8 degrees in the long-term group ($N = 22$), reflecting a transition from a concave to a convex facial profile. Upper lip position advanced by a mean of 9.2±4.0mm and 9.6±3.8mm, respectively, while lower lip position showed more modest advancement of 1.3±3.3 mm and 1.5±2.5 mm relative to the E-line. (Table 9)

Over the follow-up period, profile angle showed significant relapse toward a straighter profile in both groups: short-term (mean increase of 11.4±7.2°; 95% CI: 9.4, 13.2; $p < .001$) and long-term (mean increase of 13.9±8.0°; 95% CI: 10.6, 17.1; $p < .001$) groups. The GEE linear regression across the entire follow-up period did not reveal a statistically significant continuous trend (slope = 1.0°/year; $p = 0.110$). (Appendix XI)

Upper lip position was the only soft tissue measure to demonstrate a significant longitudinal trend. GEE linear regression revealed a significant progressive retrusion over time (slope = −0.7 mm/year; $p = 0.011$). (Figure 3) In the short-term group, the upper lip retruded by a mean of 4.3±3.4 mm from the T1 position (95% CI: −5.2, −3.4; $p < .001$), and this continued in the long-term group with a mean retrusion of 6.2±3.7 mm (95% CI: −7.7, −4.7; $p < .001$). (Table 9)

Changes in lower lip position were smaller but statistically significant at both follow-up intervals. The lower lip retruded by a mean of 0.9±3.2 mm in the short-term group (95% CI: −1.7, −0.02; $p = .045$) and 1.4±2.7 mm in the long-term group (95% CI: −2.4, −0.3; $p = .014$). The GEE linear regression did not identify a significant continuous trend over time (slope = −0.2 mm/year; $p = 0.519$). (Table 9, Appendix XII)

Figure 3. Longitudinal Change in Upper Lip Position Relative to Ricketts' Esthetic Line (E-line) During the Post-Distraction Follow-up Period in Subjects with Cleft Maxillary Hypoplasia Following MXDO-RED (N = 54 Observations from 39 Subjects)



Each point represents one follow-up observation of upper lip positional change relative to Ricketts' esthetic line (E-line), measured as the perpendicular distance from the most anterior point of the upper lip to a line drawn from Pronasale to Soft tissue Pogonion, compared to the immediate post-distraction position (T1). The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation across repeated observations. The slope of −0.7 mm per year (95% CI: −1.2 to −0.1; p = .011; r = −0.30) indicates a statistically significant progressive retrusion of the upper lip from its advanced post-distraction position.

Table 9. Soft Tissue Changes in Profile Angle and Lip Position Following MXDO-RED in Subjects with Cleft Maxillary Hypoplasia: Surgical Change (T0–T1) and Post-distraction Relapse during Short-term (0.5–3 Years) and Long-term (>3–6.5 Years) Follow-up Periods

Characteristic	Surgical Change (T0–T1)	Follow-up Change from T1	95% CI	p-value
Short-term Follow-up (T2, 0.5–3 years post-distraction)				
Profile Angle, degrees (N = 52)				
Mean (SD)	−21.4 (10.2)	11.4 (7.2)	9.4, 13.2	<.001
Median (IQR)	−23.9 (−28.7, −16.4)	10.7 (6.5, 15.2)		
Min to Max	−45.0, 3.1	−1.5, 27.9		
Upper Lip Position Relative to E-line, mm (N = 54)				
Mean (SD)	9.2 (4.0)	−4.3 (3.4)	−5.2, −3.4	<.001
Median (IQR)	10.0 (6.2, 12.3)	−4.1 (−6.3, −2.2)		
Min to Max	−1.4, 16.1	−15.9, 2.5		
Lower Lip Position Relative to E-line, mm (N = 54)				
Mean (SD)	1.3 (3.3)	−0.9 (3.2)	−1.7, −0.02	.045
Median (IQR)	1.6 (−0.3, 3.3)	−0.5 (−2.9, 1.2)		
Min to Max	−6.0, 9.2	−8.2, 5.5		
Long-term Follow-up (T3, >3–6.5 years post-distraction)				
Profile Angle, degrees (N = 22)				
Mean (SD)	−20.0 (9.8)	13.9 (8.0)	10.6, 17.1	<.001
Median (IQR)	−18.4 (−25.7, −13.0)	14.3 (10.2, 19.3)		
Min to Max	−45.0, −3.6	−6.1, 26.6		
Upper Lip Position Relative to E-line, mm (N = 22)				
Mean (SD)	9.6 (3.8)	−6.2 (3.7)	−7.7, −4.7	<.001
Median (IQR)	9.8 (6.1, 12.9)	−6.1 (−9.3, −2.7)		
Min to Max	3.3, 15.8	−13.8, −1.0		
Lower Lip Position Relative to E-line, mm (N = 22)				
Mean (SD)	1.5 (2.5)	−1.4 (2.7)	−2.4, −0.3	.014
Median (IQR)	2.2 (−0.3, 3.5)	−0.7 (−2.1, 0.0)		
Min to Max	−5.2, 5.7	−7.6, 2.8		

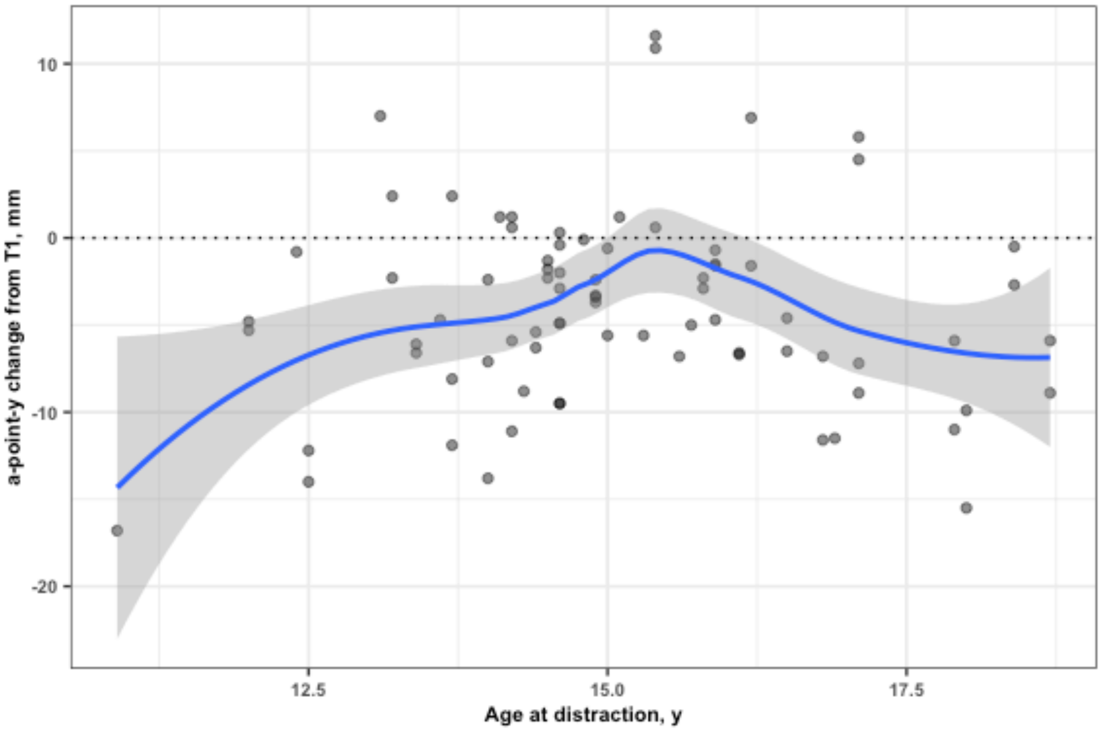
Surgical change = T0–T1 change. Follow-up change = change from T1 to follow-up time point. For profile angle: negative surgical change indicates transition from convex toward concave profile; positive follow-up change indicates relapse toward convexity. For lip positions: positive surgical change indicates advancement (protrusion) relative to E-line; negative follow-up change indicates retrusion from the advanced position. p-values from *generalized estimating equations (GEE) linear regression testing whether mean change from T1 differs significantly from zero. Sample sizes differ slightly across variables due to data availability.*

Abbreviations: MXDO-RED, maxillary distraction osteogenesis with rigid external distractor; E-line, Ricketts' esthetic line (pronasale to soft tissue pogonion); SD, standard deviation; IQR, interquartile range; CI, confidence interval; T0, pre-distraction; T1, post-MXDO.

Age at Distraction

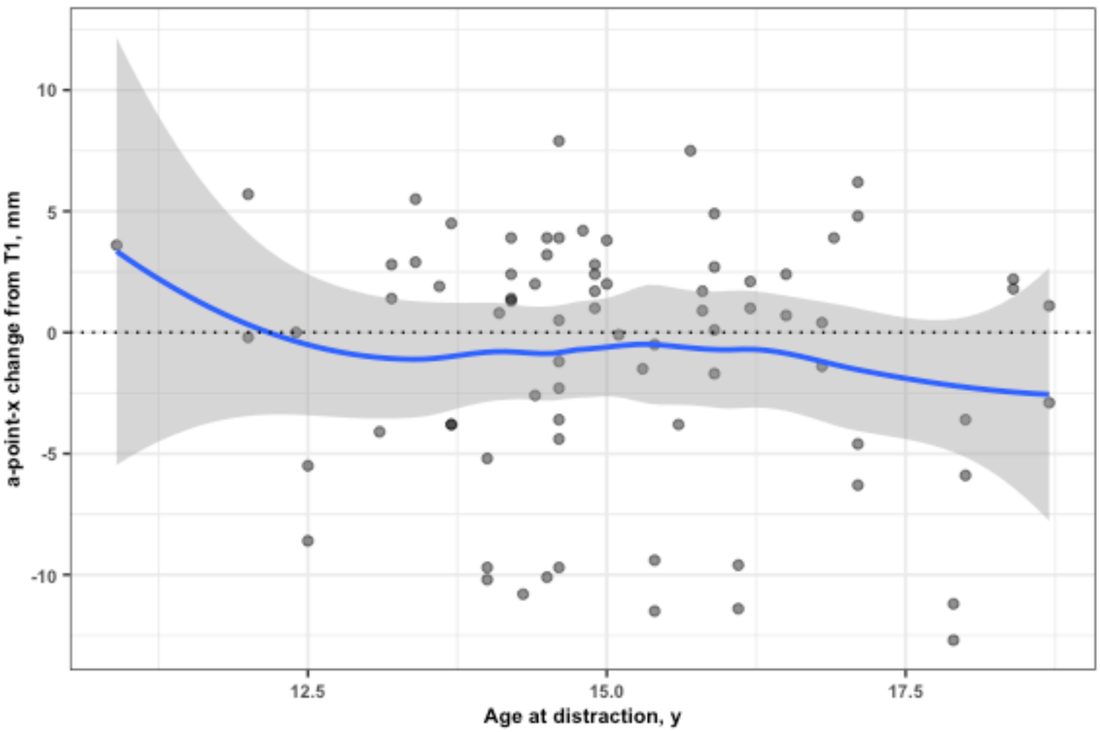
GEE linear regression adjusted for follow-up duration revealed no significant association between patient age at the time of MXDO-RED and subsequent sagittal change in A-point position (slope = 0.06 mm per year of age; 95% CI: −0.81 to 0.93; $p = .892$) or vertical change in A-point position (slope = −0.32 mm per year of age; 95% CI: −0.98 to 0.33; $p = .338$). (Figure 4, Figure 5) Across the age range in this cohort (10.9–18.7 years), skeletal stability following MXDO-RED was not significantly influenced by the patient's age at the time of MXDO-RED.

