Is there any determinant for successful distraction osteogenesis of the mandible in hemifacial microsomia patients?

Seung-Hak Baek

Dept. of Orthodontics, College of Dentistry, Seoul National University

ABSTRACT

반안면왜소증 환자의 하악골 신장술시 치료결과의 차이에 기여하는 인자

백승학

서울대학교 치과대학 치과교정학교실

본 연구의 목적은 반안면왜소증 환자의 하악골 신장술시 초기 치아골격 특성들 중에서 치료결과의 차이에 기여하는 인자를 찾는 것이다. 치료전의 치아골격 특성, 골신장술의 효과와 그 유지상태를 관찰하기 위하여 골신장술 직전(T0), 직후(T1), 추적 2년후(T2)에 측모와 정모 두부방사선 계측사진을 촬영하여 전후방, 수직, 치아, 비대칭 항목들을 계측하였다. T2 시기의 계측 결과에 따라서 환자들을 1군(양호군, 10명)과 2군(불량군, 9명)으로 분류하였다. 두 군에서 각 시기와 T0-T1, T1-T2 동안의 변화량의 차이를 Mann-Whitney U test, Wilcoxon signed independent t-test, rank test, ANOVA test를 사용하여 분석하였다. Pruzansky type이 골신장술의 성공과 실패 여부와 관계가 깊게 나타났다. T0 시기에 2군은 1군에 비하여 하악골이 후방위치되었고, 하악지 고경(ramus height)이 짧았고, 하악각(gonial angle)이 컸으며, 이환측 하악지가 내측경사되었고, 이환측으로의 이부변위(chin point deviation)가 크게 나타났다. 1군에서 골신장술의 주요한 효과는 하악지 고경의 증가, 하악골의 전방위치, 하악각의 증가, articular angle의 감소에 따른 하악골의 반시계방향 회전, 이환측의 하악지 경사의 증가, 교합면경사와 이부변위의 개선으로 나타났다. 그러나 2군에서는 골신장술을 시행했을 때 1군에 비하여 하악골이 시계방향으로 회전되었고 하악지 고경의 증가량이 작게 나타났다. T2 시기에 2군에서는 하악골의 반시계방향 회전이 나타났고, 하악지 성장이 일어나지 않았으나, 1군은 반대의 경향을 보였다. 이러한 인자들이 골신장술 결과의 차이에 기여하는 것으로 생각된다.

Key word: 골신장술, 반안면왜소증, 하악골, 종단적 결과

Introduction

Hemifacial microsomia (HFM) is the second most prevalent congenital craniofacial anomaly,[1-3] which involves skeletal, soft tissue, and neuromuscular components of the first and second branchial arches.[1, 4-6] Twenty to thirty percent of these patients show some degree of bilateral deformities.[7]

HFM represented a spectrum of clinical findings such as hypoplasia of the mandibular ramus and condyle, confinement of the maxillary vertical growth, external and/or middle ear defects, involvement of some cranial suture, buccal soft tissue, facial nerve, and muscles in the affected side.[8-12] Several classification systems have been developed in an attempt to both qualify and quantify the severity of the skeletal and soft tissue deformity seen in these patients.[3, 6, 9, 13-20]

The spectrum of mandibular malformation ranges from a small but normally shaped ramus and temporomandibular joint to complete absence of these structures.[4] Asymmetric growth of the mandible results in a canting of the occlusal plane superiorly toward the affected side. Tooth development is delayed on the affected side and abnormality of tooth dimension, especially the posterior teeth, in the affected side is proportionate to the degree of mandibular deformity.[21]

Bone lengthening by osteotomy (corticotomy) and distraction osteogenesis (DO) of long bones was first described by Codvilla[22] and popularized by Ilizarov.[23] Since McCarthy *et al.* [24] successfully elongated the human mandible by DO, it has been advocated as an effective and safe technique to increase both ramus height and mandibular body length in young patients with HFM.

Advantages of DO in comparison to conventional surgical techniques are minimalization of surgery and admission time, no bone graft, no donor morbidity, no intermaxillary fixation, remodelling of the mandible, simultaneous increase of soft tissues, and possible improvement of symmetry at an early age.[25-27] Disadvantages and complications of DO are pin tract infection and scar formation in extraoral device case, osteomyelitis, pain, nerve injury, tooth germ injury, angular deviation, failed or delayed ossification, possible relapse, and the frequent need for a second operation after growth has completed.[28]

After DO, most cases still require placement of the removable mandibular occlusal splint and/or the fixed orthodontic appliance to complete treatment.[29-31]

Although there are several follow up studies about results of facial asymmetry correction using DO,[32, 33] there are a few studies about the sagittal and vertical relationships between the maxilla and the mandible. Since the same treatment protocol may produce a good result in one but other patient, it is necessary to divide samples according to retention results. Purpose of the present study was to investigate sagittal, vertical, and asymmetric skeletal characteristics that could determine success or failure of DO of the mandible in HFM patients using longitudinal follow-up data.

Materals and methods

The records of 19 HFM patients (10 males, 9 females) who were treated with DO of the mandible in the Department of Plastic Surgery, Seoul National University Hospital and orthodontic therapy in the Department of Orthodontics, Seoul National University Dental Hospital from 1998 to 2002 were examined.

Posteroanterior (PA) cephalometric radiograph was used for asymmetry analysis and lateral cephalometric radiograph for sagittal and vertical relationships between the maxilla and the mandible. The patient was positioned in cephalostat so that the Frankfort horizontal (FH) and interpupillary lines were parallel to the floor. To locate ear rode accurately and reliably, the amounts of distance from the alar base of the nose to the pina of the ear and from distal canthus of the eye to pina of the ear in the non-affected side were measured. In the affected side, the same amounts of distance were copied along with FH and interpupillary lines.

