Maxillary distraction osteogenesis and a Le Fort I osteotomy for severe maxillary retrognathia in cleft lip and palate: a case report

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Background: The correction of severe maxillary retrognathia in patients presenting with a cleft palate is challenging due to the complexity of the orthodontic preparation and the magnitude of the surgical movements required, along with the relatively high risk of relapse.
Materials and methods: An 18-year-old Caucasian male with a repaired left-side unilateral cleft lip and palate presented with concerns relating to poor facial aesthetics and poor occlusion. Multidisciplinary treatment involving orthodontics and orthognathic surgery were undertaken to correct the severe maxillary retrognathia. The correction involved the use of internal distraction osteogenesis followed by a conventional maxillary Le Fort I advancement with rotation.
Results: Pre- and post-treatment lateral cephalogram measurements showed the maxilla was advanced 18 mm, rotated clockwise producing a 9 mm increase in vertical dimension at A point and a 7 mm gain in relative arch width across the first molars. Follow-up CBCT superimpositions showed excellent skeletal stability of the achieved anterior-posterior, lateral and vertical corrections over a 2.4-year period, although there was some minor dental relapse.
Conclusion: This case report illustrates the successful use of orthodontics and distraction osteogenesis followed by conventional Le Fort I advancement surgery to correct a severely retrognathic maxilla in a patient with a repaired unilateral cleft lip and palate.
(Aust Orthod J 2020; 36: 205-210)

Received for publication: January 2020
Accepted: August 2020

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Introduction

Midface retrusion is a common consequence of a repaired cleft lip and palate, with maxillary retrognathia becoming more noticeable as facial growth continues.[1] The surgical correction of the severely retrusive maxilla secondary to the cleft presents challenges relating to the significant vertical, lateral and horizontal deficiencies, the difficulty in mobilising the maxilla due to previous scarring, the risk of avascular necrosis and a greater tendency for relapse.[2] As part of a multidisciplinary treatment approach to manage cleft lip and palate patients, orthodontics plays a crucial role.[3] The surgical treatment of the retrognathic maxilla is often achieved by either a conventional maxillary Le Fort I advancement osteotomy or distraction osteogenesis (DO) once facial growth has ceased.[4] DO can involve either external or internal devices and traditionally involves the entire maxilla[5,6] or, more recently, the maxillary anterior segment.[7] The combination of DO followed by Le Fort I maxillary advancement surgery is sometimes undertaken in severe cases in which the occlusal outcome requires further surgical correction.[8]

Prior to undertaking maxillary DO advancement, orthodontic alignment and coordination of the collapsed maxillary dental arches is often required.[9] Adequate bone infill from previous bone grafting of the alveolar cleft site is also likely for an entire maxillary DO advancement. The advantages of DO surgery include a greater magnitude of maxillary advancement

with achievable multidirectional movements during the distraction process, which cannot be managed by a Le Fort I conventional osteotomy.[10] Other advantages include greater stability of the surgical movements, although similar outcomes have been reported for speech improvement when advancements have been undertaken by either surgical procedure.[4]

The purpose of the present report is to describe the multidisciplinary treatment of a patient who presented with extreme maxillary retrognathia and an anterior open bite as a result of a unilateral cleft lip and palate (UCLP). Treatment involved orthodontic therapy and both DO and Le Fort I maxillary advancements, which were reviewed over 2.4 years. Written informed consent was obtained from the patient for the publication of the clinical records.

Case history

An 18-year-old Caucasian male with a left-side UCLP presented with concerns relating to poor facial aesthetics and a poor occlusion. Primary surgical repair of the lip occurred at three months and palatal repair at 12 months of age. Secondary alveolar bone grafting (ABG) was undertaken at 11.2 years and a second bone graft and an upper lip dermal fat graft at 12.1 years. A persistent palatal fistula remained following both ABGs and poor speech was evident despite ongoing speech therapy.

Assessment

Clinical examination

The patient was asymmetric due to a deviated nose and maxilla to the left plus accompanying severe midface retrusion. There was an increased Frankfort mandibular plane angle (FMA) and an associated increase in lower facial height (Figure 1A). The maxillary arch was severely restricted and displayed moderate to severe crowding, while the 22 was agenic. The 23 was unerupted and potentially impacted while the 13 was buccally displaced from the arch form. There was mild to moderate lower anterior crowding affected by lower incisor retroclination. Furthermore, there was a reverse overjet of 13 mm and an anterior open bite that extended posteriorly to the first molars. There were no TMJ nor OSA symptoms reported and the patient had no other significant medical history.