Figure 4. Association Between Age at Distraction and Sagittal Change in A-point Position During the Post-Distraction Follow-up Period Following MXDO-RED (N = 86 Observations from 60 Subjects)



Each point represents one follow-up observation of sagittal A-point change (distance from Y-axis) relative to the immediate post-distraction position (T1). The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation. In the unadjusted model, the slope was 0.06 mm per year of age (95% CI: −0.81 to 0.93; p = .892; r = 0.02), indicating no significant association between age at distraction and subsequent sagittal change in A-point. After adjusting for follow-up duration, the association remained non-significant (slope = -0.004 mm per year of age; 95% CI: −0.84 to 0.83; p = .992). Negative values on the Y-axis denote posterior repositioning (relapse); positive values denote continued anterior positioning relative to T1.

Figure 5. Association Between Age at Distraction and Vertical Change in A-point Position During the Post-Distraction Follow-up Period in Subjects with Cleft Maxillary Hypoplasia Following MXDO-RED (N = 86 Observations from 60 Subjects)

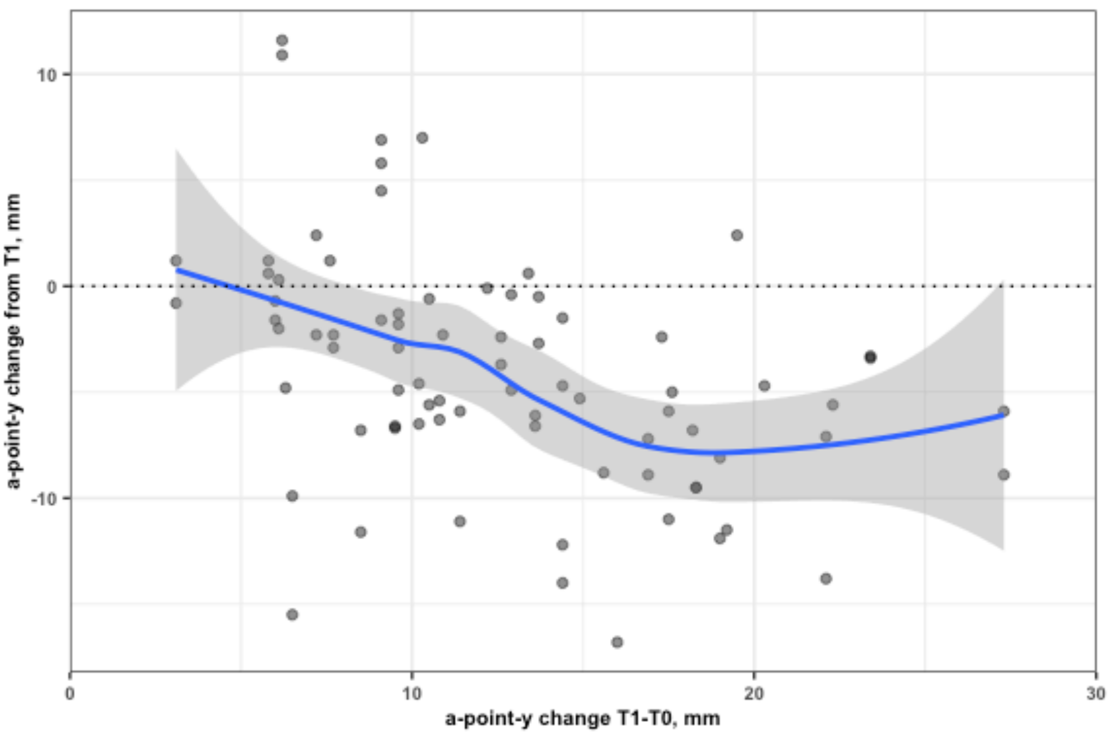


Each point represents one follow-up observation of vertical A-point change (distance from X-axis) relative to the immediate post-distraction position (T1). The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation. In the unadjusted model, the slope was −0.32 mm per year of age (95% CI: −0.98 to 0.33; p = .338; r = −0.10), indicating no significant association between age at distraction and subsequent vertical change in A-point. After adjusting for follow-up duration, the association remained non-significant (slope = −0.33 mm per year of age; 95% CI: −0.98 to 0.32; p = .332). Negative values on the Y-axis denote superior displacement (intrusion); positive values denote inferior displacement relative to T1.

Magnitude of Advancement

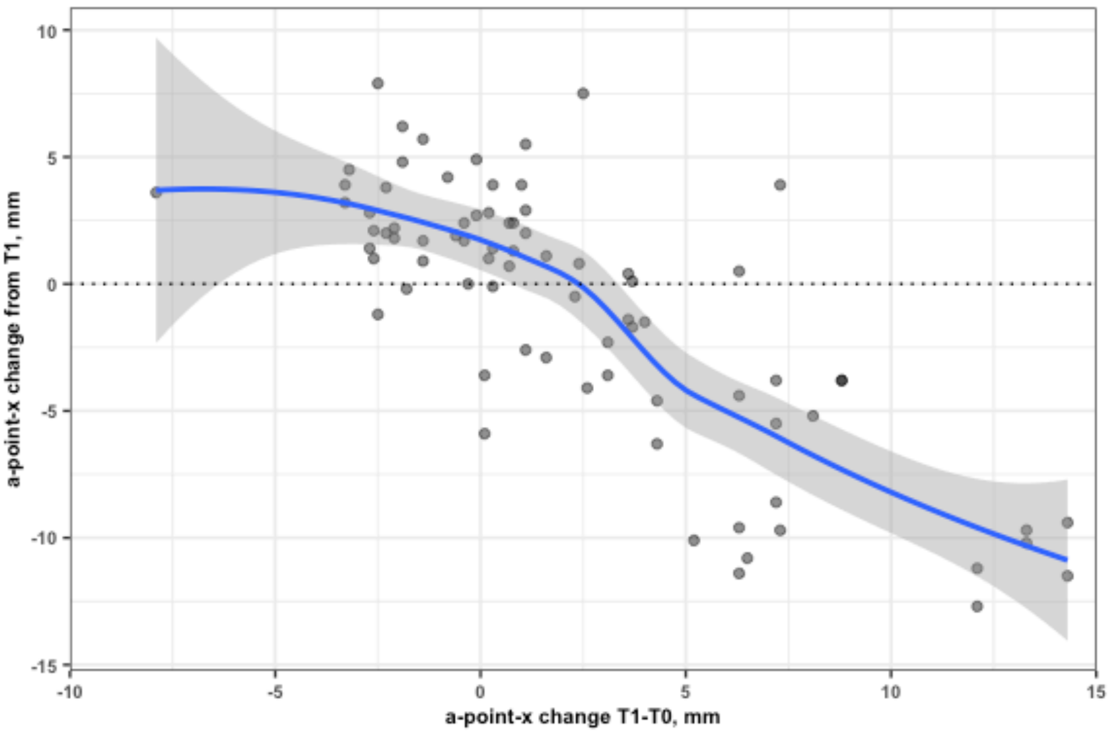
The magnitude of surgical advancement was the strongest predictor of post-distraction skeletal change. In the sagittal plane, GEE linear regression adjusted for follow-up duration demonstrated that each additional millimeter of advancement from T0 to T1 was associated with 0.42 mm of posterior relapse during the follow-up period (95% CI: −0.604 to −0.237; p < .001; r = −0.43). (Figure 6) The association was most pronounced in the vertical dimension, where each additional millimeter of vertical displacement from T0 to T1 was associated with 0.88 mm of vertical relapse (95% CI: −1.025 to −0.739; $p < .001$; $r = -0.79$), suggesting near-complete reversal of the surgical vertical change over time. (Figure 7) Both associations remained significant after adjusting for follow-up duration, indicating that the relationship between advancement magnitude and relapse was not attributable to longer observation periods capturing more change.

Figure 6. Association Between Magnitude of Surgical Advancement and Sagittal Change in A-point Position During the Post-Distraction Follow-up Period in Subjects with Cleft Maxillary Hypoplasia Following MXDO-RED (N = 86 Observations from 60 Subjects)



Each point represents one follow-up observation of sagittal A-point change (distance from Y-axis) relative to the immediate post-distraction position (T1), plotted against the magnitude of surgical advancement achieved from T0 to T1. The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation. In the unadjusted model, the slope was −0.426 mm of relapse per mm of advancement (95% CI: −0.620 to −0.232; $p < .001$; $r = -0.43$), indicating a significant negative association: greater surgical advancement was associated with greater subsequent posterior repositioning (relapse). After adjusting for follow-up duration, the association remained significant (slope = −0.420 mm per mm of advancement; 95% CI: −0.604 to −0.237; $p < .001$), suggesting that for each additional millimeter of sagittal advancement, approximately 42.0% is lost to relapse over the follow-up period. Negative values on the Y-axis denote posterior repositioning (relapse); positive values denote continued anterior positioning relative to T1.