All patients were treated by extraoral bi-directional distractor appliance (Orthopix Pennig Minifixator, Orthopix, Germany) because it can control vector during distraction period. The oblique osteotomy line was designed in Pruzansky type I and IIa and the horizontal osteotomy line in Pruzansky type IIb and III. The direction of pin fixation is defined by the technique described by Losken *et al.*[34]

Presurgical orthodontic preparation consisted of the lingual arch at the maxillary and the mandibular dentition in order to use elastic traction during distraction. The distraction protocol was as follows: latency period, 5 days; distraction amount, 1 or 2mm a day; distraction rhythm, 0.5mm twice a day or 1.0mm twice a day; consolidation period, 8~10 weeks. Overcorrection of the mandibular dental midline to the non-affected side as much as the width of the mandibular central incisor was done. Postsurgical orthodontic treatment started with the removable mandibular occlusal splint to preclude loading of the distraction site by maintaining the posterior vertical relationship. Three months after stabilization, the affected maxillary dentition was allowed to extrude by selective reduction of this splint and the fixed orthodontic treatment.

Lateral and PA cephalometric radiographs before DO (T0), after DO (T1), and after retention (T2) were traced. The affected side of the mandible in unilateral HFM and more severe side of the mandible in bilateral HFM cases were analyzed to get more objective change of cephalometric measurements. From the measurements at T2, patients were classified into Group 1 showing good retention results (N=10, 5 males and 5 females) and Group 2 showing poor results (N=9, 6 males and 3 females). Relationship among group, Pruzansky type, gender, and lateralism were analyzed by bivariate analysis. Sagittal, vertical, dental and soft tissue, and asymmetric variables were measured (Fig.1-4). Differences at each stage and during T0-T1 and T1-T2 between two groups were compared by Mann-Whitney U test, Wilcoxon signed rank test and ANOVA test.

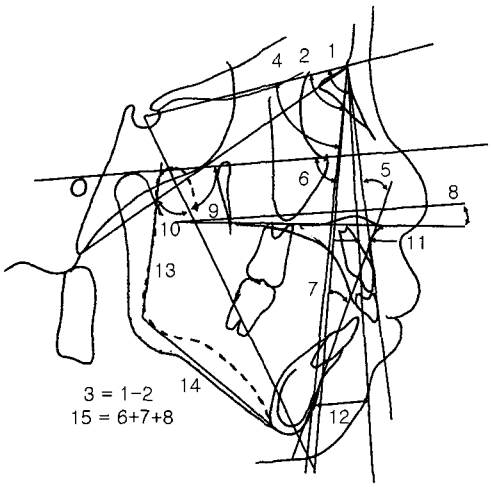

Fig. 1. Sagittal variables

1. SNA (°), 2. SNB (°), 3. ANB (°), 4. Facial plane angle (°), 5. Facial convexity angle (°), 6. Facial angle (Downs) (°), 7. A-B plane angle (°), 8. Palatal plane angle (°), 9. Y-axis to SN (°), 10. Facial axis (Ricketts) (°), 11. A-N perpendicular (mm), 12. Pog-N perpendicular (mm), 13. Ramus height (mm), 14. Mandibular body length (mm), 15. APDI (°); The dotted line is the affected side and the solid line is the non-affected side of the mandible.

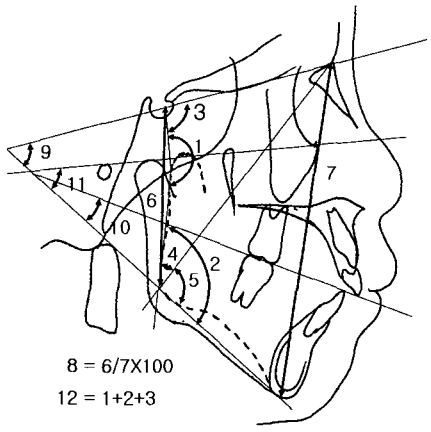

Fig. 2. Vertical variables

1. Articular angle (°), 2. Gonial angle (°), 3. Saddle angle (°), 4. Upper gonial angle (°), 5. Lower gonial angle (°), 6. Posterior facial height (mm), 7. Anterior facial height (mm), 8. Facial height ratio (%), 9. SN-GoMe (°), 10. Occlusal plane to GoMe (°), 11. FMA (°), 12. Bjork sum (°); The dotted line is the affected side and the solid line is the non-affected side of the mandible.

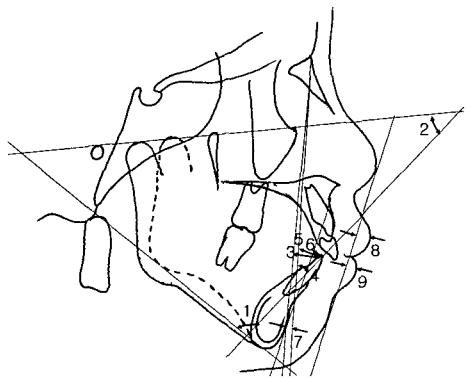

Fig. 3. Dental and soft tissue variables

1. IMPA (°), 2. FMIA (°), 3. L1 to facial plane (mm), 4. L1 to NB (°), 5. L1 to NB (mm), 6. L1 to A-Pog (mm), 7. Pog to NB (mm), 8. Esthetic line to upper lip (mm), 9. Esthetic line to lower lip (mm); The dotted line is the affected side and the solid line is the non-affected side of the mandible.

