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CASE SERIES

Anterior Segmental Distraction Osteogenesis in the Hypoplastic Cleft Maxilla

Report of five cases

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افتراق التكون العظمي للفاصل الأمامي في مرضى نقص تنسج فلج الفك العلوي

عرض لخمس حالات

سروثي راو، أس كوتراشيتي، جي لنجراج، بي بنتو، كي كيلوسكار، سيدراث جان، بيوش سوني، سانتوش راو

الملخص: جراحات تقويم الفك وافتراق التكون العظمي تلعب دورا أساسيا في تصحيح نقص تنسج الفك العلوي لمرضى فلج الشفة والحنك. التقديم الأمامي للفك العلوي بدون التداخل مع مصرة الشراع البلعومي قد تكون مفيدة لمرضى الفلج الذين دائما ما يعانون من عجز في الكلام وتزاحم في الأسنان. نعرض هنا مجموعة لخمسة مرضى مصابين بنقص تنسج الفك العلوي الذين خضعوا لعمليات افتراق التكون العظمي للفاصل الأمامي مع متابعة لمدة سنة. تم استخدام جهاز افتراق مصنع لغرض التثبيت السني مع مفك الوبر المركب بالاتجاه الأمامي والخلفي. تم تقييم تحليل الأنسجة اللينة والصلبة وكذلك تقويم الكلام. بعد مرور سنة على عملية اقتران تكون العظم، تم استقرار تطابق الأسنان والتراكب الأفقي الإيجابي وكذلك تصحيح تزاحم الأسنان بدون الحاجة لقلع الأسنان.تم تحسين المظهر الجانبي للوجه والشفة ولم يكن هناك أي تدهور في الكلام.

مفتاح الكلمات: تكون العظم؛ افتراق؛ جراحة تقويم الفك؛ فلج الحنك؛ قطع عظم الفك العلوي؛ تقرير حالات؛ الهند.

abstract: Orthognathic surgery and distraction osteogenesis play a prime role in the correction of maxillary hypoplasia in patients with cleft lip and palate (CLP). Advancement of the anterior maxilla alone without interfering with the velopharyngeal sphincter may be advantageous in cleft patients, who more commonly have speech deficits and dental crowding. We present a case series of anterior maxillary segmental distraction for maxillary hypoplasia in 5 CLP patients with a one-year follow-up. A custom-made tooth-borne distraction device with a hyrax screw positioned anteroposteriorly was used. The evaluation comprised of hard and soft tissue analysis and speech assessment. A stable occlusion with positive overjet and correction of dental-crowding without extraction was achieved at one year post-distraction. Facial profile and lip support improved. There was no deterioration in speech.

Keywords: Distraction Osteogenesis; Orthognathic Surgeries; Cleft palate; Maxillary Osteotomy; Case Reports; India.

Of all patients born with cleft lip and/or palate (CLP), 25–60% require maxillary advancement to correct the maxillary hypoplasia and improve aesthetic facial proportions.[1] This can be accomplished with the help of orthognathic surgery or distraction osteogenesis, or both of these.[2] The first successful clinical application of anterior maxillary segmental distraction (AMSD) using an intraoral tooth-borne distractor on dogs was reported in 1994 by Block and Brister and on humans in 2003 by Dolanmaz.[3,4] Subsequent clinical research was carried out to determine the optimal protocols for AMSD.[5–10] Here we present a series of five CLP cases treated with AMSD.

Case Series

A total of 5 patients with hypoplastic maxilla secondary to CLP and anterior crossbite with dental-crowding underwent AMSD at our centre. They were from Karnataka and Maharashtra, India,

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Figure 1: Distraction appliance *in situ* showing regenerate (patient 3).

with a mean age of 16.8 years (range: 13–26 years; 4 male, 1 female). All patients had permanent dentition. None of them had previously undergone alveolar bone grafting. Three of the 5 patients had an anterior palatal fistula, one had a tongue flap and one had undergone fistula repair. Written informed consent was taken from the patients and ethical permission was obtained. The patients were followed-up for a period of one year post-distraction.

