Change of Lip and Occlusal Cant After Simultaneous Maxillary and Mandibular Distraction Osteogenesis in Hemifacial Microsomia

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Abstract

Background The purpose of this study was to investigate the lip and occlusal cant changes in hemifacial microsomia (HFM) cases after simultaneous maxilla and mandibular distraction osteogenesis (DO) of the mandible.

Patients and Methods Retrospective analysis of all HFM cases at Balaji Dental and Craniofacial Hospital were performed. Patient of either gender with all medical imaging records and pre and post-operative (6 months) facial photographs in natural head position were included in the study. The lip cant change was assessed by the angle of each labial commissure and the bi-pupillary reference line. The line joining the frontozygomatic unions and a parallel line is drawn at the level of anterior nasal spine. The occlusal plane is then traced. A vertical line is traced perpendicular to the frontozygomatic union. The deviation of the occlusal plane from the horizontal is measured as the occlusal cant and a change, between pre and post-operative records was considered as the angle and linear measurements.

Result With the linear measurement, the mean change in occlusal cant was 7.18 ± 1.47 mm while for the mean change in lip cant was 3.31 ± 0.52 mm ($P = 0.120$). For the angular measurement, the mean angle change in occlusal cant was $13.86 \pm 2.69^\circ$ and mean change in angle of lip cant was $8.54 \pm 0.7^\circ$ ($P = 0.01$).

Discussion and Conclusion For type I HFM, DO corrects the occlusal and lip cant. In present study, the lip cant change relative to occlusal cant change was $47.54 \pm 10.71\%$ in linear measurements while for angular

measurements it was $63.19 \pm 10.07\%$ ($P = 0.476$; Pearson's correlation coefficient = -0.241).

keywords Hemifacial microsomia · Distraction osteogenesis · Lip cant · Occlusal cant

Introduction

Hemifacial microsomia (HFM) is a relatively common syndrome involving craniofacial anomalies, with reported incidences of 1 per 3,500 live births [1]. The clinical appearance of the patient is dominated by facial asymmetry together with ear defects. Deviation of the mandible upward and toward the affected side is observed clinically and is associated with canting of the occlusal plane [2]. Lip cant is a soft tissue facial deformity that is usually associated with a skeletal component [3]. Jaw skeletal asymmetry results in an occlusal cant and has been defined as an inconsistency between the amount of dentition and gingival exposure [4]. A soft tissue asymmetry, lip cant change, is determined by the difference in height of the commissure bilaterally [4]. The occlusal cant and lip cant are often notable in patients with HFM [2].

Distraction osteogenesis (DO) is the biological treatment process in which bone formation occurs during slow separation of the segments of bone after a planned osteotomy. McCarthy et al. [5] brought DO to present status in 1992 when they employed DO to lengthen mandibles in children. From then on, DO has been used to alleviate mandibular hypoplasia and asymmetries, hemifacial microsomia and Goldenhar's syndrome, maxillary arch width discrepancies, kleeblattschadel skull deformity, and severe midface deficiencies [6]. In these craniofacial deformities, new bone has been generated in deficient areas

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to restore esthetics, physiologic function and to correct growth. DO has been used in patients with HMO with good results in the past two decades [5, 6]. Facial symmetry in these patients are obtained by the progressive elongation of the hypoplastic mandible of the affected side. Satisfactory esthetic results are often obtained using this versatile technique as the soft tissue remain attached to the bones are expanded slowly, simultaneously and often with adequate time to adapt to the newer position [7].

Scientific literature has a quite number of manuscripts on the use of DO in HFM [8]. Though the various aspects of this modality of therapy have been presented, very few studies have concentrated on the esthetic component of the treatment in terms of lip and occlusal cant. The aim of this retrospective study is to present the effect of mandibular distraction osteogenesis on the lip and occlusal cant in a defined South Indian HFM population.

