ORIGINAL ARTICLE

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Five year follow-up of mandibular distraction osteogenesis on the dentofacial structures of syndromic children

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Structured Abstract

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Objectives – To determine long-term treatment outcomes of mandibular distraction osteogenesis (DO) in syndromic children with severely hypoplastic lower jaws.

Design – Descriptive clinical study.

Setting and Sample Population – Cleft Palate and Craniofacial Centre, Department of Plastic Surgery, Helsinki University Central Hospital. Ten children (mean age 7.6 years) with various syndromes (craniofacial microsomia, Treacher Collins syndrome, Nager syndrome).

Subjects and Methods – Ten growing children with severely retrognathic lower jaws were distracted with an extraoral bicortically fixed DO device (mean, 34 days) followed by a consolidation period (mean, 9 weeks). Orthodontic treatment was performed pre- and/or postoperatively with fixed or functional appliances (mean 14 months).

Outcome Measure – The landmarks on standard lateral cephalometric X-rays were digitized and angular and linear measurements were compared using Student's *t*-test to assess changes in pre-distraction, post-distraction, post-consolidation and 1 year follow-up. Long-term follow-up (2 and 5 years) was interpreted according to mean values.

Results – The measurements of SNB, ANB, facial convexity angle, overjet and overbite, and soft tissue facial profile showed significant correction of the mandibular retrognathia and malocclusion. The mandibular divergence decreased and mandibular, corpus, and ramus lengths remained stable during the consolidation period. The regression of mandibular measurements towards pre-distraction values was observed in skeletal and dental parameters and soft tissue profiles during the first postoperative year and continued during the 2- to 5-year follow-up period. The achieved mandibular corpus and ramus lengthening was stable while the mandible displayed considerable posterior rotation but no further growth. Orthodontic treatment could not overcome the regression of mandibular measurements to their original values. The maxilla continued its normal growth during the long-term follow-up. Two patients had tracheostomies, but the tube was removed from both during or immediately after distraction.

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Conclusion – Excellent short-term structural results of mandibular DO are not stable during the growth of syndromic children because of restricted mandibular growth. Thus, a re-evaluation of DO timing in the improvement of facial aesthetics must be considered. However, mandibular DO in children with severe airway problems is recommended.

Key words: distraction osteogenesis; mandible; maxillofacial surgery; stability

Introduction

The timing of mandibular lengthening has been revolutionized after the introduction of distraction osteogenesis (DO). Early DO in children provides a great treatment opportunity in cases of life-threatening airway problems, and in children who are dependent on tracheostomy tubes or have severe eating and speech problems (1). It is also believed that surgical intervention before skeletal maturity may help prevent secondary growth deformities and reduce the need for later surgery. Early repair has also been thought to improve body image and socialization in children. The psychosocial benefit of early surgery for children however has not been reported.

Mandibular DO has been considered a safe and predictable procedure with a low incidence of major complications (2). Surgical and technical complications include hypoesthesia of nerve, temporal nerve paresis or device-related problems (3–9). Recently, a high percentage of dental injuries was reported in small children as a consequence of the osteotomy cut and the bicortically fixed pins (10). Another major negative treatment outcome is the failure in decannulation from the tracheostomy tube (2, 9).

Although DO has been used for the correction of jaw deformities for over 15 years, very few reports exist on the stability of mandibular DO in the long term. Relapses in mandibular DO are reported to be minimal, because gradual distractions also lengthen the associated soft tissue and the restrictive muscle of mastication (2, 11, 12). The statement of good stability is supported by studies performed in adults or subjects nearing skeletal maturity (7, 8, 13). In syndromic children, long-term studies of mandibular DO are rare and include a single case report or reports of less than a dozen subjects (4, 14–18). These results, however, seem to show long-term instability and relapse in the mandibular DO of growing children (17–20).

The purpose of this clinical cephalometric follow-up study was to determine the effects of mandibular DO on dentofacial structure. Furthermore, this study aims at showing how treatment outcome is maintained during a 5-year follow-up of growing syndromic children.