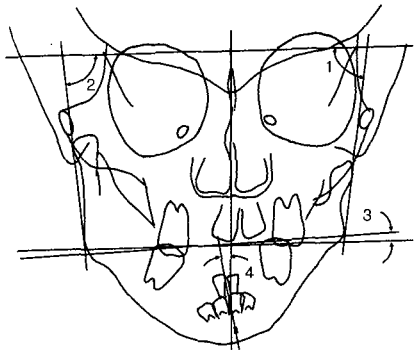

Fig. 4. Asymmetric variables

1. Ramus inclination of the affected side (°) : angle between ramus line of the affected side and zygomaticofrontal suture line, 2. Ramus inclination of the non-affected side (°) : angle between ramus line of the non-affected side and zygomaticofrontal suture line, 3. Occlusal plane canting (°) : angle between occlusal plane and zygomaticofrontal suture line, 4. Chin point deviation (°) : angle between midpoint of the chin and midsagittal line.

Results

The ratio of Pruzansky type I, IIa, IIb, and III was approximately 2.6 : 3.2 : 3.2 : 1.0 and the prevalence of male and female was approximately 6 : 4 (Table 1). Gender did not significantly relate with the prevalence of Pruzansky type (Table 2). The overall distributions of unilateral and bilateral HFM were approximately 9 : 1 (Table 3). Although Pruzansky type did not significantly influence the prevalence of literalism, Group 2 had more tendency of bilateral involvement than Group 1 (Table 4). Group 1 and 2 did not show statistically significant differences in the stages (Table 5). Bivariate analysis showed that Pruzansky type significantly influenced the success or failure of DO ($p<0.01$) (Table 6). When the degree of ramus involvement increased, so did the incidence of failure of DO result.

Cephalometric measurements that showed significant differences between two groups at T0 were: SNB, ANB, facial plane angle, facial convexity angle, Ricketts' facial axis angle, gonial angle, upper gonial angle, ramus height, ramus inclination of the affected side ($p<0.01$), APDI, Y-axis to SN, Pog-N perpendicular, articular angle, occlusal plane to GoMe angle, ramus inclination of the non-affected side, chin point deviation ($p<0.05$). (Table 7-10). Those mean that, at T0, Group 2 showed more posterior positioning of the mandible, smaller ramus height (Table 7), more vertical growth pattern of the mandible, smaller articular angle (Table 8), more labioversion of the mandibular incisor (Table 9), more inward ramus inclination of the affected side, more outward ramus inclination of the non-affected side,

Table 1. Distribution of hemifacial microsomia patients by Pruzansky type and gender.

Pruzansky type		Male	Female	Total
I	unilateral	2	3	5
	bilateral	0	0	0
	total	2 (40%)	3 (60%)	5 (26.3%)
IIa	unilateral	3	3	6
	bilateral	0	0	0
	total	3 (50%)	3 (50%)	6 (31.6%)
IIb	unilateral	3	2	5
	bilateral	1	0	1
	total	4 (66.7%)	2 (33.3%)	6 (31.6%)
III	unilateral	1	0	1
	bilateral	1	0	1
	total	2 (100%)	0 (0%)	2 (10.5%)
Total		11 (57.9%)	8 (42.1%)	19

Table 2. Bivariate analysis of Pruzansky type by gender.

Pruzansky type	Male	Female	% of Female	Spearman's rho (ρ)	*p*-value
I	2	3	60.0		
IIa	3	3	50.0		
IIb	4	2	33.3		
III	2	0	0.0	-0.335	0.161

Table 3. Distribution of hemifacial microsomia patients by Pruzansky type and lateralism.

Pruzansky type		Unilateral	Bilateral
I	Left	3	0
	Right	2	
	total	5(100%)	0 (0%)
IIa	Left	3	0
	Right	3	
	total	6 (100%)	0 (0%)
IIb	Left	5	1
	Right	0	
	total	5 (83.3%)	1 (16.7%)
III	Left	1	1
	Right	0	
	total	1(50%)	1 (50%)
Sum		17 (89.5%)	2 (10.5%)

Table 4. Bivariate analysis of Pruzansky type by lateralism

Pruzansky type	Unilateral	Bilateral	% of bilateral	Spearman' s rho (ρ)	*p*-value
I	5	0	0.0		
IIa	6	0	0.0		
IIb	5	1	16.7		
III	1	1	50.0	0.424	0.070

Table 5. Distribution of hemifacial microsomia patients' group by stages.

Stages	Group 1 (N=10)		Group 2 (N=9)		Sig.
	Mean	SD	Mean	SD	
Before DO (T0)	5Y 9M	1Y 10M	7Y 11M	3Y 1M	NS
After DO (T1)	6Y 6M	1Y 6M	8Y 3M	3Y 3M	NS
After retention (T2)	8Y 7M	10M	10Y 0M	2Y 9M	NS

SD means standard deviation; Sig., Significance; NS : not significant.
Mann-Whitney U test, * $p<0.05$, ** $p<0.01$, *** $p<0.001$

Table 6. Bivariate analysis of Pruzansky type by Group

Pruzansky type	Group 1	Group 2	% of Group 2	Spearman's rho (ρ)	*p*-value
I	5	0	0.0		
IIa	3	3	50.0		
IIb	2	4	66.7		
III	0	2	100.0	0.612	0.005**

** $p<0.01$.

Table 7. Comparison of sagittal measurements in facial skeletal pattern between Group 1 and 2 before DO (T0), after DO (T1), and after retention (T2)