Figure 7. Association Between Magnitude of Surgical Vertical Change and Vertical Change in A-point Position During the Post-Distraction Follow-up Period Following MXDO-RED (N = 86 Observations from 60 Subjects)



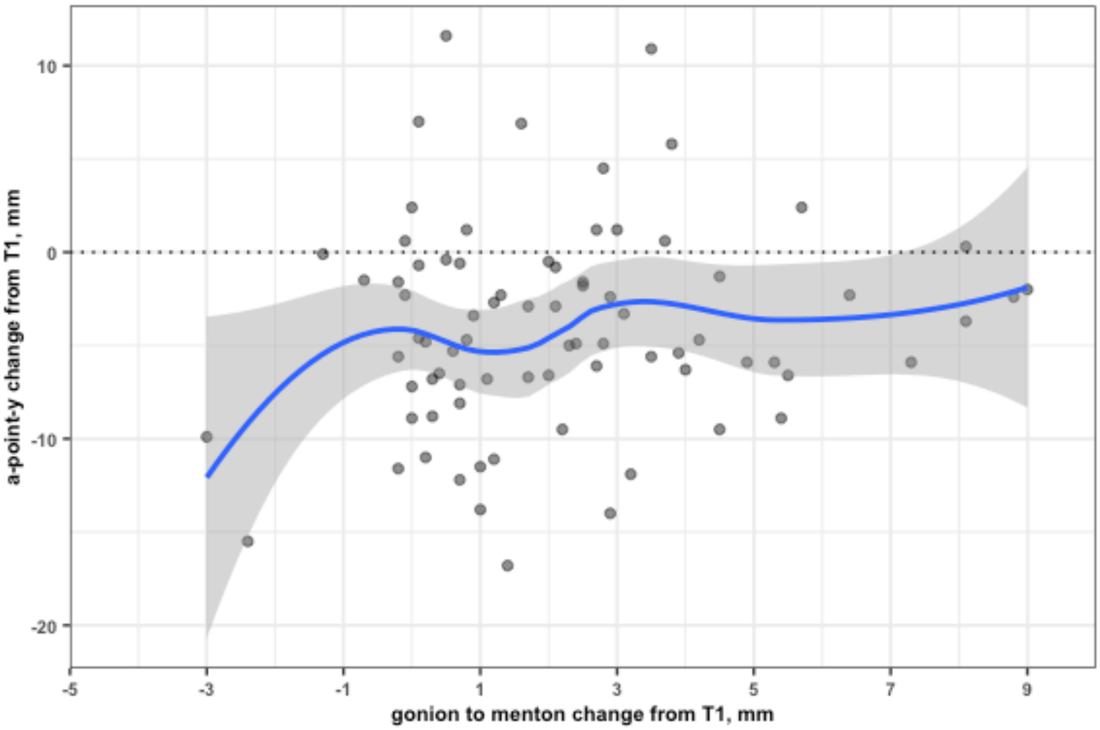
Each point represents one follow-up observation of vertical A-point change (distance from X-axis) relative to the immediate post-distraction position (T1), plotted against the magnitude of vertical displacement achieved from T0 to T1. The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation. After adjusting for follow-up duration, the association was found to be highly significant (slope = −0.881 mm per mm of initial displacement; 95% CI: −1.025 to −0.739; p < .001), suggesting near-complete reversal of the surgical vertical change over time. Negative values on the Y-axis denote superior movement (reversal of inferior displacement); positive values denote continued inferior positioning relative to T1. The correlation coefficient of −0.79 reflects a strong association, substantially exceeding the corresponding sagittal relationship (r = −0.43; Figure 6)

Mandibular Growth

Increase in the mandibular body length, measured via Gonion-Menton (Go-Me), was used to assess mandibular growth. In the short-term group, Go–Me increased by a mean of 1.8±2.3 mm from T1. In the long-term group, Go-Me increased by 3.0±2.7 mm. (Appendix XIII)

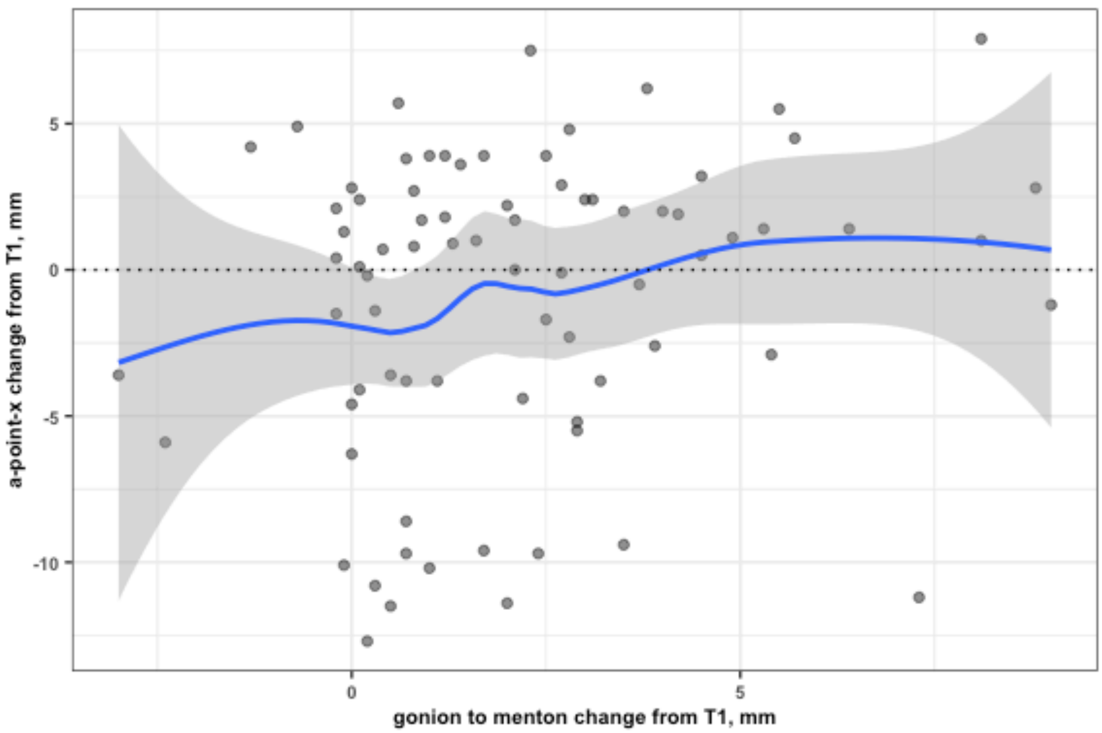
After adjusting for follow-up duration, Go–Me growth was significantly associated with changes in A-point position in both the sagittal plane (0.52 mm of anterior A-point repositioning per 1 mm of Go–Me growth; 95% CI: 0.16 to 0.90; p = .005) and the vertical plane (0.47 mm of inferior A-point displacement per 1 mm of Go–Me growth; 95% CI: 0.02 to 0.93; p = .041). (Figure 8, Figure 9) Neither association was significant in the unadjusted models (sagittal p = .121; vertical p = .061), suggesting that the relationship between mandibular body growth and A-point position was partially masked by confounding with follow-up duration. The emergence of significance after adjustment for follow-up duration suggests that mandibular growth and follow-up time are correlated confounders. Subjects with longer follow-up experienced both greater mandibular growth and greater anterior and inferior maxillary displacement, and once the independent effect of time was accounted for, the association between growth and A-point position became apparent.

Figure 8. Association Between Mandibular Body Growth (Gonion–Menton) and Sagittal Change in A-point Position During the Post-Distraction Follow-up Period Following MXDO-RED



Each point represents one follow-up observation of sagittal A-point change (distance from Y-axis) relative to the immediate post-distraction position (T1), plotted against mandibular body growth (Gonion–Menton change from T1 to follow-up). The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation. In the unadjusted model, the slope was 0.32 mm of sagittal A-point change per mm of mandibular body growth (95% CI: −0.08 to 0.72; p = .121; r = 0.14), suggesting a trend that did not reach statistical significance. After adjusting for follow-up duration, the association became significant (slope = 0.52 mm per mm of growth; 95% CI: 0.16 to 0.90; p = .005), indicating that each additional millimeter of mandibular body growth from T1 to follow-up was associated with 0.52 mm of anterior repositioning of A-point over the same interval. Positive values on the Y-axis denote anterior movement of A-point relative to T1; negative values denote posterior repositioning (relapse).

Figure 9. Association Between Mandibular Body Growth (Gonion–Menton) and Vertical Change in A-point Position During the Post-Distraction Follow-up Period Following MXDO-RED



Each point represents one follow-up observation of vertical A-point change (distance from X-axis) relative to the immediate post-distraction position (T1), plotted against mandibular body growth (Gonion–Menton change from T1 to follow-up). The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation. In the unadjusted model, the slope was 0.42 mm of vertical A-point change per mm of mandibular body growth (95% CI: −0.02 to 0.87; $p = .061$; $r = 0.20$), suggesting a trend that did not reach statistical significance. After adjusting for follow-up duration, the association became significant (slope = 0.47 mm per mm of growth; 95% CI: 0.02 to 0.93; $p = .041$), indicating that each additional millimeter of mandibular body growth from T1 to follow-up was associated with 0.47 mm of inferior displacement of A-point over the same interval. Positive values on the Y-axis denote inferior movement of A-point relative to T1; negative values denote superior displacement.

Mandibular Plane Angle

Mandibular plane angle at T0, was categorized as low (<27°, N = 8, 10.3%), normal (27–37°, N = 33, 42.3%), or high (>37°, N = 37, 47.4%). (Appendix XIV) No significant differences in sagittal or vertical A-point change from T1 were found across mandibular plane angle categories in either the short-term (sagittal p = 0.810; vertical p = 0.366), long-term (sagittal p = 0.178; vertical p = 0.150), or combined (sagittal p = 0.287; vertical p = 0.113) analyses. (Appendix XV) Although high-angle subjects showed a trend toward greater sagittal relapse (−5.1±5.3 mm) compared to low-angle subjects (−2.4±6.8 mm) in the combined analysis, the small number of low-angle subjects (N = 8) limits the statistical power to detect a meaningful difference. (Appendix XV)

DISCUSSION

Psychosocial Context and Rationale for Intervention

Children and adolescents with cleft lip and palate frequently experience significant psychosocial burden related to facial appearance. Research consistently demonstrates that individuals with visible craniofacial differences report higher rates of social anxiety, negative self-image, peer victimization, and reduced quality of life compared to unaffected peers. (39–41) The psychological impact is often most acute during adolescence, a developmental period in which facial appearance is closely tied to identity formation, social acceptance, and self-esteem. For patients with severe cleft-related maxillary hypoplasia, the midface retrusion creates a distinctly concave facial profile with a sunken appearance, relative mandibular prominence, and retruded upper lip that can be a source of psychosocial distress throughout this critical developmental period.

Conventional orthognathic surgery with Le Fort I advancement has traditionally required patients to defer definitive correction until skeletal maturity. In contrast, maxillary distraction osteogenesis can achieve large midface advancements prior to reaching skeletal maturity, offering visible esthetic and functional improvements to adolescents with severe midface deficiency during a psychologically vulnerable time.