Radiographic examination

The pretreatment OPG confirmed the absence of the 22 and the impaction of the 23. The 48 was also missing but the 18, 28 and 38 were present and potentially impacted (Figure 2A). The lateral cephalometric radiograph (Figure 3A) revealed a severe skeletal Class III malocclusion as a result of maxillary retrusion plus an increased FMA and mandibular prognathism (Table I).

Figure 1. Intraoral and extra-oral clinical photographs (A) Pre-orthognathic orthodontic preparation (B) Pre-distraction placement (C) Orthodontic appliance removal four months post-surgery (D) 2.4 year follow-up.

Table I. Lateral cephalogram analysis: (A) Pretreatment (B) Pre-distraction (C) a) Immediate post-distraction, b) Post-Le Fort I and orthodontic appliance removal (D) 2.4 year follow up.

		A	B	C(a)	C(b)	D
Skeletal AP						
	SNA (°)	73.2	74.9	80.8	82.8	81.3
	SNB (°)	83.5	81.9	82.6	84.1	83.7
	ANB (°)	-10.2	-7.0	-2.2	-1.3	-2.4
	Wits (FOP) (mm)	-20.3	-15.1	-5.1	6.1	-8.3
Dental						
	U1 - SN (°)	88.0	124.8	107.1	113.8	110.1
	U1 - Palatal Plane (°)	99.4	130.3	123.4	126.9	121.1
	L1 - GoGn (°)	71.7	93.7	77.2	72.6	77.8
	IMPA (L1-MP) (°)	69.9	90.8	74.5	69.9	73.6
	Interincisal Angle (U1-L1) (°)	162.3	102.5	133.5	135.1	136.5
	Overbite (mm)	-4.5	-17.3	-0.6	-0.7	-1.0
	Overjet (mm)	-16.3	-8.4	2.1	1.1	-0.2
Facial proportions						
	GoMe/SN (%)	118.6	115.5	119.8	119.6	111.2
	Posterior Face Height (SGo) (mm)	84.8	87.3	84.9	94.3	91.7
	Anterior Face Height (NaMe) (mm)	135.7	145.3	147.4	146.7	141.7
	Anterior Facial Height (N-Me) (mm)	149.2	159.9	162.1	161.4	155.9
	Post/Ant Face Height (%)	63.7	64.9	61.6	64.8	66.8
	AUFH (ant upper facial height) (mm)	55.8	55.7	63.1	59.6	60.0
	ALFH (ant lower facial height) (mm)	73.5	80.3	74.6	77.4	73.2
Vertical						
	FMA (FH-MP) (°)	29.6	36.7	36.3	33.2	28.6
	Palatal-Mand Angle (PP-MP) (°)	28.4	36.3	28.7	28.0	28.9
	Y-axis (SGn - SN -7) (º)	61.8	63.6	65.1	62.3	62.1
	SN - PP (°)	4.5	-1.4	9.3	6.1	4.0
	MP - SN (°)	39.9	41.9	45.0	41.1	39.9
Facial convexity						
	Convexity (NA-APo) (°)	-22.7	-17.1	-3.0	-5.5	-10.6
	Holdaway Angle (NB to H-line) (°)	-8.3	-3.3	4.0	0.4	-1.5

Treatment plan and progress

The horizontal and vertical aspects of the severely retrognathic maxilla were planned for DO surgical correction. Prior to this undertaking, the surgical exposure of the unerupted 23 (along with the removal of the potentially impacted 18, 28 and 38) was prescribed followed by the placement of upper and lower fixed appliances (0.022" preadjusted edgewise) in order to retrieve and align the unerupted 23. In addition, the collapsed minor segment of the maxillary dental arch required orthodontic expansion and the dental arches co-ordinated to facilitate the surgical advancement.

Treatment commenced at age 18.1 years. At 19.1 years, prior to placement of the maxillary distractors, maxillary arch expansion was achieved through the use of co-ordinated 0.019" × 0.025" stainless steel arch wires (Figures 1B and 2B), including the elevation and alignment of the 23 as well as arch coordination. To reduce the risk of maxillary fragmentation during the distraction phase of treatment, a hard-acrylic splint with full palatal coverage was wired to the maxillary fixed appliance at the time of internal distractor placement (Figure 2C). The activation of the distractors was initiated 48 hours after the placement surgery to a prescribed advancement of 1.0 mm per day by twice daily turns of the distractor arms. Light anterior vertical elastics (250 g force) were applied to achieve a downward rotation of the maxilla and a levelling of the upper occlusal plane as the maxilla was advanced. At the completion of the distraction phase, a small residual open bite and a reverse overjet remained. Following a period of consolidation, the distractors were removed and a Le Fort I advancement and rotation was undertaken in order to establish a positive overjet, overbite and correct the maxillary midline (Figures 2 B-D).