The patients were prepared using extraoral and intraoral photographs, study casts and digital radiographs, including lateral cephalogram, orthopanthomograph, and intraoral periapical radiographs. In order to standardise the cephalograms, the patients' heads were placed in a natural position with lips in repose and in centric occlusion. Lateral cephalograms were traced using Dolphin Imaging software, Version 11.0 (Dolphin Imaging and Management Solutions, Chatsworth, California, USA). A treatment simulation was performed in order to calculate the amount of advancement required in each case.

Based on the root angulation assessed from preoperative radiographs and the size of the premaxilla, the osteotomy site was chosen as mesial to the first molar in 4 patients and between the two premolars in one patient. With the use of preoperative casts, the appliance was custom-made for each patient. A 12 mm hyrax expansion screw number 140–005 (Great Lakes Orthodontics Ltd, Tonawanda, New York, USA) was used in an anteroposterior direction, parallel to the sagittal plane. This was soldered to a unit with complete tooth coverage using acrylic in two patients, nickel chromium alloy in two patients, and titanium in one patient [Figure 1]. All of the patients underwent presurgical oral prophylaxis, restoration of dental caries and speech assessment by a speech pathologist. Divergence of the roots orthodontically at the osteotomy site was not required for any of the patients.

Anterior maxillary osteotomy was carried out under general anaesthesia with nasoendotracheal intubation. A modified Cupar technique was used with a maxillary buccal vestibular incision from the lateral incisor to the first molar on either side, maintaining the central pedicle. The anterior maxillary segment was mobilised adequately and the prefabricated distractor device was then placed. It was tested to ensure the anterior segment was moving without resistance, and the appliance was then cemented intraoperatively using glass ionomer luting cement (GC FujiCEM™, GC Corporation, Tokyo, Japan). In two patients, the posterior component of the distractor had only a single tooth, and additional palatal screws were fixed to secure the appliance. Postoperatively, all the patients were kept on regular antibiotics, analgesics, and steroids.

A latency phase of 2 to 4 days was given varying proportionately with the age of the patient. Distraction was carried out at a rate of 1 mm/day with a twice daily rhythm. Two of the five patients experienced pain and showed mucosal inflammatory changes at a distraction rate of 1 mm/day due to premature consolidation. When the rate was increased to 1.5 mm/day, these effects ceased. The activation was stopped once the required advancement predicted from the simulation was achieved. This amounted to a 10–12 mm distraction [Figure 1]. The distractor was sealed and retained through a consolidation period of 6–8 weeks.

At the end of the consolidation phase, the distractor was removed and orthodontic treatment was started with banding and bonding carried out at the same sitting. Follow-up records were repeated at the end of activation phase, after the consolidation phase, and at three months, 6 months and one year after the distraction. Each speech analysis was rated according to the Universal Parameters Ratings of Speech Outcomes in Cleft Palate.[11] Statistical analysis was carried out using a paired t–test.

Figure 2: Occlusion pre-distraction and one year post-distraction (patient 1).

Results

The interdental space created by distraction at the osteotomy site was utilised orthodontically to erupt partially-erupted lateral incisors in two patients and correct dental-crowding in the anterior maxilla in all patients, without the need for dental extractions. A functionally stable occlusion with alignment of teeth in the arch and a positive overjet was established at the end of one year [Figure 2]. No patient reported any clinical worsening of the anterior palatal fistula.

Hard and soft tissue parameters were evaluated using the Cephalometrics for Orthognathic Surgery (COGS) analysis for hard and soft tissue, and Steiner analysis [Table 1].[12–14] The preoperative and one-year follow-up values were taken into consideration. On studying the landmarks, there appeared to be an anticlockwise rotation of the maxillary plane together with an increase in the maxillary horizontal length. There was a concomitant opening of the mandibular plane angle. The mean value of the nasion-A point- pogonion angle (N-A-Pg) improved by 12.9º (P = 0.006). The nasion-A point linear measurement (NA) of the horizontal plane value improved by 8.56 mm (P = 0.008). The mandibular plane to horizontal plane angle increased by 2.62º (P = 0.001). The posterior nasal spine-anterior nasal spine linear measurement (PNS-ANS) distance increased by 9.82 mm (P = 0.00). The mean sella-nasion-A point angle (S-N-A) increased from 71.56º to 78.2º with an average increase of 6.64º (P = 0.03). The facial soft tissue contour showed an average decrease in concavity of 7.14º (P = 0.022), thereby establishing a straight to convex profile from the preexisting concave profile. Clinically, there was a significant improvement in the appearance of the patients [Figure 3]. The facial balance was restored and the previously retruded upper lips attained normal protrusion.