Facial Esthetic Outcomes

Facial esthetic outcomes demonstrated significant initial improvements in both profile angle and lip position, with subjects transitioning from a severely concave profile with retruded upper lips to a convex profile with markedly improved lip positioning. Over time, the facial profile showed a trend toward a return to concavity, although this change did not reach statistical significance. In contrast, upper lip position exhibited gradual and statistically significant retrusion over the follow-up period, a finding that correlates with the sagittal relapse pattern observed at the maxilla. These results are consistent with the existing literature, which reports that soft tissue changes generally follow hard tissue movement, and that initial improvements in concave facial profile and lip position tend to diminish as skeletal relapse occurs. (26) The significant improvement in facial profile and lip position achieved by MXDO-RED, even if partially attenuated by subsequent relapse, may carry important psychosocial implications. For adolescents with cleft lip and palate, the immediate transformation from a severely concave to a convex profile can be a meaningful event in terms of self-perception and social engagement.

Skeletal Outcomes

The sagittal stability of correction is an important determinant of long-term treatment outcomes. This study demonstrates that while maxillary distraction osteogenesis with a rigid external distractor (MXDO-RED) is effective in achieving substantial horizontal advancement of the maxilla, relapse of approximately one-third in the short-term and almost half in the long-term group was seen over the study period. These findings are broadly consistent with those reported in the literature. Systematic reviews report sagittal relapse rates ranging from 16% to 54% in growing patients with cleft maxillary hypoplasia over follow-up periods of one to thirteen years.(30) In contrast, studies of non-growing patients have consistently reported lower relapse rates, ranging from 4% to 8%, with some demonstrating no clinically significant relapse. (27,28,30) The substantially higher relapse rates observed in the present study's predominantly growing cohort are likely attributable in part to continued mandibular growth in the setting of minimal maxillary growth. Previous studies have reported poor vertical stability following

MXDO-RED in growing patients, with one systematic review reporting vertical relapse at A-point as high as 137% and 208%, indicating superior relapse beyond the original preoperative position. (42) The substantial variability in vertical maxillary position observed in the present study similarly suggests that the distracted maxilla is highly unstable in the vertical dimension.

Dental Outcomes

Interpretation of dental outcomes following MXDO-RED is inherently complicated by the fact that these patients routinely undergo orthodontic treatment both before and after the distraction procedure. Pre-distraction orthodontics typically aims to align the arches and coordinate arch forms, while post-distraction orthodontics addresses residual malocclusion. The dental relationships observed at each follow-up time point reflect the combined effects of skeletal change, orthodontic tooth movement, and dental compensation. Additionally, most subjects in this study had maxillary incisor, premolar, or molar agenesis, which presents a considerable challenge to achieving ideal post-distraction occlusion regardless of the skeletal correction obtained.

Given this consideration, overjet showed the most robust changes among the dental variables. Overjet improved substantially following distraction but a trend of mild to moderate relapse was evident over time. The most notable dental finding pertained to crossbite correction. All subjects presented with some form of crossbite prior to distraction, with 70.9% having complete crossbite. Following MXDO-RED, more than one-third of subjects achieved complete resolution of crossbite in the short term, and nearly one-quarter maintained this correction in the long term. Long-term stability of dental relationships following MXDO-RED is not well documented in the literature. However, given the pattern of skeletal relapse observed, concurrent relapse in dental relationships over time would not be unexpected. Future studies examining dental outcomes following MXDO-RED should document the timing and nature of concurrent orthodontic treatment to better distinguish between skeletal relapse and orthodontic-related changes.

Factors Associated with Skeletal Stability

Several factors were associated with skeletal stability following MXDO-RED. The most significant predictor of post-distraction skeletal change was the magnitude of initial advancement, with greater advancement associated with proportionally greater relapse in both the sagittal and vertical planes. This finding has important clinical implications, which is that patients who require the largest corrections are also those at greatest risk for relapse, creating a paradox that should be addressed in preoperative counseling. The positive association between mandibular body growth and anterior and inferior repositioning of A-point suggests that, as the mandible grows, the distracted maxilla also grows downward and forward but in a stalled manner compared to the mandible. As the mandible outgrows the distracted maxilla, the maxillomandibular relationship progressively deteriorates, reducing the net skeletal correction achieved by MXDO-RED. This distinction is clinically important: there is absolute relapse in the distracted maxilla but also proportional relapse due to the continued growth of the mandible in growing individuals, leading to higher risk of recurrence of skeletal Class III occlusion and the need for re-operation. These findings suggest that monitoring mandibular body growth specifically may serve as a useful prognostic indicator for identifying patients at higher risk of deteriorating maxillomandibular relationships following MXDO-RED.

Need for Subsequent Orthognathic Surgery

During the follow-up period, half of subjects required or were planned for subsequent surgery, reflecting the substantial sagittal relapse observed in this study, compounded by continued mandibular growth that further deteriorates the maxillomandibular relationship over time. This rate underscores the importance of counseling patients and families preoperatively that MXDO-RED in growing patients should be viewed not as a definitive, single-stage correction but rather as an intervention that provides meaningful functional and esthetic improvements during adolescence, with the understanding that secondary orthognathic surgery at skeletal maturity may be necessary to achieve a stable, definitive result.

Limitations

The retrospective design of the present study introduces the possibility of selection bias and limits control over confounding variables. The sample was heterogeneous with respect to cleft type, and the cohort was drawn from a single institution, which may limit generalizability. The inability to standardize the timing of follow-up cephalograms introduced variability in the observation intervals. The retention protocol following distraction was not standardized and may have influenced relapse. The confounding effect of concurrent orthodontic treatment on dental outcomes could not be controlled for in this study. Missing data for certain dental variables reduced the power to detect associations and may have introduced bias if data were not missing at random.

Future Directions

Several directions for future research emerge from these findings. First, incorporating validated patient-reported outcome measures assessing psychosocial well-being, self-image, and quality of life would allow researchers to determine whether the functional and esthetic gains from MXDO-RED translate into measurable psychological benefit, and whether these benefits persist despite partial relapse. Second, prospective longitudinal studies following patients from MXDO-RED through completion of skeletal growth are needed, with standardized follow-up intervals and three-dimensional imaging, to more precisely characterize the trajectory and determinants of relapse.

CONCLUSION

MXDO-RED is an effective intervention for achieving substantial correction of severe maxillary hypoplasia in growing patients with cleft lip and palate, delivering meaningful improvements in skeletal relationships, facial esthetics, and dental occlusion during a period of psychosocial vulnerability. However, the considerable sagittal relapse observed remains a significant clinical challenge. The strong association between advancement magnitude and subsequent relapse, along with the independent contribution of mandibular body growth, highlights the complex and multifactorial nature of post-distraction maxillary instability. For patients with severe midface deficiency during adolescence, MXDO-RED provides an early intervention that can profoundly alter facial appearance and function, but patients and families must be counseled that it is best understood as one stage in a potentially multi-stage treatment course, with secondary orthognathic surgery at skeletal maturity remaining a likely possibility for a substantial proportion of patients.

REFERENCES

1. McWilliam J. Hypoplasia of the middle third of the face -a morphological study. Angle Orthod. 1976 Jul;46(3):260–7. doi:10.1043/0003-3219(1976)046%3C0260:HOTMTO%3E2.0.CO;2 PubMed PMID: 1066977.

2. Posnick JC, Makan S, Bostock D, Tremont TJ. Primary Maxillary Deficiency Dentofacial Deformities: Occlusion and Facial Esthetic Surgical Outcomes. J Oral Maxillofac Surg. 2018 Sep;76(9):1966–82. doi:10.1016/j.joms.2018.02.022

3. Wangsrimongkol B, Flores RL, Staffenberg DA, Rodriguez ED, Shetye Pradip R. Skeletal and Dental Correction and Stability Following LeFort I Advancement in Patients With Cleft Lip and Palate With Mild, Moderate, and Severe Maxillary Hypoplasia. Cleft Palate Craniofacial J. 2022 Jan;59(1):98–109. doi:10.1177/1055665621996108

4. Lee JC, Slack GC, Walker R, Graves L, Yen S, Woo J, et al. Maxillary Hypoplasia in the Cleft Patient: Contribution of Orthodontic Dental Space Closure to Orthognathic Surgery. Plast Reconstr Surg. 2014 Feb;133(2):355–61. doi:10.1097/01.prs.0000436857.63021.f0

5. Da Costa AL, Manica D, Manzini M, Bernardi A, Pérez JA, Sekine L, et al. Maxillary Hypoplasia: Differential Diagnosis of Nasal Obstruction in Infants. J Craniofac Surg. 2017 Oct;28(7):e697–700. doi:10.1097/SCS.0000000000003868

6. Molina F, Ortiz Monasterio F, de la Paz Aguilar M, Barrera J. Maxillary distraction: aesthetic and functional benefits in cleft lip-palate and prognathic patients during mixed dentition. Plast Reconstr Surg. 1998 Apr;101(4):951–63. doi:10.1097/00006534-199804040-00010 PubMed PMID: 9514327.

7. Zhang Z, Zhang P, Li S, Cheng J, Yuan H, Jiang H. Skeletal, dental and facial aesthetic changes following anterior maxillary segmental distraction by tooth-borne device in patients with cleft lip and palate. Int J Oral Maxillofac Surg. 2021 Jun;50(6):774–81. doi:10.1016/j.ijom.2020.09.010

8. Fariña R, Lolas J, Moreno E, Alister JP, Uribe MF, Pantoja R, et al. Cleft Lip and Palate Midfacial Hypoplasia: Criteria to Choose the Treatment. J Craniofac Surg. 2022 Mar;33(2):496–501. doi:10.1097/SCS.0000000000007973

9. Proffit WR, Turvey TA, Phillips C. The hierarchy of stability and predictability in orthognathic surgery with rigid fixation: an update and extension. Head Face Med. 2007 Dec;3(1):21. doi:10.1186/1746-160X-3-21

10. Saltaji H, Major MP, Altalibi M, Youssef M, Flores-Mir C. Long-term skeletal stability after maxillary advancement with distraction osteogenesis in cleft lip and palate patients: A systematic review. Angle Orthod. 2012 Nov;82(6):1115–22. doi:10.2319/011212-27.1

11. Dowling PA, Espeland L, Sandvik L, Mobarak KA, Hogevold HE. LeFort I maxillary advancement: 3-year stability and risk factors for relapse. Am J Orthod Dentofacial Orthop. 2005 Nov;128(5):560–7. doi:10.1016/j.ajodo.2004.07.051

12. Valls-Ontañón A, Fernandes-Ciaccia L, Haas-Junior OL, Hernández-Alfaro F. Relapse-related factors of Le Fort I osteotomy in cleft lip and palate patients: A systematic review and meta-analysis. J Cranio-Maxillofac Surg. 2021 Oct;49(10):879–90. doi:10.1016/j.jcms.2021.09.002

13. Jiang L, Zheng Y, Li N, Chen X, Lu Z, Tong H, et al. Relapse rate after surgical treatment of maxillary hypoplasia in non-growing cleft patients: a systematic review and meta-analysis. Int J Oral Maxillofac Surg. 2020 Apr;49(4):421–31. doi:10.1016/j.ijom.2019.08.012

14. Velasques BD, Moura LB, Martins JR, Damian MF, Xavier CB. What is the relapse after Le Fort I maxillary advancement in cleft lip and palate patients? A systematic review. Oral Maxillofac Surg. 2021 Jun;25(2):139–48. doi:10.1007/s10006-020-00906-y

15. Watts GD, Antonarakis GS, Forrest CR, Tompson BD, Phillips JH. Is Linear Advancement Related to Relapse in Unilateral Cleft Lip and Palate Orthognathic Surgery? Cleft Palate Craniofacial J. 2015 Nov;52(6):717–23. doi:10.1597/14-061.1

16. Fahradyan A, Wolfswinkel EM, Clarke N, Park S, Tsuha M, Urata MM, et al. Impact of the Distance of Maxillary Advancement on Horizontal Relapse After Orthognathic Surgery. Cleft Palate Craniofacial J. 2018 Apr;55(4):546–53. doi:10.1177/1055665617739731

17. Polley JW, Figueroa AA. Management of severe maxillary deficiency in childhood and adolescence through distraction osteogenesis with an external, adjustable, rigid distraction device. J Craniofac Surg. 1997 May;8(3):181–5; discussion 186. doi:10.1097/00001665-199705000-00008 PubMed PMID: 9482064.