Materials and Methods

From the archival records (1995–2009) of the Balaji Dental and Craniofacial Hospital, all diagnosed (by history and phenotypic expression) [9] and treated cases of HFM satisfying the following criteria were included in the study. All simple HFM, belonging to type I (Pruzansky's classification) [10], who had undergone mild over correction (2 or 3 mm) of distraction for the shortened ramus height with simultaneous distraction of mandible and maxilla after Lefort I, with all medical imaging records and pre and post-treatment (6 months) facial photographs in natural head position were. Patient who had other interventions including other surgical treatments, orthodontics were not included. Similarly earlier corrective surgeries were also not included. All surgical treatment and measurements were done by the author himself.

Demographical details were collected from case records. For the lip cant measurements, from diagnostic photographs, the method by Namano et al. [11] was employed to assess the angle (measured in degrees) created between horizontal line joining both labial commissure and the bi-pupillary reference line. The lines were extended using a corelDRAW, version 12.0 software and the angle measured. A vertical line was drawn to on the horizontal reference line between the commissures and the distance was used to detect the amount of cant in each patient (measured in mm). Facial height was used to standardize the actual measurement and to account for any photographic magnification (Figs. 1, 2).

The occlusal cant was measured from the posteroanterior cephalometric tracings. The line joining the frontozygomatic unions and a parallel line is drawn at the level of anterior nasal spine. The occlusal plane is then traced at the

Fig. 1 Pre-operative photograph of measuring linear and angular lip cant

Fig. 2 Post-operative photograph of measuring linear and angular lip cant

level of first molar occlusion. A vertical line is traced perpendicular to the frontozygomatic union. The deviation of the occlusal plane from the horizontal, on each side, is measured as the occlusal cant and a change, between pre and post-operative records were considered as the angle after measurements. A vertical line is drawn from the lowest point of orbital rim to occlusal plane in both sides. The difference between sides will yield the occlusal cant (in mm). The tracings and measurement were performed by author and two blinded assistant surgeons and an average taken as final value (Figs. 3, 4).

All the values were entered in SPSS version 16.0. Descriptive statistics are presented. Unpaired *t* test was employed to find the relation between lip and occlusal cant. Pearson's correlation coefficient was employed to find the relative change between the occlusal and lip cant. A *P* value of less than 0.05 was taken as statistically significant.

Fig. 3 Pre-operative cephalometric tracing for measuring linear and angular lip cant

Fig. 4 Post-operative cephalometric tracing for measuring linear and angular lip cant

Results

There were 11 HFM patients satisfying the criteria mentioned. There were 3 males (27%) and 8 females (73%) aged between 14 and 29 years. The mean age was 21.82 ± 4.64 years.

With the linear measurement, the mean change in occlusal cant was 7.27 ± 1.46 mm (Range: 4.18–9.2 mm) while for the mean change in lip cant was 3.31 ± 0.52 mm (Range: 2.45–4.01 mm). Though the difference was approximately one half of the measurement value of the occlusal cant in each case, the correlation was 0.497 with $P = 0.120$ (Graph 1).

For the angular measurement, the mean angle change in occlusal cant was $13.86 \pm 2.69^\circ$ (Range 10° – 18°) and mean change in angle of lip cant was $8.54 \pm 0.7^\circ$ (Range 7.75° – 9.85°). The difference approximately about three fourth of the occlusal cant in each case, the correlation was

Graph 1 Difference between linear lip and occlusal cant changes 6 months after distraction ($P = 0.12$)

0.734 with a high statistical significance ($P = 0.01$) (Graph 2).

The lip cant change relative to the occlusal cant change was $47.54 \pm 10.71\%$ in the linear measurements while for angular measurements it was $63.19 \pm 10.07\%$. The Pearson's correlation test between the occlusal cant changes to the lip cant change revealed no statistical significance ($P = 0.476$; Pearson's correlation coefficient = -0.241) (Tables 1 and 2).