Measurements	Before DO (T0)					After DO (T1)					After retention (T2)				
	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.
	Mean	SD	Mean	SD		Mean	SD	Mean	SD		Mean	SD	Mean	SD	
SNA (°)	79.15	2.82	77.12	2.36		78.44	2.86	77.42	2.84		78.01	2.69	76.95	2.34	
SNB (°)	73.66	2.23	68.98	2.31	**	76.12	3.05	71.49	3.00	*	75.33	2.69	71.48	3.32	
ANB (°)	5.49	1.40	8.14	1.22	**	2.32	1.68	5.93	1.62	**	2.68	2.19	5.47	1.50	*
Facial plane angle (°)	71.67	2.56	65.82	3.20	**	74.37	2.39	68.13	3.81	**	74.19	1.99	68.23	3.27	**
Facial convexity angle (°)	15.25	4.65	22.88	2.18	**	8.40	5.18	18.50	4.05	**	7.80	5.21	17.54	4.41	**
Facial angle (Downs) (°)	81.41	2.56	76.82	4.54		84.10	2.34	78.74	4.72	*	83.93	2.69	79.22	4.25	
A-B plane angle (°)	-6.04	1.65	-8.10	3.21		-1.71	2.41	-4.86	3.73		-2.48	3.29	-4.62	2.83	
APDI (°)	75.05	4.02	69.35	1.67	*	82.37	4.82	75.68	3.57	*	81.12	5.97	73.99	6.83	
Y-axis to SN (°)	75.70	1.99	80.88	3.38	*	74.19	1.36	80.28	3.56	**	75.35	1.76	80.51	2.81	**
Facial Axis (Ricketts) (°)	80.22	2.98	73.71	3.03	**	83.10	3.34	75.63	5.02	*	80.42	2.74	74.66	3.89	*
A-N perpendicular (mm)	-1.11	2.86	-1.56	3.46		-1.73	2.65	-1.59	2.48		-2.22	2.50	-1.87	2.07	
Pog-N perpendicular (mm)	-15.76	5.54	-24.11	6.23	*	-10.73	4.11	-21.34	8.67	*	-11.74	5.40	-21.18	8.40	*
Ramus height (mm)	29.73	4.43	22.13	2.57	**	34.76	6.12	24.92	5.74	*	28.57	4.27	24.53	8.16	
Man. body length (mm)	61.71	5.68	52.22	7.63		61.80	9.61	57.93	9.61		71.20	5.02	74.73	8.05	

DO means distraction osteogenesis, SD, standard deviation; Sig., Significance
Mann-Whitney U test, * $p<0.05$, ** $p<0.01$, *** $p<0.001$

Table 8. Comparison of vertical measurements in facial skeletal pattern between Group 1 and 2 before DO (T0), after DO (T1), and after retention (T2)

Measurements	Before DO (T0)					After DO (T1)					After retention (T2)				
	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.
	Mean	SD	Mean	SD		Mean	SD	Mean	SD		Mean	SD	Mean	SD	
Articular angle (°)	143.00	4.98	133.11	9.27	*	131.02	7.99	140.94	5.05	**	138.51	9.90	132.28	8.52	*
Gonial angle (°)	139.80	5.12	150.67	8.03	**	149.69	10.53	150.74	9.57		145.38	10.50	155.96	11.11	
Bjork sum (°)	409.01	4.89	410.68	8.63		408.52	4.65	416.33	6.84	*	410.71	4.05	415.60	7.15	
Upper Gonial angle (°)	56.94	2.18	66.15	8.89	**	63.69	9.50	60.97	6.68		60.51	6.81	69.37	10.38	
Lower Gonial angle (°)	82.85	3.85	89.01	8.35		86.00	2.96	89.77	7.30		84.87	4.95	86.59	5.32	
Post. Facial Height (mm)	58.56	8.33	60.19	6.22		62.97	8.55	59.64	9.58		62.01	7.92	57.58	10.03	
Ant. Facial Height (mm)	110.48	10.56	112.96	9.05		114.40	8.91	116.51	9.17		120.73	7.51	120.49	10.36	
Facial Height Ratio (%)	52.94	5.28	53.36	4.91		55.04	6.28	51.14	6.95		51.28	4.90	47.65	6.02	
SN-GoMe (°)	49.01	4.89	55.16	5.46		48.52	4.65	56.33	7.84		50.71	4.05	55.94	7.03	
Occlusal plane to GoMe (°)	16.86	3.02	23.25	4.50	*	22.30	4.35	28.27	7.30		22.97	4.41	25.81	9.02	
FMA (°)	39.24	6.37	44.29	5.80		38.80	5.04	45.75	9.06		40.95	4.23	44.96	9.20	

DO means distraction osteogenesis, SD, standard deviation; Sig., Significance.
Mann-Whitney U test, * $p<0.05$, ** $p<0.01$, *** $p<0.001$

Table 9. Comparison of dental and soft tissue measurements between Group 1 and 2 before DO (T0), after DO (T1), and after retention (T2)

Measurements	Before DO (T0)					After DO (T1)					After retention (T2)				
	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.
	Mean	SD	Mean	SD		Mean	SD	Mean	SD		Mean	SD	Mean	SD	
IMPA (°)	83.54	5.92	87.01	10.82		77.31	3.55	84.35	10.53		79.22	8.81	87.36	10.41	
FMIA (°)	57.22	8.22	48.70	6.93		63.90	5.01	49.90	5.00	***	59.83	8.39	47.68	8.35	*
L1 to facial plane (mm)	5.81	3.33	12.54	3.89	*	6.09	1.00	12.19	3.73	**	7.63	3.06	13.55	5.18	*
L1 to NB (°)	26.33	8.02	31.27	7.58		21.95	5.19	32.20	6.25	*	25.24	6.29	34.78	8.55	
L1 to NB (mm)	4.62	1.26	8.32	3.83		3.81	1.40	7.59	4.74		5.96	2.08	8.89	4.00	
L1 to A-Pog (mm)	1.91	3.11	6.21	3.46		3.80	1.75	6.84	3.38		5.41	1.96	8.14	4.49	
Pog to NB (mm)	-3.59	2.27	-5.87	2.42		-3.21	2.54	-6.46	3.84		-2.35	2.94	-6.59	2.97	*
Esthetic line to upper lip (mm)	3.18	1.73	5.62	2.38		1.64	1.40	4.87	2.69	*	2.77	1.72	5.45	3.11	
Esthetic line to lower lip (mm)	4.18	1.31	6.36	2.24		3.52	1.21	6.58	2.04	*	5.70	3.16	7.45	1.45	