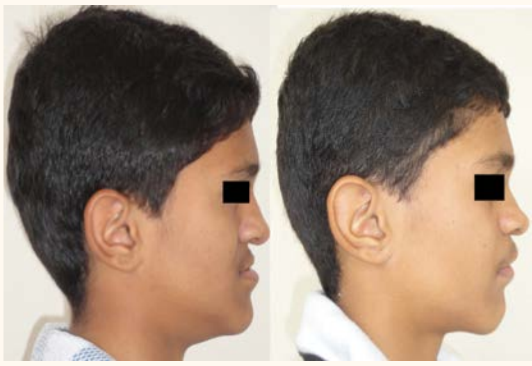

Figure 3: Profile pre-distraction and one year post-distraction (patient 5).

The results of the speech analysis were not statistically significant. However, in two cases there was an improvement in speech understandability, and in two there was an improvement in speech acceptability [Table 2].

In one case, where a high anterior maxillary osteotomy was carried out as directed by the cephalometric tracing, extension prongs were adapted and soldered to the intraoral distractor. Two-hole titanium plates (Synthes, Inc., Sydney, Australia) were fixed bilaterally at the upper end of the osteotomised segment, in the infraorbital region. A pull-through wire was secured around the plate on either side, which exited the skin and was connected to the prongs. This enabled the bodily distraction of the anterior maxilla. The transcutaneous wires were removed along with the intraoral distractor at the end of the consolidation period. There was minimal scarring.

In one patient, the appliance fractured during the consolidation period. Thereafter, an acrylic retention appliance was immediately fabricated and retained *in situ* until the end of consolidation.

Discussion

Various treatment modalities have been used and recommended by different authors for the correction of secondary deformities in CLP patients. The literature reveals that about 25% of patients with maxillary hypoplasia secondary to CLP do not respond to orthodontic-orthopaedic therapy alone and require further intervention.[15] Orthognathic surgery consists of single stage corrective procedures accompanied by internal fixation. Distraction osteogenesis consists of slow regeneration of the bone following corticotomy or

Table 1: Cephalometric analysis for orthognathic surgery

	Preoperative Mean	1 year postoperative Mean	Preoperative SD	1 year postoperative SD	Paired differences Mean	Paired differences SD	Significance
			Hard tissue analysis				
Horizontal skeletal							
N-A-Pg (°)	-14.36	-1.46	5.34	0.92	-12.90	5.29	0.006
N-A (HP) (mm)	-9.06	-0.50	3.99	0.94	-8.56	3.95	0.008
N-B (HP) (mm)	-1.72	-1.66	0.94	1.43	-0.06	0.60	0.834
N-Pg (HP) (mm)	-0.38	-0.24	1.31	1.18	-0.14	0.27	0.311
Vertical (skeletal, dental)							
N-ANS (perp HP) (mm)	40.60	40.50	5.27	5.28	0.10	0.85	0.806
ANS-Me (perp HP) (mm)	62.96	63.90	6.91	6.49	-0.94	0.63	0.03
PNS-N (perp HP) (mm)	40.00	39.80	5.46	5.25	0.20	0.23	0.13
Mandibular Plane-HP (°)	21.42	24.04	1.52	1.63	-2.62	0.76	0.001
Maxilla, mandible							
PNS-ANS (HP) (mm)	40.74	50.56	4.10	4.45	-9.82	1.05	0.00
			Soft tissue analysis				
UL Protrusion (mm)	2.30	3.60	2.43	2.74	-1.30	1.85	0.19
LL Protrusion (mm)	6.68	5.70	2.43	2.86	0.98	1.29	0.17
Nasolabial Angle (°)	96.54	96.00	22.76	17.10	0.54	12.57	0.93
			Steiner analysis selected parameters				
SNA (°)	71.56	78.20	4.45	4.70	-6.64	2.32	0.003
SNB (°)	75.62	78.74	3.83	4.23	-3.12	4.09	0.16
Soft Tissue Convexity (°)	144.1	136.96	3.11	3.04	7.14	4.36	0.02
SN-GoGn(°)	34.64	32.62	3.84	6.04	2.02	5.96	0.49