Discussion

HFM is the most frequently encountered form of isolated facial asymmetry. It affects approximately one in 5,000 births and ranking second only to cleft lip and palate as the most common facial anomaly. HFM is a congenital malformation resulting in a deficiency in the amount of hard

Graph 2 Difference between angular lip and occlusal cant changes 6 months after distraction ($P = 0.01$)

Table 1 Linear measurements of cant

	Occlusal cant change	Lip cant change
Average (in mm)	7.27	3.31
Standard deviation	1.46	0.52
Minimum (mm)	4.18	2.45
Maximum (mm)	9.2	4.01

Table 2 Angular measurements of cant

	Occlusal cant change	Lip cant change
Average (in mm)	13.86	8.54
Standard deviation	2.69	0.7
Minimum (mm)	10	7.75
Maximum (mm)	18	9.85

and soft tissue on one side of the face. It is primarily a syndrome of the first branchial arch, involving underdevelopment of the temporomandibular joint, mandibular ramus, mastication muscles and the ear. The affected ear may have an external soft-tissue malformation in addition to being lower set than on the contralateral side. Hearing loss may result from underdevelopment of the osseous components of the auditory system and a diminished or absent external auditory meatus. Occasionally, second branchial arch defects involving the facial nerve and facial muscles coexist with HFM. Patients who have Goldenhar's syndrome, the most severe form of HFM, may have eye tumors and fused spines in addition to the characteristic facial asymmetry [12]. The occlusal cant and lip cant are often notable in patients with HFM [2].

In non-growing adult patients, skeletal deformities of HFM traditionally were treated with osteotomies followed by acute orthopedic movement and osseous fixation. This procedure carried the inherent risk of relapse caused by the inability of muscles to be acutely stretched often compromising the final outcome. Furthermore, poor soft-tissue adaptation often lead to compromised function and esthetics. When an autogenous costochondral grafting was used in more severe deformities, infection, pain and donor site morbidity posed important postoperative concerns along with overgrowth of the graft [12].

Distraction osteogenesis (DO) has become widely accepted method to overcome such drawbacks and treat such conditions. DO is a process in which new bone is formed between the surface of disrupted bone segments that gradually are separated by incremental traction. This method of expanding bone after a surgical corticotomy was first described by Gavriil A. Ilizarov in orthopedics surgeon and has gained wide applicability in the area of craniofacial deficiencies since then [12]. DO delivers a traction forces applied to bone also create tension in the soft tissue,

initiating a sequence of adaptive changes termed distraction histogenesis. This concomitant expansion of the soft-tissue functional matrix allows for multidimensional expansion of the lower jaw. Under the influence of tension produced by distraction, active histogenesis also occurs in skin, fascia, blood vessels, nerves, muscle ligaments and periosteum. Because the lengthening procedure progresses slowly over several weeks, the soft tissue is able to stretch and adapt gradually without loss of sensation. For patients who have HFM, this secondary gain is significant because they lack both bone and overlying soft tissue [12].

DO has been successfully used to treat HMO patients with pleasing results in the past two decades [5, 6]. Though English literature has a quite number of manuscripts on the use of simultaneous DO in HFM [8], very few studies have concentrated on the esthetic component of the treatment in terms of lip and occlusal cant in these patients. The result of the present study has proved that linear occlusal cant change and lip cant change are related by a significant difference at the end of the observatory period while the angular measurement had a high statistical significant correlation of 0.734 ($P = 0.01$) (Graph 2).

Literature has reports of DO elongating all anatomical elements including soft tissues such as muscles, ligaments, vessels and nerves along with the osseous component [7]. Author's observation were consistent with the findings of reported literature of Monasterio FO [7]. This phenomenon causes the clinically pleasing appearance of the subjects.

The ratio of the quantitative amount of soft tissue changes to that of the hard tissue changes by exclusive orthognathic surgery or conventional orthodontic treatment or any treatment that involves bone modification has seldom been reported [7, 13]. In such studies, the factors that could influence this ratio were reported as soft tissue thickness variability or maximal muscle tonicity and these factors might have contributed to the individual variation of this ratio [13] (Table 3).