18. Lucchese A, Gherlone EF, Asperio P, Baena RRY. The Distraction Osteogenesis in Midfacial Hypoplasia. J Craniofac Surg. 2014 May;25(3):831–4. doi:10.1097/SCS.0000000000000738

19. Pandey S, Chug A, Reddy SG, Simre SS. Comparison of Lefort-1 advancement by internal and external distraction osteogenesis in non-syndromic cleft lip and palate: A systematic review and meta-analysis. J Oral Maxillofac Surg Med Pathol. 2025 Jan;37(1):23–30. doi:10.1016/j.ajoms.2024.08.002

20. Hopper RA, Kapadia H, Susarla SM. Surgical-Orthodontic Considerations in Subcranial and Frontofacial Distraction. Oral Maxillofac Surg Clin N Am. 2020 May;32(2):309–20. doi:10.1016/j.coms.2020.01.005

21. Scolozzi P. Distraction Osteogenesis in the Management of Severe Maxillary Hypoplasia in Cleft Lip and Palate Patients. J Craniofac Surg. 2008 Sep;19(5):1199–214. doi:10.1097/SCS.0b013e318184365d

22. Eid A, Ahmed R, Yehia M, Abdelfattah S. Maxillary Distraction Osteogenesis Versus Conventional Orthognathic Surgery in Cleft Lip and Palate Patients: Systematic Review. J

Craniofac Surg. 2025 Jun 1;36(4):1180–4. doi:10.1097/SCS.0000000000011237 PubMed PMID: 40202314.

23. Kinter S, Susarla S, Delaney JC, Chapman K, Kapadia H, Weiss N. Does Distraction Lower Risk of VPI Compared to Conventional Maxillary Advancement? A Retrospective Cohort Study of Adolescents with Cleft Palate. Cleft Palate Craniofacial J. 2024 Mar;61(3):422–32. doi:10.1177/10556656221138895

24. Vincent V, Reddy SG, Markus A. Comparing the socio-economic and emotional implications of management of a hypoplastic cleft maxilla with distraction osteogenesis or orthognathic surgery in a developing country. Br J Oral Maxillofac Surg. 2025 May;63(4):298–302. doi:10.1016/j.bjoms.2025.02.006

25. Cho BC, Kyung HM. Distraction Osteogenesis of the Hypoplastic Midface using a Rigid External Distraction System: The Results of a One- to Six-Year Follow-Up: Plast Reconstr Surg. 2006 Oct;118(5):1201–12. doi:10.1097/01.prs.0000243563.43421.0b

26. Gao WL, Lee YH, Tsai CY, Wu TJ, Lai JP, Lin SS, et al. One-Year Treatment Outcome of Profile Changes After Transcutaneous Maxillary Distraction Osteogenesis in Growing Children With Cleft Lip and Palate. Cleft Palate Craniofacial J. 2022 Mar;59(3):299–306. doi:10.1177/10556656211005638

27. Kanno T, Mitsugi M, Hosoe M, Sukegawa S, Yamauchi K, Furuki Y. Long-Term Skeletal Stability After Maxillary Advancement With Distraction Osteogenesis in Nongrowing Patients. J Oral Maxillofac Surg. 2008 Sep;66(9):1833–46. doi:10.1016/j.joms.2007.10.013

28. Meazzini MC, Basile V, Mazzoleni F, Bozzetti A, Brusati R. Long-term follow-up of large maxillary advancements with distraction osteogenesis in growing and non-growing cleft lip and palate patients. J Plast Reconstr Aesthet Surg. 2015 Jan;68(1):79–86. doi:10.1016/j.bjps.2014.08.069

29. Rachmiel A, Turgeman S, Shilo D, Aizenbud D, Emodi O. Skeletal stability in patients with clefts after large maxillary advancements using intraoral distraction. Br J Oral Maxillofac Surg. 2020 Jul;58(6):663–8. doi:10.1016/j.bjoms.2020.03.023

30. Nimbalkar KA, Datana S, Agarwal SS, Chopra SS, Bhandari SK. Comparison of long-term skeletal stability following maxillary advancement using rigid external distraction in growing and non-growing patients with cleft lip and palate: a systematic review and meta-analysis. Eur J Orthod. 2022 Jan 25;44(1):22–9. doi:10.1093/ejo/cjab017

31. Chua HDP, Cheung LK. Soft Tissue Changes From Maxillary Distraction Osteogenesis Versus Orthognathic Surgery in Patients With Cleft Lip and Palate—A Randomized Controlled Clinical Trial. J Oral Maxillofac Surg. 2012 Jul;70(7):1648–58. doi:10.1016/j.joms.2011.06.226

32. Gao WL, Lee YH, Tsai CY, Wu TJ, Lai JP, Lin SS, et al. One-Year Treatment Outcome of Profile Changes After Transcutaneous Maxillary Distraction Osteogenesis in Growing

Children With Cleft Lip and Palate. Cleft Palate Craniofacial J. 2022 Mar;59(3):299–306. doi:10.1177/10556656211005638

33. Gürsoy S, Hukki J, Hurmerinta K. Five-Year Follow-Up of Maxillary Distraction Osteogenesis on the Dentofacial Structures of Children With Cleft Lip and Palate. J Oral Maxillofac Surg. 2010 Apr;68(4):744–50. doi:10.1016/j.joms.2009.07.036

34. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)—A metadata-driven methodology and workflow process for providing translational research informatics support. J Biomed Inform. 2009 Apr;42(2):377–81. doi:10.1016/j.jbi.2008.08.010

35. Harris PA, Taylor R, Minor BL, Elliott V, Fernandez M, O'Neal L, et al. The REDCap consortium: Building an international community of software platform partners. J Biomed Inform. 2019 Jul;95:103208. doi:10.1016/j.jbi.2019.103208

36. Cicchetti DV. Guidelines, Criteria, and Rules of Thumb for Evaluating Normed and Standardized Assessment Instruments in Psychology. *Psychol Assess.* 1994;6(4):284–290.

37. Kim HY. Statistical notes for clinical researchers: Evaluation of measurement error 2: Dahlberg's error, Bland-Altman method, and Kappa coefficient. Restor Dent Endod. 2013;38(3):182. doi:10.5395/rde.2013.38.3.182

38. Zeger SL, Liang KY. Longitudinal Data Analysis for Discrete and Continuous Outcomes. Biometrics. 1986 Mar;42(1):121. doi:10.2307/2531248

39. Hunt O, Burden D, Hepper P, Stevenson M, Johnston C. Self-Reports of Psychosocial Functioning among Children and Young Adults with Cleft Lip and Palate. Cleft Palate Craniofacial J. 2006 Sep;43(5):598–605. doi:10.1597/05-080

40. Salinero LK, Romeo DJ, Pontell ME, Friedman L, Ahluwalia VS, Wagner CS, et al. Psychosocial Status and Self-Perception in Patients with Cleft Lip and/or Palate. Cleft Palate Craniofacial J. 2025 Jun;62(6):1079–86. doi:10.1177/10556656241236369

41. Lorenz FJ, Ma C, Parikh AC, Lighthall JG. Cleft Lip and/or Palate Is Associated with an Increased Prevalence of Mental Health and Behavioral Disorders. Cleft Palate Craniofacial J. 2025 Apr 30;10556656251331329. doi:10.1177/10556656251331329

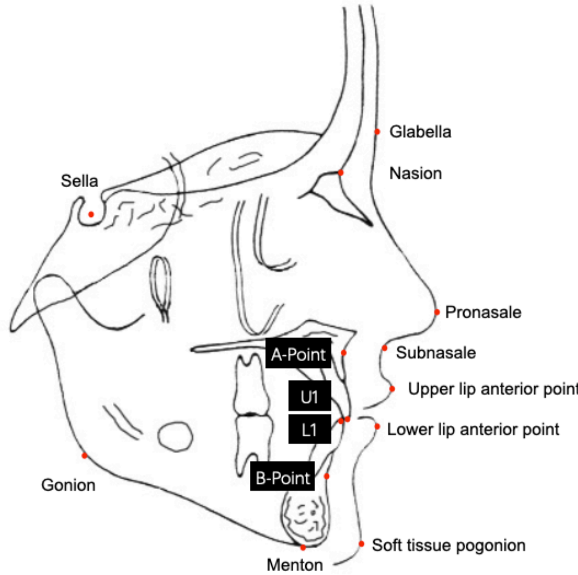
42. Liu K, Zhou N. Long-Term Skeletal Changes After Maxillary Distraction Osteogenesis in Growing Children With Cleft Lip/Palate. J Craniofac Surg. 2018 Jun;29(4):e349–52. doi:10.1097/SCS.0000000000004294

APPENDICES

Appendix I. Cephalometric Landmarks Used in the Assessment of Skeletal, Dental, and Soft Tissue Outcomes Following MXDO-RED

