



## Dual Midfacial Distraction of Le Fort III minus I and Le Fort I Advancement in Syndromic Craniosynostosis: Extension of application to Younger Ages

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### Abstract

**Objective:** Le Fort III distraction or mono bloc fronto-facial distraction has been widely applied for the midfacial recession in syndromic craniosynostosis patients. Conversely considering the degree of upper and lower half of the midfacial recession, dual midfacial distraction of Le Fort III minus I and Le Fort I advancement in Syndromic craniosynostosis is a useful procedure to reduce morbidity of adolescence and adult patients. Here the authors describe the application of extension to younger patients, exhibiting several case reports.

**Method:** 14 cases of Crouzon disease and 4 Apert syndrome are included in this series. Age ranges 10 to 32 years of age. In three cases with severe maxilla-mandibular discrepancy, mandibular set-back surgery with sagittal splitting osteotomy was associated with. 4 cases of 10-12 years of age, 4 cases of 13-16 years of age, and 6 cases of 17-32 years of age have been distributed. In all cases, after the Le Fort III osteotomy, Le Fort I osteotomy is added. In the upper portion of Le Fort III minus I, the internal device was used, and in the lower half of Le Fort I segment, bone borne distraction using soft wire connected to Halo device is used. Distraction has been completed from 3-5 days after the surgery to the end of gradual distraction of 1 mm per day.

**Result:** The amount of distraction was noticed in 15-20 mm in the upper half of the midface and in 12-18 mm in the lower half of midface. No particular complications were noticed including tooth germ injury at the osteotomy and in the distraction phase. Halo device was able to be removed in around 3-4 weeks.

**Conclusion:** Dual midfacial distraction of Le Fort III minus I and Le Fort I is an ideal technique to apply midfacial recession in syndromic craniosynostosis. This report verified the application to younger ages of 10 to 12 years of aged patients.

**Keywords:** Syndromic craniosynostosis; Dual midfacial distraction; Maxilla-mandibular discrepancy

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### Introduction

Although Monoblocdistraction [1] has currently been the champion in particular for children with the craniofacial deformity in syndromic craniosynostosis, Le Fort III distraction has separately been performed worldwide after the fronto-orbital advancement for the midfacial retrusion in syndromic craniosynostosis [2]. The midface distraction by either Monobloc or Le Fort III advancement induces effectively movement of the midface enbloc. Since the relapse or growth impairment of the midface for children has been noticed after the midfacial distraction, over correction has been postulated for the midfacial distraction even if substantial degree of malocclusion is created. The surgical timing is very important for this procedure, but midfacial distraction can be performed even in any age group. However, Le Fort III distraction alone results malocclusion for elder children to adults. The procedure separating upper and lower portion is good candidates respecting the occlusion for the adolescence and adults. Dual midfacial distraction of separating Le Fort III minus I and Le Fort I portion for the adolescence and adults patients in syndromic craniosynostosis has been an ideal procedure and reported in 2004 [3]. Herein the follow-up series extending much younger patients will be described and the appropriate protocol to reduce the patients' burden by long maintenance of Halo device has been proposed by dual midfacial distraction described above.

**Table 1:** Demography of cases.

No	Age	Sex	Disease	Operative procedure	Device for upper part	Device for lower part	Associated Mandibular set-back
1	26	F	Crouzon	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
2	13	F	Apert	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
3	19	M	Apert	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
4	16	F	Crouzon	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
5	17	M	Crouzon	Le Fort III and I osteotomy	Z-Z type internal	Halo	yes 5 mm
6	32	F	Crouzon	Le Fort III and I osteotomy	Z-Z type internal	Halo	yes 5 mm
7	10	M	Crouzon	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
8	12	F	Crouzon	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
9	12	M	Crouzon	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
10	12	M	Crouzon	Le Fort III and I osteotomy	Halo	Halo	no
11	16	M	Crouzon	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
12	14	F	Apert	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
13	19	M	Crouzon	Le Fort III and I osteotomy	Z-Z type internal	Halo	no
14	18	M	Apert	Le Fort III and I osteotomy	Z-Z type internal	Halo	yes 5 mm

**Table 2:** Detail treated by this procedure.

No	Distraction amount of upper part	Distraction amount of lower part	Retention period of Halo	Postoperative follow up
1	18 mm	13 mm	3 weeks with Halo brace	16 years
2	17 mm	13 mm	3 weeks with face mask	14 years
3	18 mm	14 mm	3 weeks with Halo brace	14 years
4	16 mm	12 mm	3 weeks with Halo brace	13 years
5	17 mm	18 mm	4 weeks with Halo mask	14 years
6	16 mm	13 mm	4 weeks with Halo mask	13 years
7	20 mm	16 mm	4 weeks with Halo mask	9 years
8	20 mm	15 mm	3 weeks with Halo brace	7 years
9	18 mm	14 mm	3 weeks with Halo brace	4 years
10	18 mm	15 mm	4 weeks with Halo mask	3 years
11	16 mm	10 mm	3 weeks with Halo brace	2 years
12	18 mm	15 mm	3 weeks with Halo brace	3 years
13	15 mm	12 mm	4 weeks with Halo mask	2 years
14	16 mm	13 mm	4 weeks with Halo mask	2 years