DO means distraction osteogenesis, SD, standard deviation; Sig., Significance
Mann-Whitney U test, * $p<0.05$, ** $p<0.01$, *** $p<0.001$

and more deviation of chin point to the affected side (Table 10) than Group 1. Especially, ramus inclination of the affected side and non-affected side of Group 2 at T0 showed reverse tendency when compared with Group 1 (Table 10).

The major effects of DO on Group 1 were lengthening of the ramus height, forward positioning of the mandible (Table 7, 11), increase of the gonial angle, counterclockwise rotation due to decrease of articular angle (Table 8, 12), increase of ramus inclination of the affected side, and improvement of occlusal canting and chin point deviation (Table 10, 14). However, after DO (T1), Group 2 showed still retrognathic mandible and less increase of ramus height (Table 7, 11), clockwise rotation of the mandible due to increase of articular angle (Table 8, 12), and more labioversion of the mandibular incisor and less improvement of lip protrusion to E-line (Table 9, 13), less improvement of occlusal plane canting and chin point deviation (Table 10, 14) than Group 1. Although improvement of the sagittal relationship of the retrognathic mandible was maintained after retention (T2), Group 2 at T2 showed still retrognathic mandible and little growth of ramus height (Table 7), counter-clockwise rotation of the mandible due to decrease of articular angle (Table 8) and more labioversion of the mandibular incisor than Group 1 (Table 9).

Both Group 1 and Group 2 showed significant difference in ramus inclination between the affected and non-affected side before DO (T0), after DO (T1), after retention (T2) (Table 10). Occlusal plane canting showed significant change in Group 1 among before DO (T0), after DO (T1), and after retention (T2). In Group 2, occlusal plane canting showed significant change between before DO (T0) and after retention (T2) (Table 10).

Cephalometric measurements such as ramus height, articular angle, ramus inclination of the affected side, occlusal plane canting, chin point deviation ($p<0.01$), facial convexity angle, Pog-N perpendicular, gonial angle, upper gonial angle ($p<0.05$) showed significant differences during DO (T0-T1) and occlusal plane canting ($p<0.01$), articular angle, upper gonial angle ($p<0.05$) during retention (T1-T2) between the two groups (Table 11-14).

Since Group 2 showed more severe sagittal, vertical, and asymmetric skeletal relationships than Group 1 at T0, effect of DO on Group 2 was less than Group 1 during DO (T0-T1). Group 1 showed more forward positioning of the mandible, more lengthening of the ramus height rather than body length (Table 11), more increase of gonial angle, counterclockwise rotation of the mandible due to decrease of articular angle (Table 12), uprighting of the ramus inclination of the affected side, improvement of occlusal plane canting and chin point deviation (Table 14) than Group 2.

During retention (T1-T2) Group 2 showed counterclockwise rotation of the mandible due to decrease of articular angle and more increase of upper gonial angle (Table 12) than Group 1. Decrease of occlusal plane canting in Group 2 during retention (T1-T2) (Table 10, 14) seems to relate with bilateral cases which occlusal plane canting is not severe and less vertical growth of the ramus of both sides.

Table 10. Comparison of facial asymmetry pattern between Group 1 and 2 before DO (T0), after DO (T1), and after retention (T2).

Measurements	T0					T1					T2				
	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.
	Mean	SD	Mean	SD		Mean	SD	Mean	SD		Mean	SD	Mean	SD	
Ramus inclination of the affected side (°)	83.75	2.27	86.50	1.18	**	86.75	4.04	83.67	4.13	NS	85.50	2.78	84.67	4.93	NS
Ramus inclination of the non-affected side (°)	76.50[a]	1.20	70.67[a]	5.39	*	78.00[b]	6.19	74.67[b]	2.73	NS	76.00[a]	4.21	74.17[b]	2.62	NS
Occlusal plane canting (°)[c,d]	7.13	0.79	5.83	2.21	NS	2.88	1.09	3.83	2.91	NS	4.75	1.95	2.17	1.29	*
Chin point deviation (°)	8.25	2.77	14.17	4.70	*	5.00	1.56	7.33	6.71	NS	7.63	3.47	7.83	4.25	NS

DO means distraction osteogenesis, SD, standard deviation; Sig., Significance; NS : not significant.
Mann-Whitney U test, * $p<0.05$, ** $p<0.01$, *** $p<0.001$
Wilcoxon signed rank test between the affected and non-affected side of Group 1 and Group 2 at each stage, a<0.001, b<0.01.
one-way ANOVA test among T0, T1, and T2 of Group 1 and Group 2, c means that Group 1 showed significant changes in (T0,T1)***, (T1,T2)*, and (T0,T2)**, d means that Group 2 showed significant changes in (T0,T2)*.