Landmark	Definition
Skeletal Landmarks — Maxillary	
A-point	The deepest midline point on the curved bony outline between the anterior nasal spine and the alveolar crest of the maxilla
Skeletal Landmarks — Mandibular	
B-point	The deepest midline point on the bony contour of the mandible between the infradentale and the pogonion
Menton	The most inferior point on the bony outline of the mandibular symphysis
Gonion	The most posteroinferior point on the angle of the mandible, constructed as the bisection of the posterior ramus border and the inferior mandibular border
Skeletal Landmarks — Cranial Base	
Sella	The geometric center of the pituitary fossa (sella turcica)
Nasion	The most anterior point on the frontonasal suture in the midsagittal plane
Dental Landmarks	
Upper incisor tip	The incisal edge of the most prominent maxillary central incisor
Lower incisor tip	The incisal edge of the most prominent mandibular central incisor
Soft Tissue Landmarks	
Glabella	The most anterior point on the soft tissue contour of the forehead in the midsagittal plane
Pronasale	The most anterior point on the soft tissue contour of the nose
Subnasale	The point where the columella of the nose merges with the upper lip in the midsagittal plane
Upper lip anterior	The most anterior point on the vermilion border of the upper lip
Lower lip anterior	The most anterior point on the vermilion border of the lower lip
Soft tissue pogonion	The most anterior point on the soft tissue contour of the chin in the midsagittal plane

Appendix II. Skeletal, Dental, and Soft Tissue Cephalometric Landmarks Traced for Assessment of Skeletal, Dental, and Facial Esthetic Outcomes Following MXDO-RED



Appendix III. Variables Extracted by Record Type for Assessment of Skeletal, Dental, and Soft Tissue Outcomes Following MXDO-RED

Record Type	Category	Variables Extracted
Electronic Health Record	Demographics	Age at MXDO, sex, race/ethnicity, craniofacial diagnoses (cleft type, syndrome status)
	Operative details	Le Fort osteotomy level, operating surgeon
	Treatment timeline	Operation date, distraction start/end dates, consolidation period, distraction-related complications
	Subsequent treatment	History of subsequent orthognathic surgeries
Lateral Cephalogram	Maxillary position	A-point distance from Y-axis (sagittal), A-point distance from X-axis (vertical)
	Mandibular dimensions	Gonion–Menton (mandibular body length), SN-MP angle (mandibular plane angle)
	Dental relationships[a]	Overjet, overbite
Dental Model	Occlusal relationships	Overjet, overbite, Crossbite
Extraoral Photograph	Facial profile	Profile angle (Glabella–Subnasale–Soft tissue Pogonion)
	Lip position	Upper lip distance to E-line, lower lip distance to E-line

[a] Overjet and overbite were extracted from dental models when available; lateral cephalograms were used when dental models were not present for a given time point.

Appendix IV. Distraction Protocol and Operative Characteristics of Subjects Receiving MXDO-RED (N = 60)

Characteristic	N = 60
Age at Distraction Start, years	
Mean (SD)	14.9 (1.7)
Median (IQR)	14.6 (13.9, 15.9)
Min to Max	10.9, 18.7
Skewness	0.1
Distraction Duration, days (N = 57)[a]	
Mean (SD)	24.2 (9.0)
Median (IQR)	22.0 (18.0, 27.0)
Min to Max	9.0, 50.0
Skewness	0.9
Distraction Rate, mm/day, n (%) (N = 38)[b]	
1.0 mm/day (1 turn/day)	34 (89.5%)
1.5 mm/day	1 (2.6%)
2.0 mm/day (2 turns/day)	2 (5.3%)
4.0 mm/day (4 turns/day)	1 (2.6%)
Total Number of Distraction Turns (N = 10)[c]	
Mean (SD)	42.0 (6.9)
Median (IQR)	43.0 (40.0, 48.0)
Min to Max	28.0, 50.0
Skewness	−0.7
Operating Surgeon, n (%)	
Surgeon A	40 (66.7%)
Surgeon B	15 (25.0%)
Surgeon C	5 (8.3%)

All subjects underwent Le Fort I level maxillary osteotomy with rigid external distractor (RED). The active distraction phase began after a short latency period following osteotomy.

[a] Distraction duration data unavailable for 3 subjects. [b] Distraction rate data unavailable for 22 subjects. [c] Total number of turns data available for only 10 subjects.

Abbreviations: MXDO-RED, maxillary distraction osteogenesis with rigid external distractor; SD, standard deviation; IQR, interquartile range.

Appendix V. Distraction-Related Complications During MXDO-RED in Subjects with Cleft Maxillary Hypoplasia (N = 60)

Characteristic	n	%
Occurrence of Distraction-Related Complications (N = 53)[a]		
Any complication	16	30.2%
No complications	37	69.8%
Type of Complication Among Affected Subjects (N = 16)[b]		
Reverse turn(s) required	3	18.8%
Modification to distractor (other than pin height)	7	43.8%
Other complication	8	50.0%
Pin height modification	0	0.0%
Distraction pause	0	0.0%

[a] Complication status unavailable for 7 subjects; percentages based on 53 subjects with available data. [b] Categories are not mutually exclusive; individual subjects may have experienced more than one complication type.

Abbreviations: MXDO-RED, maxillary distraction osteogenesis with rigid external distractor.

Appendix VI. Intra-examiner Reliability of Cephalometric and Soft Tissue Measurements at Pre-distraction (T0) and Post-distraction (T1) Time Points (N = 20 Re-traced Cephalograms)

Measurement	1st Mean (SD)	2nd Mean (SD)	Diff (SD) [95% CI]	ICC (95% CI)	DE (Min, Max)
Skeletal — Maxillary					
A-pt Y-axis, mm	57.5 (8.6)	58.1 (8.1)	−0.6 (1.1) [−1.1, −0.1]	0.99 (0.97, 1.00)	0.8 (0.0, 4.8)
A-pt X-axis, mm	48.9 (5.4)	48.3 (6.0)	0.6 (1.0) [0.1, 1.1]	0.98 (0.95, 0.99)	0.8 (0.0, 2.4)
Skeletal — Mandibular					
Go–Me, mm	64.2 (4.0)	64.0 (4.1)	0.3 (1.4) [−0.4, 0.9]	0.94 (0.87, 0.98)	1.0 (0.0, 3.6)
SN-MP (°)	40.7 (6.0)	40.4 (6.1)	0.3 (2.7) [−1.0, 1.6]	0.90 (0.77, 0.96)	1.9 (0.0, 6.0)
Dental					
Overjet, mm	−4.6 (8.9)	−4.6 (8.8)	0.1 (0.7) [−0.2, 0.4]	1.00 (0.99, 1.00)	0.5 (0.0, 1.7)
Overbite, mm	−0.4 (6.3)	−0.7 (6.8)	0.3 (2.6) [−1.2, 1.7]	0.92 (0.80, 0.97)	1.8 (0.0, 9.2)
Soft Tissue					
Profile angle (°)	176.2 (16.1)	176.3 (15.2)	−0.1 (7.9) [−3.8, 3.6]	0.88 (0.72, 0.95)	5.4 (0.0, 21.0)
UL to E-line, mm	−1.8 (5.6)	−1.9 (6.0)	0.2 (0.8) [−0.2, 0.5]	0.99 (0.97, 1.00)	0.6 (0.0, 3.3)
LL to E-line, mm	3.2 (4.2)	3.0 (4.1)	0.2 (0.7) [−0.1, 0.5]	0.99 (0.97, 0.99)	0.5 (0.0, 2.2)

Twenty randomly selected cephalograms from T0 and T1 were re-traced by the same examiner ≥2 weeks apart. Diff = 1st − 2nd tracing. ICC = intraclass correlation coefficient (two-way mixed, absolute agreement); ≥0.90 = excellent, 0.75–0.89 = good. DE = Dahlberg error = $\sqrt{(\sum d^2/2n)}$, representing random measurement error in original units.

Abbreviations: A-pt, A-point; DE, Dahlberg error; Go, Gonion; ICC, intraclass correlation coefficient; LL, lower lip; Me, Menton; SN-MP, Sella–Nasion to mandibular plane angle; UL, upper lip; Δ, change.

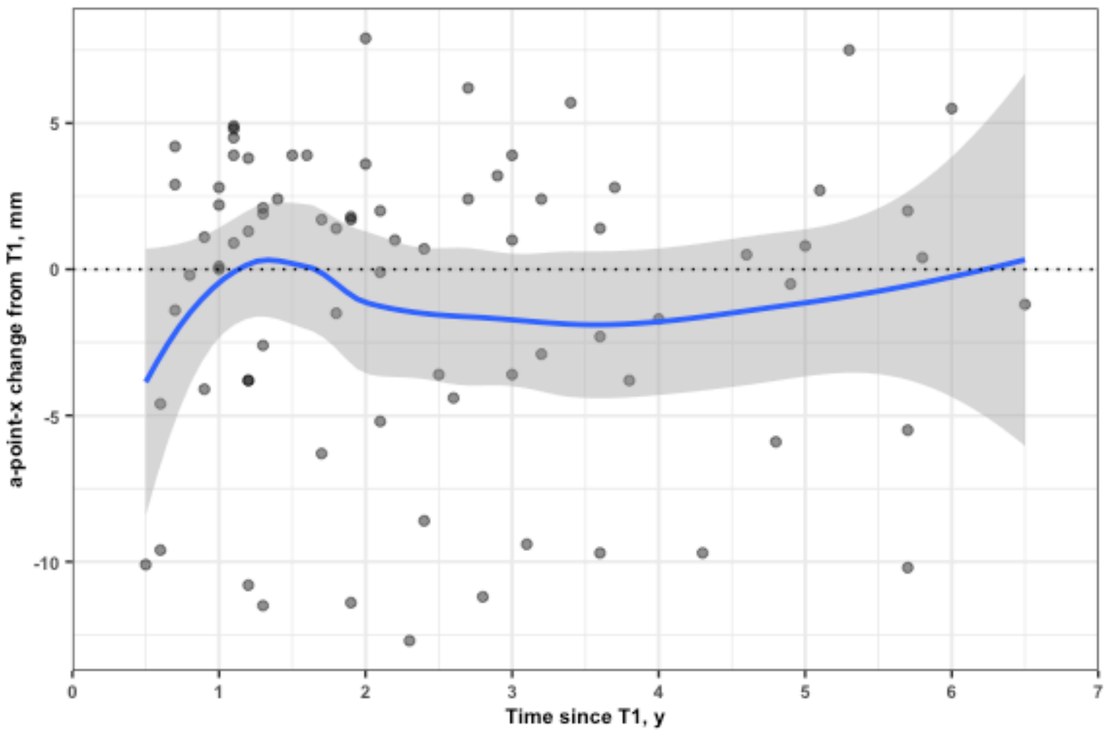
Appendix VII. Intra-examiner Reliability of Cephalometric and Soft Tissue Change Scores from Pre-distraction (T0) to Post-distraction (T1) (N = 20 Re-traced Cephalograms)