Patients and methods

Patients

Ten growing children (five boys and five girls, mean age 7,6 years) with severely hypoplastic mandibles were osteodistracted with a bilateral extraoral bicortically fixed mandibular distraction device and followed for an average of 5 (minimum 3 and maximum 7) years. Mandibular hypoplasia was caused by various syndromes: Craniofacial Microsomia (2), Treacher Collins syndrome (5) and Nager syndrome (3).

Surgical techniques and distraction procedure

Complete osteotomy with mobilization was performed in the gonial area of the lower jaw posterior to unerupted molars (21). After a latency period of 4–5 days, the bony segments were distracted by a bicortically fixed extraoral multidirectional device (Leibinger Multiguide; Freiburg, Germany). The distraction rate was 0.5 mm/12 h. The mean distraction period was 34 days (range, 24–49 days) followed by rigid consolidation period (mean 9 weeks, range, 6–12 weeks).The mean total time with the distraction devices attached was 14 weeks (range, 11–18 weeks). Two Nager patients had tracheostomies and the tracheostomy tube was removed during or immediately after distraction. The facial treatment outcome was excellent. Also, the correction of sagittal dental relation was achieved, but a posterior vertical open bite was created. Orthodontic treatment was performed in eight children pre-surgically with fixed appliances to align dental arches and in nine children post-surgically with fixed appliances and elastics or functional appliances to stabilize the achieved occlusion. The post-surgical orthodontic treatment lasted a mean of 14 months (range, 10–19 months).

Methods

Standardized lateral cephalometric follow-up measurements were taken in natural head position pre-distraction (T1), post-distraction (T2), post-consolidation (T3), 1 year (T4) and 2 years (T5) and 5 years (mean 4.9 ± 1.3 minimum 3 and maximum 7 years) (T6) postoperatively. In the analysis, 53 consecutive pre- and postoperative cephalometric radiographs were analysed on a light box using a magnifying glass. All cephalograms were traced by one investigator (S.G.) on 0.003 inch acetate paper for the comparison of anatomical landmarks from the original radiograph to subsequent radiographs. Cases requiring clarification were jointly evaluated by two senior orthodontists (K.H., S.G.). Cephalometric landmarks, planes and their definitions are presented in Fig. 1. All 19 landmarks were digitized with a computerized digitizer (X-metrix; Smartsystem, Turku, Finland) to assess the structural changes in mandibular and maxillary positions. Thirty-five skeletal, dental and soft tissue angular and linear measurements were calculated. A magnification of 10% was taken into account in the linear measurements.

To describe the positional changes, horizontal and vertical reference lines were used (Fig. 1). The horizontal line (HL) was defined as a line through the Nasion rotated 7° to upwards from the SN line. Vertical values were measured in relation to this HL. For sagittal measurements, the vertical line (VL) was defined as the line perpendicular to the HL through the Sella. Within a 2-week period, 20 (N) randomly selected cephalograms were redigitized and the distance (*d*) between the first and second tracings were measured to calculate the inter-measurement error, $\sqrt{(\sum d^2/2N)}$. The range of inter-measurement error was 0.9 mm for linear measurements and 1.3° for angular measurements.

A Student's *t*-test was used to compare the structural changes between pre-distraction, post-distraction, post-consolidation and 1-year follow-up. Long-term follow-up (2 and 5 years) were interpreted according to mean values due to the small sample size (eight patients). The research protocol was approved by the Ethics Committee of Helsinki University Central Hospital (HUS 223/E6/2000).