Table 11. Comparison of changes in sagittal measurements in facial skeletal pattern between Group 1 and 2 during DO (T0-T1) and during retention (T1-T2)

Measurements	T0-T1					T1-T2				
	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.
	Mean	SD	Mean	SD		Mean	SD	Mean	SD	
SNA (°)	-0.71	0.96	0.30	2.03	NS	-0.43	1.14	-0.47	1.77	NS
SNB (°)	2.46	1.38	2.51	1.27	NS	-0.79	1.83	-0.01	1.26	NS
ANB (°)	-3.17	0.90	-2.21	1.10	NS	0.36	1.80	-0.46	1.56	NS
Facial plane angle (°)	2.70	1.33	2.30	1.45	NS	-0.19	1.67	0.10	1.76	NS
Facial convexity angle (°)	-6.85	1.16	-4.38	2.12	*	-0.60	4.07	-0.97	2.82	NS
Facial angle (Downs) (°)	2.68	1.45	1.92	1.90	NS	-0.16	1.55	0.48	2.32	NS
A-B plane angle (°)	4.34	1.66	3.24	2.10	NS	-0.78	3.04	0.23	2.43	NS
APDI (°)	7.32	1.81	6.33	3.33	NS	-1.26	4.05	-1.69	5.58	NS
Y-axis to SN (°)	-1.50	1.60	-0.59	1.68	NS	1.16	1.42	0.23	1.34	NS
Facial Axis (Ricketts) (°)	2.88	4.08	1.92	3.04	NS	-2.68	3.84	-0.97	1.70	NS
A-N perpendicular (mm)	-0.62	1.04	-0.04	1.93	NS	-0.49	1.96	-0.27	0.99	NS
Pog-N perpendicular (mm)	5.04	3.24	2.77	3.01	*	-1.02	3.51	0.16	4.22	NS
Ramus height (mm)	5.03	6.32	2.79	6.88	**	-6.19	6.54	-0.39	10.89	NS
Man. body length (mm)	0.09	6.58	5.71	9.11	NS	9.40	6.06	16.80	14.13	NS

DO means distraction osteogenesis, SD, standard deviation; Sig., Significance; NS : not significant.

Table 12. Comparison of changes in vertical measurement between Group 1 and 2 during DO (T0-T1) and after retention (T1-T2)

Measurements	T0-T1					T1-T2				
	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.
	Mean	SD	Mean	SD		Mean	SD	Mean	SD	
Articular angle (°)	-11.98	8.33	7.83	9.79	**	7.49	9.43	-8.67	9.71	*
Gonial angle (°)	9.90	8.81	0.07	4.81	*	-4.31	8.03	5.22	10.40	NS
Bjork sum (°)	-0.49	5.41	5.65	9.18	NS	2.19	3.14	-0.73	3.58	NS
Upper Gonial angle (°)	6.75	7.36	-5.18	6.57	*	-3.18	6.23	8.40	7.75	*
Lower Gonial angle (°)	3.15	4.59	0.76	7.13	NS	-1.13	4.99	-3.18	2.47	NS
Post. Facial Height (mm)	4.41	7.45	-0.55	7.11	NS	-0.96	3.40	-2.06	6.19	NS
Ant. Facial Height (mm)	3.91	2.75	3.56	3.50	NS	6.33	6.49	3.97	1.77	NS
Facial Height Ratio (%)	2.10	6.29	-2.22	5.87	NS	-3.76	2.26	-3.48	5.63	NS
SN-GoMe (°)	-0.49	5.41	1.17	3.51	NS	2.19	3.14	-0.39	3.02	NS
Occlusal plane to GoMe (°)	5.44	3.43	5.02	6.72	NS	0.67	2.39	-2.47	5.02	NS
FMA (°)	-0.45	5.18	1.46	4.22	NS	2.15	3.76	-0.79	3.84	NS

DO means distraction osteogenesis, SD, standard deviation; Sig., Significance; NS : not significant.
Mann-Whitney U test, * $p<0.05$, ** $p<0.01$, *** $p<0.001$

Table 13. Comparison of changes in dental and soft tissue measurement between Group 1 and 2 during DO (T0-T1) and after retention (T1-T2)

Measurements	T0-T1					T1-T2				
	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.
	Mean	SD	Mean	SD		Mean	SD	Mean	SD	
IMPA (°)	-6.23	6.08	-2.66	4.15	NS	1.92	8.01	3.01	3.16	NS
FMIA (°)	6.68	8.16	1.20	4.82	NS	-4.06	7.15	-2.22	5.53	NS
L1 to facial plane (mm)	0.28	2.71	-0.34	1.09	NS	1.54	2.30	1.36	1.64	NS
L1 to NB (°)	-4.39	8.25	0.93	3.54	NS	3.30	6.29	2.58	5.33	NS
L1 to NB (mm)	-0.82	2.00	-0.73	1.17	NS	2.16	2.97	1.30	1.88	NS
L1 to A-Pog (mm)	1.90	2.66	0.63	0.88	NS	1.61	2.28	1.30	1.23	NS
Pog to NB (mm)	0.38	2.17	-0.59	2.12	NS	0.86	2.04	-0.13	2.54	NS
Esthetic line to upper lip (mm)	-1.54	1.38	-0.75	1.95	NS	1.13	1.29	0.58	1.23	NS
Esthetic line to lower lip (mm)	-0.66	1.47	0.22	0.86	NS	2.18	3.02	0.88	1.65	NS

DO means distraction osteogenesis, SD, standard deviation; Sig., Significance; NS : not significant.
Mann-Whitney U test, * $p<0.05$, ** $p<0.01$, *** $p<0.001$

Table 14. Comparison of changes in vertical measurement between Group 1 and 2 during DO (T0-T1) and after retention (T1-T2)

Measurements	T0-T1					T1-T21				
	Group 1		Group 2		Sig.	Group 1		Group 2		Sig.
	Mean	SD	Mean	SD		Mean	SD	Mean	SD	
Change in ramus inclination of the affected side (°)	3.00	1.00	-2.83	2.98	***	-1.25	2.89	1.00	0.89	NS
Change in ramus inclination of the non-affected side (°)	1.50	5.48	4.00^a	3.90	NS	-2.00	2.27	-0.50^b	0.45	NS
Change in occlusal plane canting (°)	-4.25	0.89	-2.00	1.18	**	1.88	1.98	-1.67	1.86	**
Change in chin point deviation (°)	-3.25	1.44	-6.83	2.02	**	2.63	3.41	0.50	2.49	NS

SD means standard deviation; Sig., Significance; NS, not significant
Mann-Whitney U test, * $p<0.05$, ** $p<0.01$, *** $p<0.001$
Wilcoxon signed rank test between the affected and non-affected side of Group 2 during distraction (T0-T1) and during retention (T1-T2), a<0.01, b<0.01.