Measurement	1st Mean (SD)	2nd Mean (SD)	Diff (SD) [95% CI]	ICC (95% CI)	DE (Min, Max)
Skeletal — Maxillary					
A-pt Y-axis Δ, mm	−13.4 (6.4)	−12.7 (6.2)	−0.7 (1.5) [−1.8, 0.4]	1.00 (1.00, 1.00)	1.1 (0.1, 4.7)
A-pt X-axis Δ, mm	−2.2 (4.8)	−3.0 (5.4)	0.8 (1.1) [0.0, 1.6]	1.00 (1.00, 1.00)	1.0 (0.1, 2.3)
Skeletal — Mandibular					
Go–Me Δ, mm	−3.0 (3.6)	−2.3 (4.2)	−0.7 (1.5) [−1.7, 0.4]	1.00 (1.00, 1.00)	1.1 (0.2, 3.0)
SN-MP Δ (°)	−6.4 (5.8)	−5.3 (5.1)	−1.1 (1.8) [−2.4, 0.2]	1.00 (1.00, 1.00)	1.4 (0.4, 4.0)
Dental					
Overjet Δ, mm	−15.1 (4.0)	−14.8 (4.2)	−0.3 (1.1) [−1.0, 0.5]	1.00 (1.00, 1.00)	0.7 (0.0, 2.4)
Overbite Δ, mm	2.8 (8.6)	4.9 (10.1)	−2.1 (3.9) [−6.9, 2.7]	1.00 (1.00, 1.00)	2.9 (0.3, 8.9)
Soft Tissue					
Profile angle Δ (°)	22.8 (11.3)	25.2 (6.4)	−2.4 (12.2) [−11.1, 6.3]	1.00 (1.00, 1.00)	8.4 (0.2, 23.3)
UL to E-line Δ, mm	−8.8 (2.3)	−9.0 (2.2)	0.2 (1.3) [−0.7, 1.1]	1.00 (1.00, 1.00)	0.9 (0.0, 3.6)
LL to E-line Δ, mm	−0.7 (3.7)	−0.3 (3.8)	−0.4 (0.6) [−0.8, 0.0]	1.00 (1.00, 1.00)	0.5 (0.0, 1.4)

Change scores (Δ) represent T0–T1 differences calculated independently from each tracing session. All ICCs were 1.00, reflecting that between-subject variance in surgical change far exceeded within-subject tracing error. Dahlberg errors for profile angle was notably larger, indicating reduced landmark reproducibility; results should be interpreted with caution.

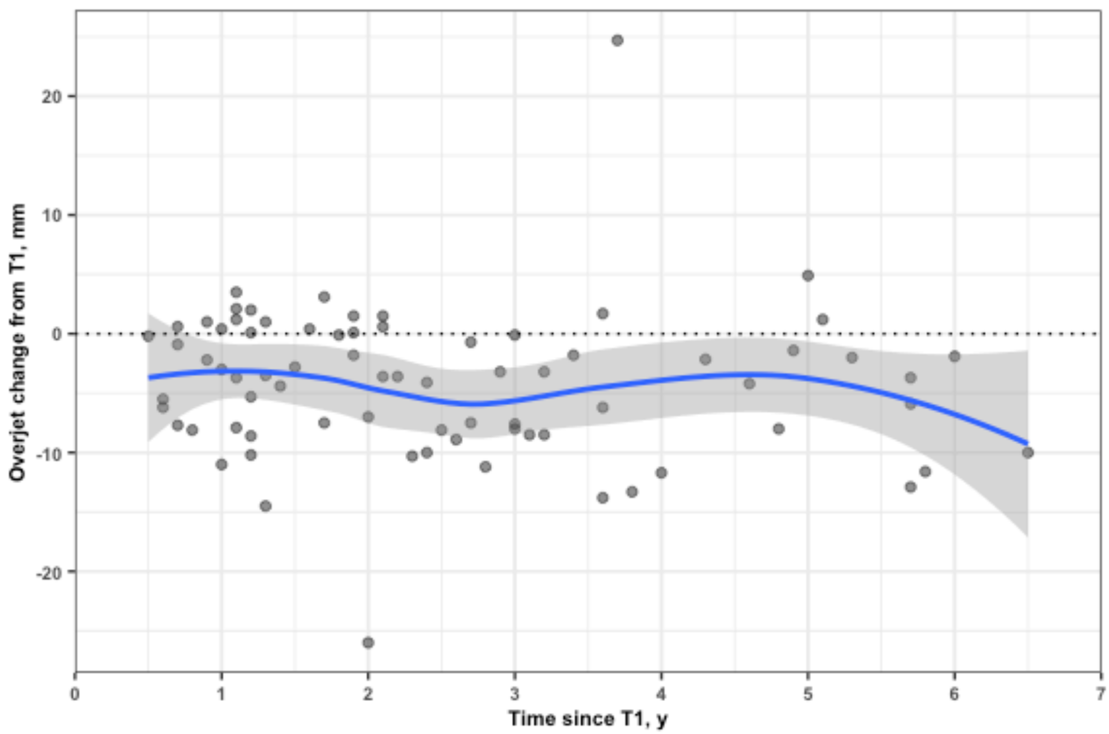
Abbreviations: A-pt, A-point; DE, Dahlberg error; Go, Gonion; ICC, intraclass correlation coefficient; LL, lower lip; Me, Menton; SN-MP, Sella–Nasion to mandibular plane angle; UL, upper lip; Δ, change.

Appendix VIII. Longitudinal Vertical Change in A-point Position (X-axis) During the Post-Distraction Follow-up Period Following MXDO-RED (N = 86 Observations from 60 Subjects)



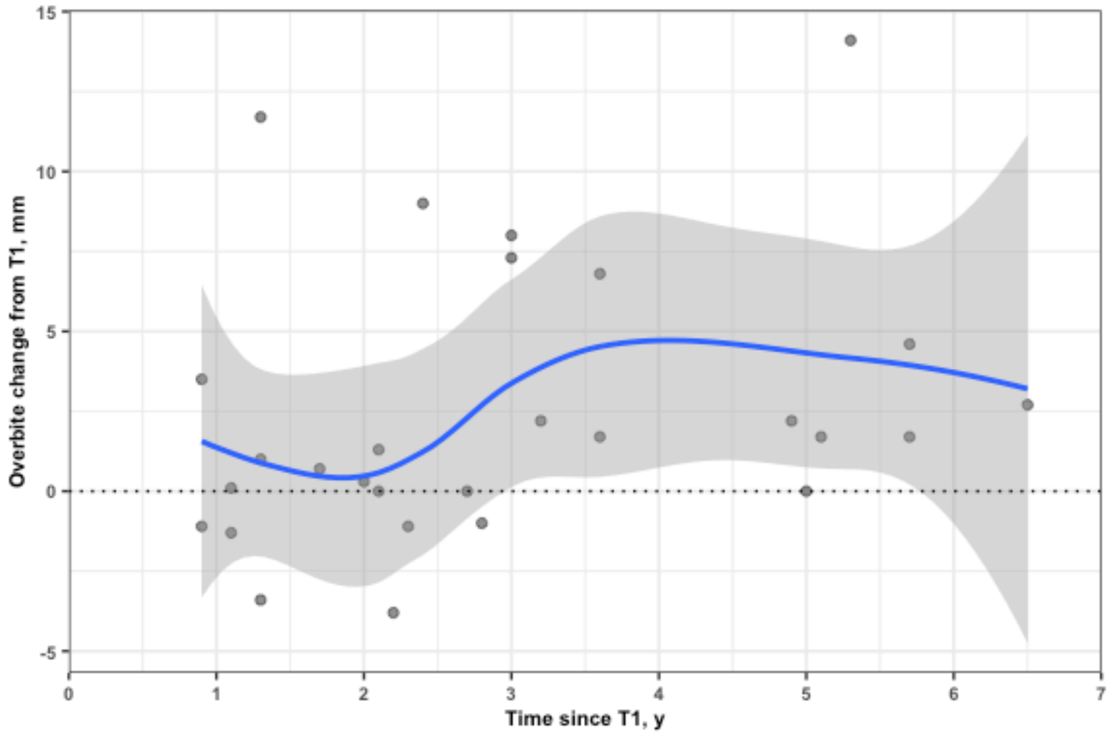
Each point represents one follow-up observation. The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation across repeated observations. The slope of −0.1 mm per year (95% CI: −0.8 to 0.6; p = .850) indicates no statistically significant vertical displacement of A-point relative to its immediate post-distraction position (T1). The correlation coefficient (r = −0.02) reflects a negligible association between follow-up duration and A-point vertical position. Negative values on the Y-axis denote superior movement (intrusion); positive values denote inferior movement relative to T1.

Appendix IX. Longitudinal Change in Overjet During the Post-Distraction Follow-up Period Following MXDO-RED (N = 52 Short-term, N = 23 Long-term Observations)



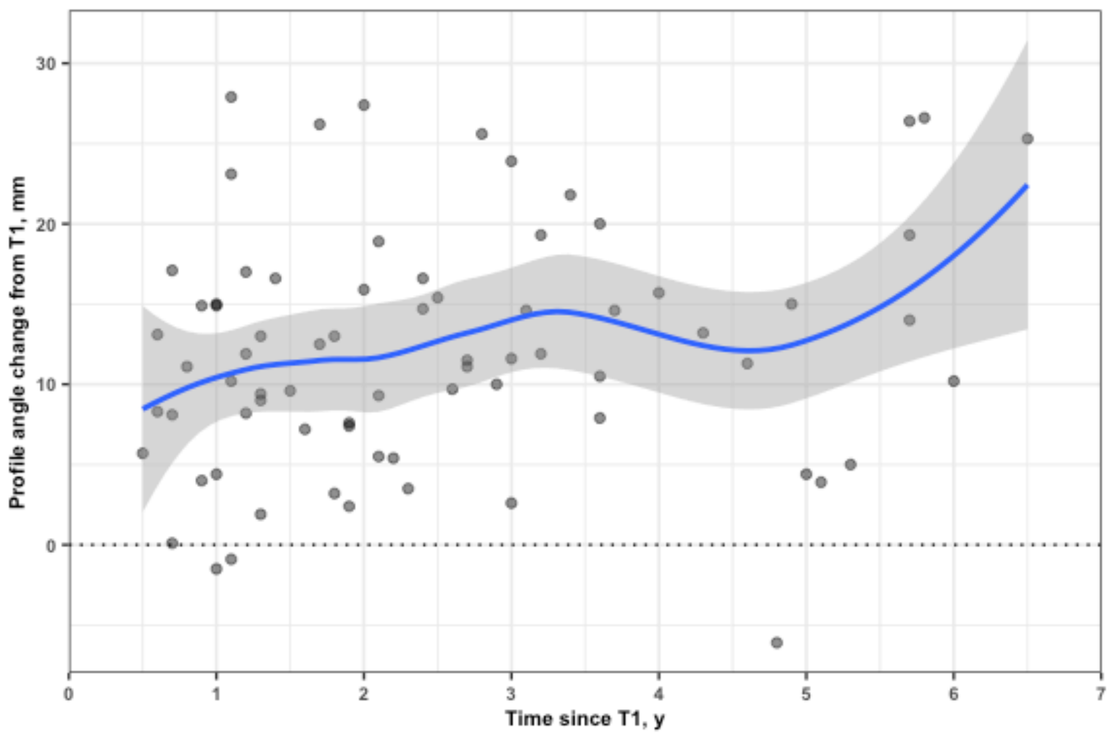
Each point represents one follow-up observation. The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation across repeated observations. The slope of −0.4 mm per year (95% CI: −1.1 to 0.4; p = .373) indicates no statistically significant continuous change in overjet relative to the immediate post-distraction position (T1). The correlation coefficient (r = −0.09) reflects a negligible association between follow-up duration and overjet. Negative values on the Y-axis denote a decrease in overjet; positive values denote an increase relative to T1. Although the overall longitudinal trend was not significant, paired comparisons at discrete time points demonstrated significant decreases in overjet from T1 (see Table 7).