Four months following the Le Fort I advancement, the fixed appliances were removed and retainers placed (Figures 1C, 2D and 3D). Long-term retention supervision was provided with follow-up records collected at age 21.1 years (2.4 years post appliance removal).

Treatment outcome

A clinical examination at appliance removal and at subsequent retention visits, including the 2.4 year follow-up appointment, showed satisfactory facial and occlusal relationships with a dramatic improvement in midface projection compared with the initial presentation. Plans to carry out an additional mandibular surgical set back or reduction genioplasty to address the mandibular prominence were declined by the patient.

Pre- and post-treatment lateral cephalogram measurements and 3D imaging superimpositions showed the maxilla was advanced 18 mm, rotated clockwise resulting in an increase in vertical dimension at A point of 9 mm and a gain of 7 mm in relative arch width across the first molars (Figure 2B-D). The 3D maxillary superimpositions showed that the distraction was stable (mostly green) after 2.4 years (Figure 2F-H), although there was some minor dental relapse seen as a reduction in overjet and a slight anterior open bite and midline discrepancy (Figure 1D).

Discussion

This patient presented with challenging clinical features including an impacted unerupted 23, a severely constricted maxillary arch with an accompanying midface deficiency. In addition, there was mild mandibular prognathism that produced an excessive reverse overjet and an extensive anterior open bite. Surgical correction was challenging, not only due to the magnitude and multidirectional nature of the movements required, but also due to the difficulties in mobilising the maxilla because of scarring related to the previous surgery and an increased risk of post-surgical relapse. At the completion of treatment, a dramatic improvement in midface projection and occlusal outcome was achieved.

The use of intermaxillary elastics during distraction resulted in a favourable clockwise rotation of the maxilla, levelling of the upper occlusal plane and closing of the anterior open bite. Post-treatment records show significant maxillary anterior advancement with favourable maxillary lateral and vertical skeletal changes that appeared stable 2.4 years following orthodontic treatment, although dentally, a slight anterior open bite, reduced overjet and midline discrepancy were noted.

An alternative treatment plan to restore the missing upper left lateral incisor by a prosthesis could have been undertaken. This may have resulted in a more ideal buccal occlusion on the left side and possibly a reduced risk of dental relapse. However, the lack of bone at the cleft site would present challenges for possible implant placement. The increased burden of care associated with the long-term maintenance of a prosthetic replacement would also be a consideration.

The patient is now awaiting final rhinoplasty to complete his treatment program; however, the palatal fistula remains despite numerous attempts of closure.

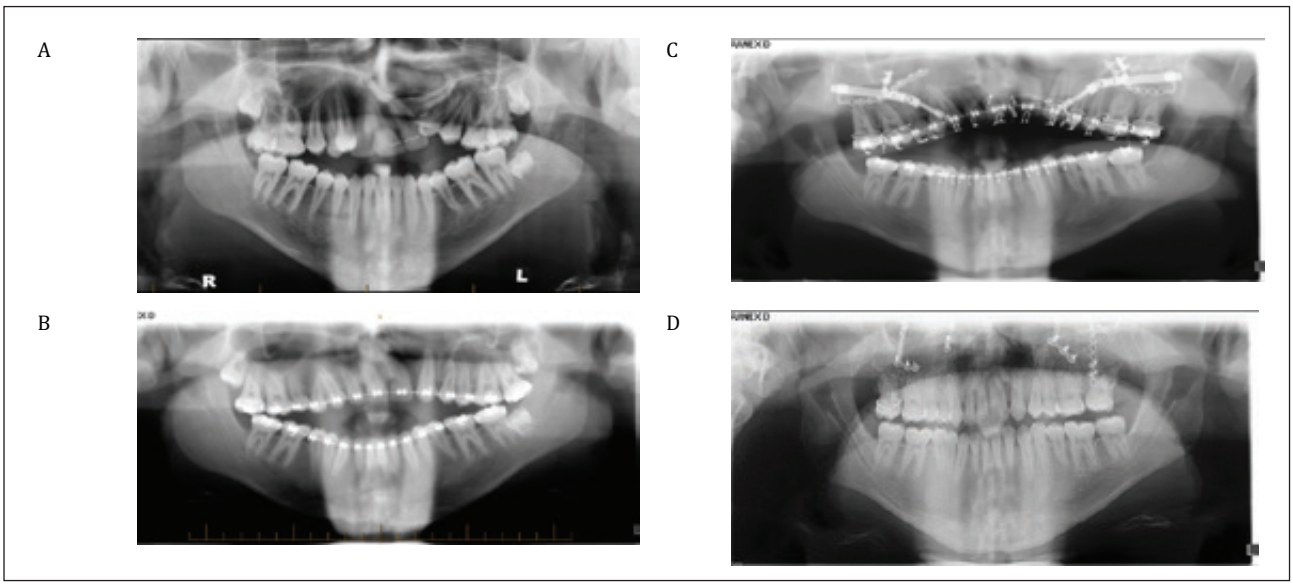

Figure 2. Panoramic radiographs (A) Pre-orthodontics (B) Pre-distractor placement (C) Pre-distractor activation with palatal acrylic splint in place (D) Orthodontic appliance removal.