Lip cant is caused by the difference in height of the labial commissures bilaterally, and the vertical height of commissure is determined by the upward pull from the zygomaticus major and levator anguli oris muscles and the downward pull of the depressor anguli oris muscle [13]. Using these 3 muscles as variables and simplifying these vectors as a 2-dimensional relationship demonstrated that the depressor anguli oris muscle contributes to the change in the commissure position postoperative owing to the disruption of the equilibrium between the adjacent

Table 3 Lip cant to occlusal cant ratio (in percentage)

	Mean $\pm$ SD
Angular	63.19 $\pm$ 10.07%
Linear	47.54 $\pm$ 10.71%

musculature around the corner of the mouth [13]. During DO the muscle attachments that determine the vertical location of these muscles usually move apart, depending on the degree of deformity. The depressor anguli oris muscle attaches in the mandibular proximal segment that moves backwards. Because the manipulation of the maxillary cant results in a change in the mandibular proximal segment's cant, the depressor anguli oris muscle attachment undergoes displacement as well and, thus, contributes to the lip cant adjustment. Moreover, the increase in soft tissue, that usually accompanies the DO in mandible also contribute to the phenomenon. This is exhibited by the significant changes in angular measurements.

Another muscle to be noted is the buccinator group of muscle fibres. The buccinator muscles form the deep layer of the orbicularis oris muscle [13]. The fibers of the middle of the muscle decussate at the angle of the mouth, and the uppermost and lowermost buccinator fibers pass without decussation across the midline into the upper and lower lips. The muscle fibers originating from the mandibular alveolar bone pass to the upper lip and those from the maxilla pass to the lower lip [13]. The lower maxillary alveolar bone, from which the buccinators muscles originate, is the region changed during DO procedures. When the mandible is moved forward, the buccinator muscle interferes with the ipsilateral lower lip and lip commissure.

In the present context, all the muscle plays an important role. However, many more factors could contribute to the change in the lip commissural height. Considering this and relying on the direction of the musculature as a vector would be controversial as pointed out by several studies [13]. Several points including the listed ones has to be further considered. First, the attachment point of the depressor anguli oris muscle can be altered, depending on the change of the mandibular position. This can occur vertically, horizontally, or sagittally. Second, the location of the labial commissure is a long distance from the muscle attachment points; thus, it is difficult to confirm that the hard tissue location change readily occurs after DO. Finally, a vertical movement of the hard tissue not only changes the muscle's length, but also the thickness [13]. These elements support the findings of the present study, which showed that the lip cant soft tissue change was less than one half that of the occlusal cant hard tissue change. Similarly mild overcorrection yields favorable results.

If a poor surgical outcome relative to the occlusal cant correction is expected owing to various practical reasons, the occlusal cant could be overcorrected (as demonstrated in the present study) to benefit additional lip cant correction. Another suggested method [7] would be to correct the lip cants using soft tissue management. During the operation, the muscle's origin and insertion points are detached from the bone and reattached after the operation; this

usually results in a shorter muscle length after reattachment [13]. Therefore, to relocate the position of the labial commissure superiorly, the depressor anguli oris muscle's insertion point could be manipulated on the mandible, and to move the commissure inferiorly, the maxilla's zygomatic major and levator anguli oris muscles could be manipulated by reattachment during the corticotomy procedure [13]. Yet another method of relocating the commissure would be an excision of a muscle or performing a face lift to manage the soft tissue. It is remembered that the above mentioned techniques compromises the fundamental goals of function and esthetics.

The advantage of simultaneous DO includes a single stage correction of several problems including occlusal discrepancy, alleviating the need for latter major orthodontic treatment and over correction will yield predictable results that are esthetically and functionally pleasing. The technique of overcorrection of the occlusal cant by a 2 or 3 mm will provide a much more stable, predictable result than normal correction.

Conclusion

Changes in Lip and occlusal cant in HFM cases after mild over correction treatment are reported. The mechanism of linear and angular change in lip and occlusal cant has been described. Linear change does not appear to be significant whereas the angular change in lip and occlusal cant appears significant. Further large scale multicentric studies are essential to highlight the findings of the present study.

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