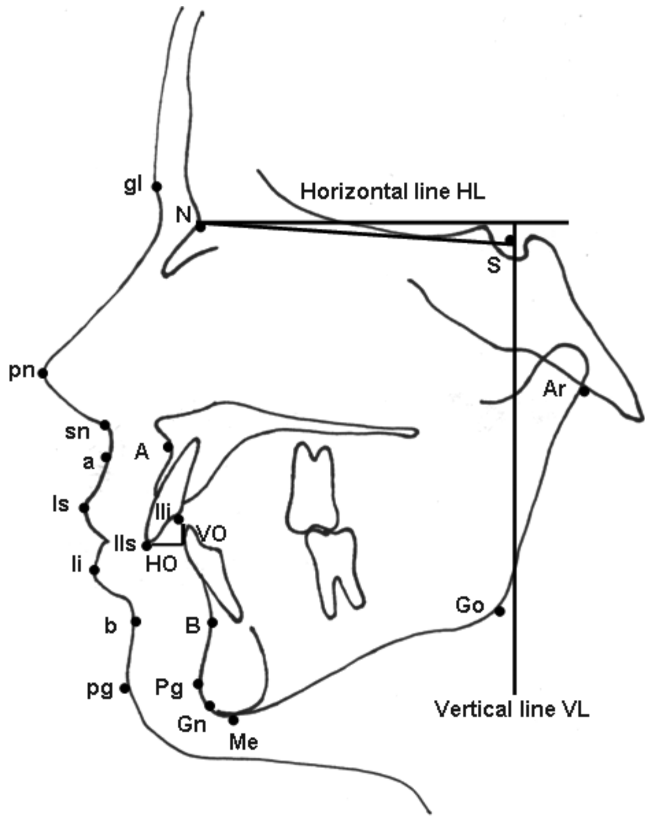

Fig. 1. Diagrammatic representation of landmarks and constructed lines used to identify craniofacial and dental parameters on cephalometric radiographs. *Skeletal and dental points*: **A** Subspinale, the most posterior point of anterior contour of the upper alveolar process; **Ar** Articulare, the point of intersection between the posterior border of the mandibular condyle and the lower border of the cranial base; **B** Supramentale, the most posterior point on the anterior contour of the lower alveolar process; **Gn** Gnathion, the most anterior and inferior point on the mandibular symphysis; **Go** Gonion, the midpoint of angle of mandible; **Ils** Incisive superior, the incisal edge of the upper central incisor; **Ili** Incisive inferior, the incisal edge of the lower central incisor; **Me** Menton, the most inferior point on the mandibular symphysis; **N** Nasion, the most anterior point of the frontonasal suture; **Pg** Pogonion, the anterior point of the chin; **S** Sella, the centre of the sella turcica. *Soft tissue points*: **a** subspinale, the most posterior soft tissue point of the anterior contour of upper lip; **b** supramentale, the most posterior soft tissue point on the anterior contour of the lover lip; **gl** glabella, the most anterior point of soft tissue forehead; **ls** labi superior, the most anterior point of upper lip; **li** labi inferior, the most anterior point of lower lip; **pg** pogonion, the most prominent point of the soft tissue chin; **pn** pronasion, the most anterior point of nasal tip; **sn** subnasale, the point at which columella merges with upper lip; *Lines*: **HO** Horizontal overjet, the distance from incisive superior to mesial surface of lower incisor; **VO** Vertical overbite; the distance between incisive inferior and superior on the vertical line VL; **Horizontal line HL** A line through Nasion rotated 7° upwards from the Sella-Nasion line; **Vertical line VL** A line perpendicular to the Horizontal line through the Sella.

Table 1. **Pre- and postoperative measurements (and significances). Angles are reported in degrees and distances in millimetres**