There were no differences in the change of the dental and soft tissue measurement during DO (T0-T1) and during retention (T1-T2) (Table 13).

discussion

Initial skeletal characteristics of Group 2 at T0 (Table 7-10) could be related with different DO results. In Group 2, the pressure from the distraction seems to make the proximal segment of the mandible be rotated clockwise instead of lengthening of the ramus height and forward positioning of the mandible (Table 7-9) and ramus inclination of both sides be swung to the non-affected side (Table 10). These could be related with less improvement of sagittal, vertical, and asymmetric relationships between the maxilla and mandible in Group 2 than in Group 1. Therefore, for case with initial skeletal characteristics such as more retrognathic mandible, smaller ramus height and articular angle, and more vertical growth pattern, delay in early DO treatment might be sometimes the best option to prevent second surgical intervention at a later stage.

After retention (T2) Group 2 showed more relapse tendency due to counterclockwise rotation of the mandible, decrease of articular angle, little growth of the ramus height, and uprighting of ramus inclination of the affected side than Group 1 (Table 7-10). Group 1 showed reverse tendency (Table 7-10).

Although the amounts of increase of SNB and decease of ANB during DO (T0-T1) were similar between both groups (Table 11), the sagittal difference between both groups was still existed due to smaller

ramus height, retrognathic position and clockwise rotation of the mandible in Group 2 (Table 11, 12).

Group 1 showed increase of the ramus height during DO (T0-T1) and decrease of the ramus height after retention (T1-T2) (Table 11). Marquez *et al.* [32] found the distracted side grew less than the non-affected side in a group of younger patients. The "growth" of the mandibular ramus on the affected side after DO is anteroposteriorly and vertically decreased or even vertically resorptive.[35] DO did not result in a permanent and obvious increase in bony volume in all cases.[28] Since the soft tissue matrix does not allow the increased length of the ramus to be "permanent", DO seems not to maintain increase of the ramus height permanently.[32]

Despite overcorrection of the mandibular dental midline to the non-affected side, there was relapse of chin point deviation and ramus inclination of both sides during retention (T1-T2) in Group 1 (Table 14). The reason why Group 2 showed stable result of chin point deviation and occlusal plane canting during retention (T1-T2) (Table 14) seems to be related with bilateral involvement and growth deficiency of the ramus of both sides. These factors seem to be related with asymmetric relapse.

The primary effects of DO on Group 1 are (1) modest increase in the sagittal length of the mandible with improved sagittal pogonion projection and transverse pogonion position, and (2) an improvement in the vertical and sagittal position of the maxillary and mandibular dentition of the affected side. But the vertical lengthening and soft tissue effects are minimal.[32]

On the contrary, the primary effect of DO in Group 2 seems to be dentoalveolar with minimal secondary changes in the mandibular ramus or body. It is because the degree of ramus involvement, tissue deficiency, surgical trauma, and formation of scar tissue could be main causative factors for failure of DO result.

A possible explanation for less effect of DO and the relapse after DO in Group 2 might be that the soft tissue matrix was less capable of reacting to the stretching forces [28, 36] and clockwise rotation of the proximal segment of the mandible (Table 7-9), and swing of ramus of both sides to the non-affected side (Table 10).

This study had some limitations in patient' s number and retention period. To overcome these limitations, it is necessary to study with more patients and longer retention period.

In conclusion, these sagittal, vertical, and asymmetric factors could contribute to the difference of long-term results between two groups. Therefore it is necessary to evaluate these items in diagnosis and treatment planning of the HFM in growing patients when one considers DO.

References

1. Grabb WC. The first and second branchial arch syndrome. *Plast Reconstr Surg* 1965;36:485-508.
2. Poswillo D. Otomandibular deformity: pathogenesis as a guide to reconstruction. *J Maxillofac Surg* 1974;2:64-72.
3. Murray JE, Kaban LB, Mulliken JB. Analysis and treatment of hemifacial microsomia. *Plast*

Reconstr Surg 1984;74:186-99.