SD = standard deviation; N-A-Pg = nasion-A point-pogonion angle; HP = horizontal plane; N-A = nasion-A point; N-B = nasion-B point; N-Pg = nasion-pogonion; N-ANS = nasion-perpendicular-anterior nasal spine; ANS-Me = anterior nasal spine-menton; PNS-N = posterior nasal spine-nasion perpendicualr; UL = upper lip; LL = lower lip; S-N-A = sella-nasion-A point angle; S-N-B = sella-nasion-B point angle; SN-GoGn = angle between sella-nasion and gonion-gnathion.

osteotomy after vector planning. In CLP patients, the relapse after orthognathic surgery is greater due to tense scar tissue from multiple previous surgeries. Larger advancements with better stability can be achieved with the help of distraction.[15]

Block *et al.* carried out a pilot study on the distraction of the anterior maxilla in dogs and described the use of a tooth-borne distractor.[3,16] Sagittal advancement of the entire maxilla in CLP patients has the risk of shortening the soft palate and inducing velopharyngeal incompetence. The use of AMSD is recommended as an alternative

Table 2: Speech analysis

	Pre-operative Mean	1 year post-operative Mean	Pre-operative SD	1 year post-operative SD	Paired differences Mean	Paired differences SD	Significance
Hypernasality	1.4	0.8	1.1402	0.4472	0.6	0.8944	.208
Speech understandability	1.6	1.2	0.8944	0.8367	0.4	0.5477	.178
Speech acceptability	1.8	1.2	1.0954	0.8367	0.6	0.5477	.07

SD = standard deviation.

method to conventional Le Fort I osteotomy and rigid external distraction systems for CLP patients with severe velopharyngeal incompetence as it does not affect the velopharyngeal sphincter.[7] Patients who have a class I molar relationship, negative or zero overjet, or impacted or malaligned anterior teeth are ideal candidates.[5,17,18] In these patients, the treatment objective should be to create space for the eruption of impacted anterior teeth or for their alignment by increasing the arch length while maintaining the class I molar relationship. An anterior crossbite and a concave profile can also be addressed using this technique.

External and internal distractors like the Dynaform system, modified hyrax appliance and the hybrid distractors have been described in relation to an anterior maxillary distraction.[8,10,19] The distractor used in this case series had the advantage of easy fabrication, minimal expense and good patient tolerance.

The modified Cupar technique used for these cases was intended to prevent vascular compromise to the previously operated-upon cleft maxillae.[20] The rate of distraction was planned according to age and executed based on the resistance of the maxilla to easy distraction, which would be related to the consolidation changes taking place.[21] In the consolidation phase, the patients had minimal discomfort and psychological trauma in retention of the intraoral tooth borne distractor, in contrast to the conspicuous extraoral distractors.

Similar to previous studies on soft tissue changes in maxillary distraction, in the current case series there appeared to be a clinically significant improvement in facial balance, with positive soft tissue changes produced by increasing the nasal projection, normalising the nasolabial angle and making the upper lip more prominent.[15,17,22,23] The results of this case series were in accordance with the study by Ho *et al*. who reported stable occlusion results after 3 years' follow-up.[17]

There has been no previous study on speech outcomes after AMSD. In this case series, there was no worsening of speech in any patient, possibly attributable to there being no change in the velopharyngeal sphincter. The speech analysis revealed no clinically or statistically significant changes in speech. Improvement in speech understandability and acceptability may be attributed to the improved dental alignment and incisor relationship.

Conclusion

Anterior segmental distraction osteogenesis of the hypoplastic cleft maxilla improves facial balance and aesthetics, and achieves stable occlusion while correcting dental-crowding without any detrimental effect on speech.

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