	Pre-distraction (T1)		Post-distraction (T2)			Post-consolidation (T3)			1 year follow-up (T4)			2 years (T5)		5 years (T6)	
Variable	Mean	SD	Mean	SD	*p*-value T2/T1	Mean	SD	*p*-value T2/T3	Mean	SD	*p*-value T2/T4	Mean	SD	Mean	SD
Maxillary protrusion, SNA	77.1	6.8	76.9	7.4		77.1	7.1		77.7	4.8		77.2	3.4	76.9	4.5
Mandibular retrusion, SNB	62.1	5.6	69.1	5.8	***	69.3	4.2		67.7	3.3	**	66.1	5.1	61.8	3.9
Jaw interrelationship, ANB	15.0	7.5	7.0	5.5	***	7.8	6.6		10.0	6.0	**	11.0	5.8	15.1	5.2
Facial height, Na-Me	92.5	10.3	97.8	7.1	*	97.3	8.2		99.7	8.1		100.7	8.8	108.3	10.8
Lower jaw divergence, SN/Me-Go	54.8	9.8	52.9	9.7	**	48.1	9.1		55.2	12.8	**	53.5	14.6	62.1	10.5
Mandibular length, Me-Ar	62.1	11.9	75.4	9.7	***	73.6	9.7		73	7.2		71.5	10	74.7	9.2
Corpus length, Me-Go	40.0	9.1	48.2	6.1	**	50.3	12.4		48.0	7.4		46.1	9.6	47.0	11.1
Ramus length, Ar-Go	25.4	6.4	32.4	8.7	*	28.9	10.2		28.6	7.4		29.0	8.2	29.1	4.7
Facial convexity, gl-sn/sn-pg	37.0	14.4	24.8	10.2	**	25.8	11.9		27.5	11.6		35.5	10.7	39.9	11.0
Horizontal overjet, HO	9.1	6.5	4.5	5.0	*	5.9	5.7		8.2	7.8		9.5	8.7	10.2	7.5
Vertical overbite, VO	–3.0	3.3	2.5	4.5	*	–1.3	4.8		–0.7	3.9		–1.3	4.0	–1.9	4.0
The distance of point to vertical line, VL (forward/backward)															
A to VL	55.9	3.3	55.7	4.3		55.4	3.1		56.2	3.3		57.4	3.5	58.4	3.6
B to VL	32.5	6.2	41.5	8.3	**	40.0	6.6		37.6	7.0		36.6	10.2	30.3	7.4
Pg to VL	39.6	9.9	45.6	10.4	**	48.6	8.4		45.1	8.0		45.3	11.7	38.9	7.3
Gn to VL	25.9	6.6	36.2	9.3	***	34.8	7.6		32.5	9.2		31.2	13.2	23.5	9.0
Me to VL	22.3	6.2	31.1	9.5	**	31.0	7.4		28.3	8.8		27.0	12.7	18.6	9.7
Go to VL	6.9	2.8	5.2	4.2		4.4	3.2		4.9	3.5		4.5	6.0	9.0	7.8
pn to VL	79.7	3.9	79.8	4.2		79.7	3.5		81.7	3.8		84.2	4.3	85.2	6.2
a to VL	67.7	4.0	68.5	4.4		67.4	3.4		69.0	2.8		71.6	4.0	70.8	6.2
ls to VL	68.4	5.2	69.8	5.4	*	69.1	4.7		70.4	3.4		72.6	4.8	70.5	5.7
li to VL	56.9	7.3	63.0	8.3	**	61.4	7.2		62.1	5.8		63.3	9.0	59.5	7.5
b to VL	47.0	9.5	55.1	9.2	**	53.4	7.9		52.4	8.0		52.4	8.6	48.1	7.4
pg to VL	29.3	6.7	39.9	8.9	**	38.1	7.6		35.4	8.0		34.2	12.4	26.8	8.7
The distance of point to horizontal line, HL (down/up)															
A to HL	44.7	4.0	44.7	4.2		44.6	3.6		45.4	3.9		47.0	5.1	51.2	5.3
B to HL	75.4	11.1	82.2	7.3	*	83.0	8.4		83.8	7.9		84.9	8.4	87.3	11.0
Pg to HL	83.9	8.7	89.2	6.1	*	88.9	5.6		92.6	6.8		91.6	7.2	95.0	10.7
Gn to HL	85.2	11.5	93.3	8.1	**	93.1	8.5		93.9	8.7		94.6	8.7	99.3	10.6
Me to HL	83.9	11.9	93.4	7.7	**	92.6	9.0		94.1	8.1		93.7	8.9	98.1	10.4
Go to HL	55.3	7.6	62.9	9.9	*	57.2	15.4		58.2	9.4		61.6	3.8	60.7	13.1
pn to HL	37.2	5.2	36.9	5.3		36.9	4.4		37.6	5.4		38.4	7.1	41.8	12.6
a to HL	50.8	4.8	51.1	5.2		50.5	5.5		52.6	4.9		53.7	6.1	53.1	12.4
ls to HL	58.6	5.1	57.9	5.1		57.9	3.9		60.8	5.8		62.2	6.7	66.1	10.8
li to HL	70.5	6.6	70.9	4.7		71.0	5.0		73.1	4.8		74.7	5.5	78.9	10.5
b to HL	73.1	7.5	77.8	6.6	**	76.7	4.8		78.9	5.8		82.5	6.5	83.8	9.3
pg to HL	82.9	11.4	90.9	8.2	**	90.7	8.7		91.1	8.4	*	92.3	8.2	96.4	10.8

*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$.