4. Pruzansky S. Not all dwarfed mandibles are alike. *Birth Defects*. 1969;2:120-129.
5. Converse JM, Coccaro PJ, Becker M, Wood-Smith D. On hemifacial microsomia: the first and second branchial arch syndrome. *Plast Reconstr Surg* 1973;51:268-79.
6. Rune B, Selvik G, Sarnas KV, Jacobsson S. Growth in hemifacial microsomia studied with the aid of roentgen stereophotogrammetry and metallic implants. *Cleft Palate J* 1981;18:128-46.
7. Ross RB. Lateral facial dysplasia (first and second branchial arch syndrome: hemifacial microsomia). *Birth Defects* 1975;11:51-9.
8. Cohen MM Jr, Rollnick BR, Kaye CI. Oculoauriculovertebral spectrum: an updated critique. *Cleft Palate J* 1989;26:276-86.
9. Figueroa AA, Pruzansky S. The external ear, mandible, and other components of hemifacial microsomia. *J Maxillofac Surg* 1982;10:200-11.
10. Lauritzen C, Munro IR, Ross RB. Classification and treatment of hemifacial microsomia. *Scand J Plast Reconstr Surg* 1985;19:33-39.
11. Kay ED, Kay CN. Dysmophogenesis of the mandible, zygoma, and middle ear ossicles in hemifacial microsomia and mandibulofacial dysostosis. *Am J Med Genet* 1989;32:27-31.
12. Vento AR, LaBrie RA, Mulliken JB. The O.M.E.N.S. classification of hemifacial microsomia. *Cleft Palate Craniofac J* 1991;28:68-76.
13. Obwegeser HL. Correction of the skeletal anomalies of oto-mandibular dysostosis. *J Maxillofac Surg* 1974;2:73-92.
14. Kaban LB, Mulliken JB, Murray JE. Three-dimensional approach to analysis and treatment of hemifacial microsomia. *Cleft Palate J* 1981;18:90-9.
15. Mulliken JB, Kaban LB. Analysis and treatment of hemifacial microsomia in childhood. *Clin Plast Surg* 1987;14:91-100.
16. Kaban LB, Moses MH, Mulliken JB. Surgical correction of hemifacial microsomia in the growing child. *Plast Reconstr Surg* 1988;82:9-19.
17. Vargervik K, Kaban LB. Hemifacial microsomia: diagnosis and management. In: Bell WH, editor. Modern practice in orthognathic and reconstructive surgery. Philadelphia: Saunders; 1992. 1532-60.
18. Horgan JE, Padwa BL, LaBrie RA, Mulliken JB. OMENS-Plus analysis of craniofacial and extracraniofacial anomalies in henifacial microsomia. *Cleft Palate Craniofac J* 1995;32:405-12.
19. Polley JW, Figueroa AA, Liou EJ, Cohen M. Longitudinal analysis of mandibular asymmetry in hemifacial microsomia. *Plast Reconstr Surg* 1997;99:328-39.
20. Huisinga-Fischer CE, Zonneveld FW, Vaandrager JM, Prahl-Andersen B. CT-based size and shape determination of the craniofacial skeleton: a new scoring system to assess bony deformities in hemifacial microsomia. *J Craniofac Surg* 2001;12:87-94.
21. Chang YI, Yang WS, Nahm DS, Kim TW, Baek SH. Analysis of the primary and the permanent teeth dimension in Korean hemifacial micorsomia

patients. *Korea J Orthod.* 2000;30:43-52.

22. Codivilla A. On the means of lengthening, in the lower limbs, the muscles and tissues which are shortened through deformity. 1904. Clin Orthop. 1994;301:4-9.
23. Ilizarov GA. The principles of the Ilizarov method. *Bull Hosp Jt Dis Orthop Inst* 1988;48:1-11.
24. McCarthy JG, Schreiber J, Karp N, Thorne CH, Grayson BH. Lengthening the human mandible by gradual distraction. *Plast Reconstr Surg* 1992; 89: 1-8.
25. Yasui N, Kojimoto H, Shimizu H, Shimomura Y. The effect of distraction upon bone, muscle, and periosteum. *Orthop Clin North Am* 1991;22:563-7.
26. McCarthy JG. The role of distraction osteogenesis in the reconstruction of the mandible in unilateral craniofacial microsomia. *Clin Plast Surg* 1994;21:625-31.
27. Molina F, Ortiz Monasterio F. Mandibular elongation and remodeling by distraction: a farewell to major osteotomies. *Plast Reconstr Surg* 1995;96:825-40.
28. Huisinga-Fischer CE, Vaandrager JM, Prahl-Andersen B. Longitudinal results of mandibular distraction osteogenesis in hemifacial microsomia. *J Craniofac Surg* 2003;14:924-33.
29. Grayson BH, Santiago PE. Treatment planning and biomechanics of distraction osteogenesis from an orthodontic perspective. *Semin Orthod* 1999;5:9-24.
30. Hanson PR, Melugin MB. Orthodontic management of the patient undergoing mandibular distraction osteogenesis. *Semin Orthod* 1999;5:25-34.
31. Tehranchi A, Behnia H. Facial symmetry after distraction osteogenesis and orthodontic therapy. *Am J Orthod Dentofacial Orthop* 2001;120:149-53.
32. Marquez IM, Fish LC, Stella JP. Two-year follow-up of distraction osteogenesis: its effect on mandibular ramus height in hemifacial microsomia. *Am J Orthod Dentofacial Orthop* 2000;117:130-9.
33. Trahar M, Sheffield R, Kawamoto H, Lee HF, Ting K. Cephalometric evaluation of the craniofacial complex in patients treated with an intraoral distraction osteogenesis device: a preliminary report. *Am J Orthod Dentofacial Orthop* 2003;124:639-50.
34. Losken HW, Patterson GT, Lazarou SA, Whitney T. Planning mandibular distraction: preliminary report. *Cleft Palate Craniofac J* 1995;32:71-6.
35. Kearns GJ, Padwa BL, Mulliken JB, Kaban LB. Progression of facial asymmetry in hemifacial microsomia. Plast Reconstr Surg. 2000;105:492-8.
36. Huisinga-Fischer CE, Zonneveld FW, Vaandrager JM, Prahl-Andersen B. Relationship in hypoplasia between the masticatory muscles and the craniofacial skeleton in hemifacial microsomia, as determined by 3-D CT imaging. *J Craniofac Surg* 2001;12:31-40.

교신 저자

서울대학교 치과대학 치과교정학교실 조교수 백승학
서울시 종로구 연건동 28번지 우편번호) 110-744 / 전화: 02-2072-3952 / E-mail: drwhite@unitel.co.kr