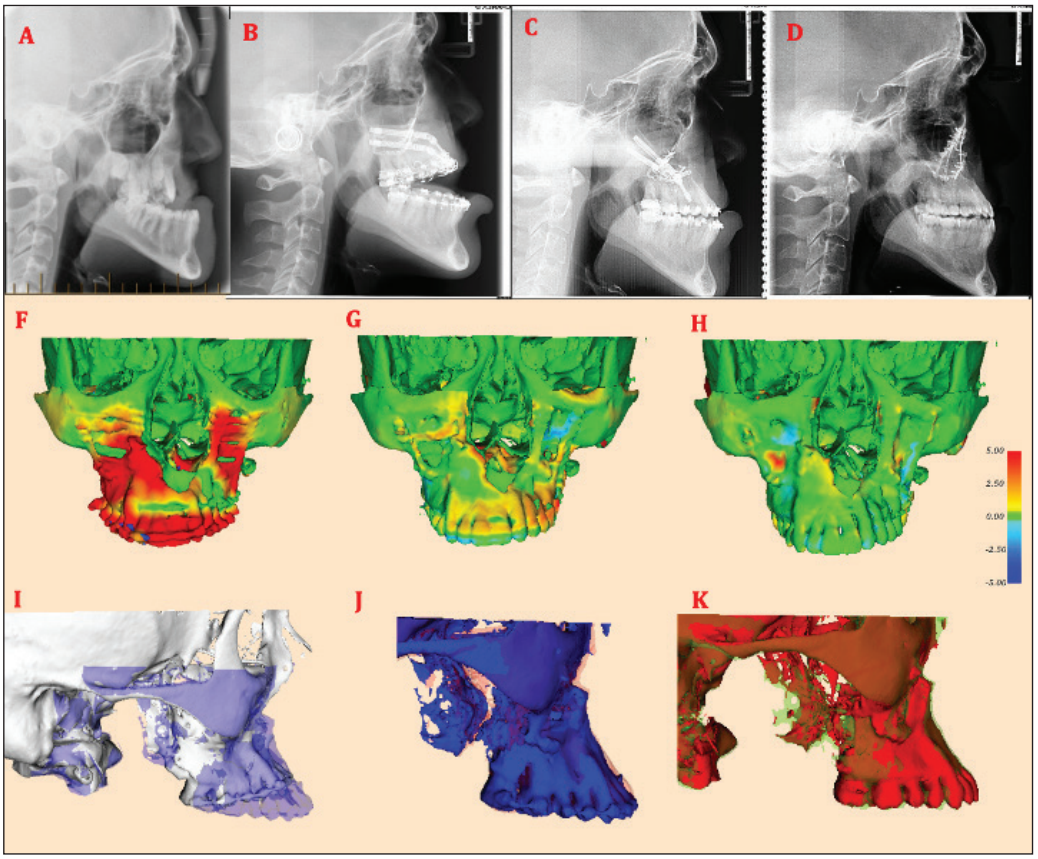

Figure 3. Lateral cephalogram radiographs and Cone Beam Computed Tomography (CBCT) imaging (A) Pre-orthodontics (B) Distractor placement and initial activation (C) Post-distractor activation and pre-Le Fort I (D) 2.4 year follow-up after orthodontic appliances were removed F&I. CBCT superimposition distractor placement and post-distractor activation G&J. CBCT superimpositions post-surgery and orthodontic appliance removal H&K. Post-surgery and 2.4 year follow up

Although speech is not ideal, post-distraction speech assessments showed minimal difference compared with his pre-distraction recordings. Immediately post-surgery, there was a sinus infection that was controlled by the administration of antibiotics. A securing wire tying the hard-acrylic splint was inadvertently left in place (tooth 17) (Figure 2D) and was subsequentially and uneventfully removed.

Summary

The present case report demonstrated the multidisciplinary treatment of a severely retrognathic maxilla in which the use of both DO and a conventional Le Fort I osteotomy was required to surgically position the maxilla in the three planes of space. The skeletal changes addressed several of the challenges of treating severe maxillary retrognathia in cleft lip and palate patients and appeared to be stable over a 2.4-year period.

Acknowledgments

The authors wish to thank Dr Peter Coghlan for his assistance with the surgery.

Declaration of conflicting interests

The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

The patient provided written consent for the publication of clinical data and images.

Funding

The authors received no financial support for the research, authorship and/or publication.

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