Results

The results are presented in Table 1.

T1-T2 period

During DO, the mandible was significantly advanced as indicated by changes in SNB and ANB angles (8°). The facial convexity (gl-sn/sn-pg) increased significantly (over 13°) and significant increases in the mandibular (Me-Ar), ramus (Ar-Go) and corpus (Me-Go) lengths were consistent findings. The facial (Na-Me) height also increased significantly and the lower jaw divergence (SN / Me-Go) decreased significantly.

There was significant forward movements (mean 6–11 mm) in all mandibular skeletal points (B, Pg, Me and Gn) when measured from VL. The anterior part of the mandible (B, Pg, Me and Gn) was also significantly moved downwards (mean 6–10 mm) when measured from HL. Soft tissue followed the underlying hard tissue and the significant increase was measured in pg, b, ls, li to VL and b and pg to HL.

Dental measurements showed significant closure of the vertical open bite (mean 4 mm) and a decrease in overjet (mean 4 mm).

T2-T3 period

All variables changed insignificantly between the post-distraction (T2) and consolidation (T3) periods.

T2-T4 period

During the first year of follow-up, SNB decreased and ANB increased significantly. The facial soft tissue convexity (gl-sn/sn-pg) increased (mean 3°) insignificantly. The length of the mandible (Me-Ar), ramus (Ar-Go), and corpus (Me- Go) remained almost the same. Lower jaw divergence (SN/Me-Go) increased and the mandible rotated backwards. Facial height (Na-Me) continued to increase insignificantly. The only significant changes related to soft tissues were observed in the b to VL and pg to HL measurements.

T2-T5-T6 period

During 5 years of follow-up, the maxillary protrusion SNA was stable. Significant increases in mandibular retrusion (SNB) and the jaw interrelationship (ANB) seen during the 1-year follow-up period continued to increase, and the measurements reached their pre-distraction values during the long-term follow-up. Also, facial height continued to increase. The significant closing of lower jaw divergence (SN/Me-Go) started to reopen significantly during the first year of follow-up and continued to increase in the long term.

Facial convexity (gl-sn/sn-pg) continued to increase to almost pre-treatment values. During the 2-year follow-up, the mandibular lengths (Me-Ar, Me-Go, Ar-Go) remained almost the same. After 5 years, there was a slight increase in these lengths.

Mandibular skeletal and soft tissue profiles (B, Pg, Gn, Me, b, and pg to VL) continued to move backwards. The maxillary and labial soft tissue points (pn, a, ls and li to VL) moved forward. All skeletal and soft tissue profile points to HP moved downwards.

The values for vertical overbite continued to open and the value for horizontal overjet reached a value greater than the pre-distraction value (mean 10 mm).

Discussion

Mandibular DO is a unique method of choice in cases of dependency on tracheostomy tubes, severe eating problems and sleep apnoea. In our study, two patients had tracheostomies and the tracheostomy tube was removed during or immediately after distraction. However, this cephalometric follow-up study of syndromic children shows how improvements in facial appearance resulting from mandibular DO gradually diminish with growth (Fig. 2). Structural relapse was not observed during the consolidation period. The gradual return of the lower jaw and the facial profile towards their original pre-distraction shape during the 5-year follow-up period was first assumed to be relapse. However, the results showed that the achieved mandibular lengthening was stable and the mandible displayed a considerable posterior rotation, but no further growth occurred. The maxilla, in contrast, continued its normal growth during long-term follow- up.