Appendix X. Longitudinal Change in Overbite During the Post-Distraction Follow-up Period Following MXDO-RED (N = 18 Short-term, N = 10 Long-term Observations)



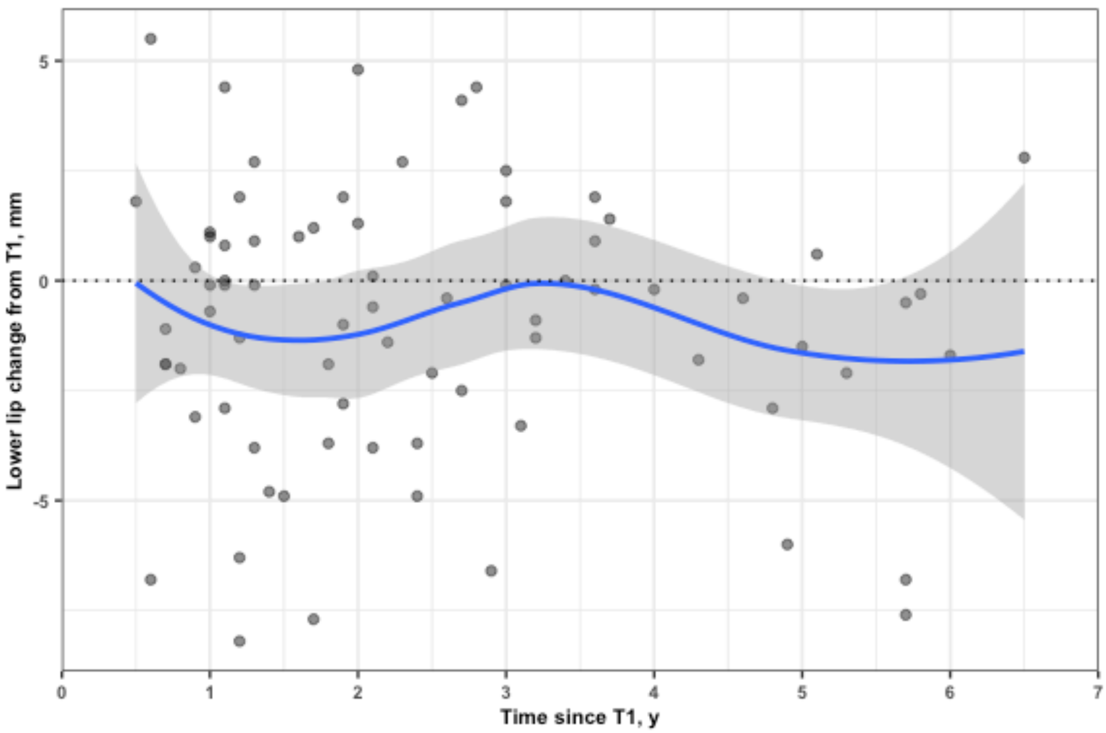
Each point represents one follow-up observation. The regression line and 95% confidence band are derived from a generalized estimating equations (GEE) linear model accounting for within-subject correlation across repeated observations. The slope of +0.7 mm per year (95% CI: −0.2 to 1.6; p = .134) indicates a trend toward increasing overbite over the post-distraction follow-up period, though this did not reach statistical significance. The correlation coefficient (r = 0.27) reflects a weak positive association between follow-up duration and overbite. Positive values on the Y-axis denote deepening of the bite; negative values denote opening of the bite relative to the immediate post-distraction position (T1).

Appendix XI. Longitudinal Change in Profile Angle (Glabella–Subnasale–Soft Tissue Pogonion) Following MXDO-RED in Subjects with Cleft Lip and Palate (N = 52)



GEE linear regression slope (95% CI) = 1.0°/year (−0.2 to 2.1), p = 0.110; correlation coefficient = 0.20. Each point represents the change in profile angle relative to T1. Positive values indicate relapse toward a more convex (straighter) profile. The trend toward increasing profile angle over time did not reach statistical significance.

Appendix XII. Longitudinal Change in Lower Lip Position Relative to E-line Following MXDO-RED in Subjects with Cleft Lip and Palate (N = 54)



GEE linear regression slope (95% CI) = −0.2 mm/year (−0.6 to 0.3), p = 0.519; correlation coefficient = −0.07. Each point represents the change in lower lip distance from E-line relative to T1. No significant change in lower lip position was observed over the follow-up period.

Appendix XIII. Mandibular Growth Measurements and Interval Changes Following MXDO-RED in Subjects with Cleft Lip and Palate: Absolute Values at Each Time Point and Growth Increments during Short-term (0.5–3 Years, N = 55) and Long-term (>3–6.5 Years, N = 23) Follow-up Periods

	Absolute Values at Each Time Point			Interval Changes		
Characteristic	**T0**	**T1**	**Follow-up**	**T1–T0**	**FU–T1**	**FU–T0**
Short-term Follow-up (T2, 0.5–3 years post-distraction; N = 55)						
Gonion to Menton, mm						
Mean (SD)	65.2 (5.5)	66.6 (5.2)	68.4 (5.3)	1.4 (1.3)	1.8 (2.3)	3.2 (2.8)
Median (IQR)	64.4 (61.5, 69.9)	66.8 (62.7, 70.1)	68.7 (64.2, 72.5)	1.1 (0.2, 2.4)	1.2 (0.2, 2.8)	2.1 (1.0, 4.9)
Min to Max	51.7, 74.9	53.2, 77.1	53.5, 79.2	−1.1, 4.5	−3.0, 8.1	−0.3, 11.3
Skewness	−0.2	−0.1	−0.3	0.5	1.0	1.1
Long-term Follow-up (T3, >3–6.5 years post-distraction; N = 23)						
Gonion to Menton, mm						
Mean (SD)	62.7 (5.2)	64.4 (4.9)	67.5 (5.3)	1.7 (1.4)	3.0 (2.7)	4.7 (3.0)
Median (IQR)	61.4 (60.2, 65.8)	64.3 (61.8, 67.2)	67.8 (63.6, 71.5)	1.5 (0.7, 2.9)	2.9 (0.8, 4.5)	3.9 (2.5, 6.0)
Min to Max	51.7, 74.9	53.2, 73.7	53.0, 77.4	−1.2, 5.1	−2.4, 9.0	0.5, 11.8
Skewness	0.3	0.0	−0.7	0.3	0.5	0.7

All values in millimeters (mm). Absolute values represent cephalometric measurements at each time point. Interval changes: T1–T0 = perioperative change; FU–T1 = post-distraction growth during follow-up; FU–T0 = total change from baseline.

Abbreviations: MXDO-RED, maxillary distraction osteogenesis with rigid external distractor; SD, standard deviation; IQR, interquartile range; T0, pre-distraction; T1, post-MXDO; FU, follow-up; T2, first follow-up (short-term); T3, last follow-up (long-term).

Appendix XIV. Distribution of Mandibular Plane Angle (SN-MP) Categories at Pre-distraction (T0) in Subjects with Cleft Lip and Palate Receiving MXDO-RED, by Follow-up Group

SN-MP Category	**Short-term Group (N = 55)**	**Long-term Group (N = 23)**	**All Observations (N = 78)**
Low angle (<27°)	6 (10.9%)	2 (8.7%)	8 (10.3%)
Normal angle (27–37°)	27 (49.1%)	6 (26.1%)	33 (42.3%)
High angle (>37°)	22 (40.0%)	15 (65.2%)	37 (47.4%)

Values are n (%). N = 78 total observations reflects subjects contributing to both follow-up groups (some subjects appear in both short- and long-term analyses).

Abbreviations: SN-MP, sella-nasion to mandibular plane angle; MXDO-RED, maxillary distraction osteogenesis with rigid external distractor; T0, pre-distraction.

Appendix XV Sagittal and Vertical A-point Change from T1 by Pre-distraction Mandibular Plane Angle (SN-MP) Category Following MXDO-RED in Subjects with Cleft Lip and Palate, by Short-term (0.5–3 Years), Long-term (>3–6.5 Years), and Combined Follow-up Periods

Outcome	Low Angle (<27°)	Normal Angle (27–37°)	High Angle (>37°)	p-value
Short-term Follow-up (T2, 0.5–3 years; N = 55)				
N	6	27	22	
Sagittal change from T1, Mean (SD)	−2.5 (7.8)	−3.3 (4.5)	−4.1 (5.5)	0.810
Vertical change from T1, Mean (SD)	1.0 (3.4)	−1.3 (5.3)	−0.5 (5.4)	0.366
Long-term Follow-up (T3, >3–6.5 years; N = 23)				
N	2	6	15	
Sagittal change from T1, Mean (SD)	−2.4 (4.2)	−3.8 (8.9)	−6.6 (4.8)	0.178
Vertical change from T1, Mean (SD)	2.6 (4.4)	−2.7 (4.4)	−1.4 (5.5)	0.150
All Observations (N = 78)				
N	8	33	37	
Sagittal change from T1, Mean (SD)	−2.4 (6.8)	−3.4 (5.4)	−5.1 (5.3)	0.287
Vertical change from T1, Mean (SD)	1.4 (3.4)	−1.6 (5.1)	−0.8 (5.4)	0.113

Values are Mean (SD) in millimeters. Sagittal change = change in A-point distance from Y-axis (negative = posterior relapse). Vertical change = change in A-point distance from X-axis (negative = superior relapse). SN-MP categories at T0: low angle (<27°), normal angle (27–37°), high angle (>37°). No statistically significant differences in sagittal or vertical A-point change were found across mandibular plane angle categories in any follow-up group. p-values from GEE linear regression.

Abbreviations: SN-MP, sella-nasion to mandibular plane angle; MXDO-RED, maxillary distraction osteogenesis with rigid external distractor; GEE, generalized estimating equations; SD, standard deviation; T0, pre-distraction; T1, post-MXDO.