There are some risk factors related to relapse. Congenital micrognathia seems to relapse more frequently than acquired micrognathia (7). The large antegonial notching and obtuse mandibular angle have been

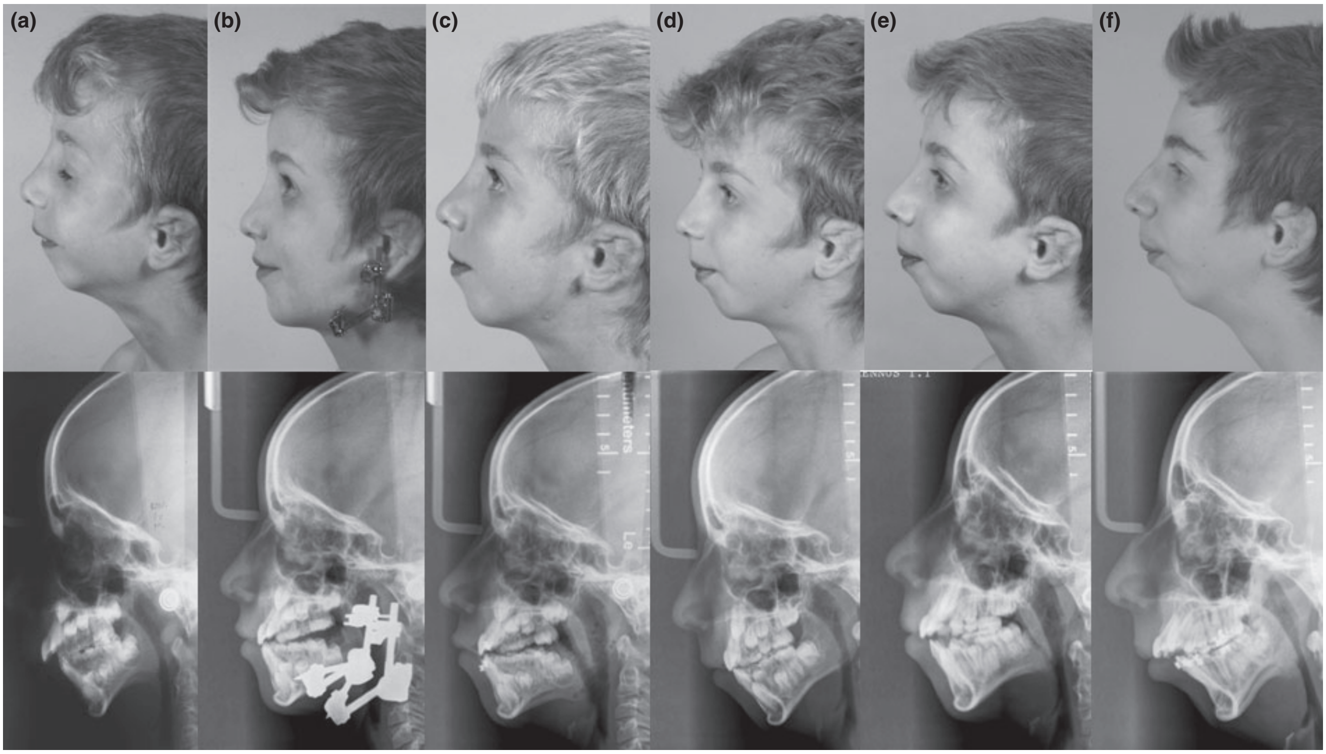

Fig. 2. The profile photographs and lateral cephalometric images of Treacher Collins boy: (a) pre-distraction, (b) post-distraction, (c) post-consolidation, (d) 1-year follow-up, (e) 2-year follow-up, (f) 5-year follow-up.

indicated to return to their original form after DO in patients with Treacher Collins and Nager syndrome (17). Even after growth, the wide divergence of the lower jaw (high mandibular angle) has also been shown to be a risk factor for relapse (13). Risk factors including congenital malformation, growth, high mandibular angle and large gonial angle were present in all patients included in this study.

In this study, a statistically significant increase in the mandibular, ramus and corpus lengths as well as the correction in mandibular skeletal and soft tissue profiles indicated a large distraction distance. Earlier, the distance of mandibular DO lengthening was not shown to be related to relapse, whereas in sagittal split osteotomy, larger advancement is believed to be a predictor for relapse (22). Careful preoperative planning of the DO vector and the control of the vector during distraction has been suggested to be important in achieving good treatment results (23–26). However, the mandibular form has been suggested to be more complex than either the amount or the direction of the DO vector, especially for a long-term period (17).

In this study, the significant correction of the mandibular retrognathia and facial convexity was not stable in the long term. During the first year, a statistically significant relapse occurred in jaw interrelationship, mandibular retrognathia, and facial convexity. During the 5-year follow-up period, the mandibular skeletal and soft tissue profiles continued to move backwards and the mandibular form and position returned almost to the original pre-distraction state, while the surrounding maxillary, nasal and labial measurements lengthened and moved forwards and downwards. The mandibular, ramus and corpus lengths were preserved, but the lower jaw divergence started to open significantly in the first year of follow-up and continued to increase in the long term. The difficulty of establishing a good posterior vertical occlusion might be one of the reasons for this backward mandibular rotation. It was impossible to compare the growth of our patients with that of untreated syndromic patients due to the small sample size in our study and a lack of reported data of syndromic growth. To include large amounts of subjects and to produce statistically comparable results, the analysis of a rare procedure like DO should be performed within multiple centres with similar documentation and protocol.

Distraction osteogenesis orthodontics differs from conventional orthognathic orthodontics because when it is performed during growth, it can utilize the eruption of teeth. The posterior open bite created by vertical lengthening of the ramus is supported postoperatively by an occlusal splint or activator with a simultaneous enhancing of the eruption of most posterior teeth with guiding elastics (24). In this study, the changes at SNB, ANB, overjet, and overbite showed skeletal and dental corrections of the sagittal malocclusion, but in the 5-year follow-up period, the lower jaw and the occlusion showed a gradual retreat to the point of virtually no gain in spite of orthodontic treatment. The observed positional drawback was more severe in the lower jaw than in the occlusion because of effects of the orthodontic treatment and adaptation to the dentoalveolar structure. An overcorrection in mandibular DO has been recommended to minimize the effect of relapse or further growth (4, 12, 24). However, even quite massive overcorrections would not compensate the structural relapse seen in this study. Furthermore, treating a patient for one malocclusion by inducing another is not recommended. In hemifacial microsomia, repeated osteodistractions are recommended over large overcorrections (19).

Hopper et al. (27) demonstrated cephalometric changes during the consolidation period in some cases and thus evaluated it as a dynamic period. The changes occurring during the consolidation period are more related to relapse than growth. However, we could not determine any statistically significant changes related to relapse during the consolidation period. The difference between the relapse and restricted growth potential of distracted tissues is not clearly distinguished. The word relapse can be defined as a loss of a treatment outcome. The conversion of the mandible towards its original shape during the 5-year follow-up period may not be called a relapse. However, the restricted growth potential caused the re-patterning of the skeleton as all the gained mandibular lengths were preserved, but did not increase as expected via normal mandibular growth. Previously, in syndromic children, the return of the mandible to its original shape was suggested to be related to the intrinsic patterning signals within the bone combined with the forces exerted by muscles acting on the mandible (17). The psychosocial effect of the treatment, burden of care and the temporal improvement of the appearance were not evaluated in this study. Also, when DO is performed for the correction of facial appearance, it may also help to facilitate the later sagittal split osteotomy.

Conclusion

This study confirms that in children with severe airway problems, early mandibular DO is recommended. However, our results showed that the improvement in facial appearance in syndromic children is gradually reduced with growth. It is apparent that there are multiple variables and interactions, many of them unrecognized, that influence post-treatment stability over a long-term period. The conversion of the mandible towards its original shape during the 5-year follow-up period may not be called a relapse, but rather is a case where the restricted growth potential of the mandible accompanies the posterior rotation of the mandible. Early application of this treatment for severe airway problems still remains effective. However, when considering DO for the correction of facial appearance, postponing the procedure until later stages of skeletal maturity should be reconsidered.

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