## Materials and Methods

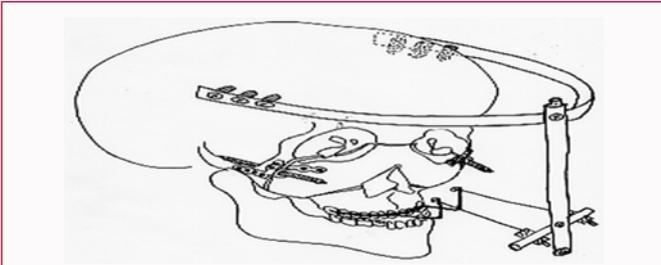
### Demographic data of cases

14 cases of 10 Crouzon disease and 4 Apert syndrome are included in this series. 8 females and 6 males are consisted of. Age ranges 10 to 32 years of age (Table 1). 4 cases of 10-12 years of age, 4 cases of 13-16 years of age, and 6 cases of 17-32 years of age have been distributed. All the patients consulted to the orthodontists and preoperative conference between us was routinely performed. Postoperative continuous manage orthodontic management is definitely required to obtain more favorable occlusion for long term. Although patients with syndromic craniosynostosis may have some degree of airway obstruction, in this series patients complained of no airway problems, and no particular assessment of airway was conducted. In two adult cases, no fronto-orbital advancement for craniosynostosis has been performed previously. In all cases, after the Le Fort III osteotomy, Le Fort I osteotomy is added simultaneously. In the upper portion of Le Fort III minus I portion, internal distraction device (zygoma-zygoma typed device, W. Lorenz, Jacksonville, USA) was attached in the zygomatic bone. In the lower half of Le Fort I segment, bone borne

distraction using soft wire attached to the maxilla and connected to the Halo device.

### Operative procedure and postoperative protocol

Through coronal incision, subciliary, and buccal incision, the subperiosteal undermining is completed and Le Fort III osteotomy was accomplished, and then Le Fort I osteotomy is added. The internal device is attached to the zygoma bilaterally and soft wire piercing to the upper part of the maxilla of Le Fort I portion bilaterally. The well-designed internal device does not disturb Le Fort I osteotomy and attachment of soft wires to Halo (Figure 1). After the scalp closure, Halo device is attached to the temporal area. Soft wires are connected to the Halo (Figure 1). The concept of this procedure is to obtain the preferable occlusion controlled by Halo device and to augment infraorbital area advancing forward using by internal device. The amount and the direction of the distraction in the upper and lower portion is changed according to the severity of respective cases. The distraction of the internal device is ceased after substantial amount of advancement. Then Halo device is taken off to release the physical burden to the patient wearing the Halo device as early as possible.



**Figure 1:** A schema of dual midfacial distraction of Le Fort III minus I and le Fort I, in the upper portion of Le Fort III minus I, internal distraction device is attached and in the lower portion of Le Fort I, Halo external device is attached.

Postoperatively, distraction has started on 3-5<sup>th</sup> day and 1 mm (2 turns) advancement per day was conducted. The internal device applied to the upper portion of the midface is kept in place for consolidation for around six months.

**Results and Complications**

In 3 cases with severe maxilla-mandibular discrepancy, mandibular set-back surgery with sagittal splitting osteotomy was associated with. The amount of mandibular set-back was 5 mm in all of them. In the upper half of Le Fort III minus I portion, distraction amount ranged 15-20 mm. In the lower half of Le Fort I, distraction amount ranged 12-18 mm to obtain pre-planned preferable occlusion (Table 2). After the distraction in the upper and lower portion, Halo device has been taken off and fixed the Le Fort I osteotomy site to the upper portion with a mini-plate at the pyriformis area bilaterally under second general anesthesia. Then the patient can be freed from Halo and internal device exposure. In the distraction phase, no particular unfavorable events including tooth germ injury was noticed in any case. The internal distraction devices for the upper portion and mini-plates for Le Fort I osteotomy site had been removed at 6-12 months

postoperatively. Although postoperative cephalometric assessment wasn't done in any case, no particular visible relapse was exhibited for long. Comparing with the conventional Le Fort III osteotomy with rigid fixation, distraction technique guarantees much more stability postoperatively. Any other complications were noticed for a postoperative period of 2-16 years.

**Representative Cases**

**Case 1**

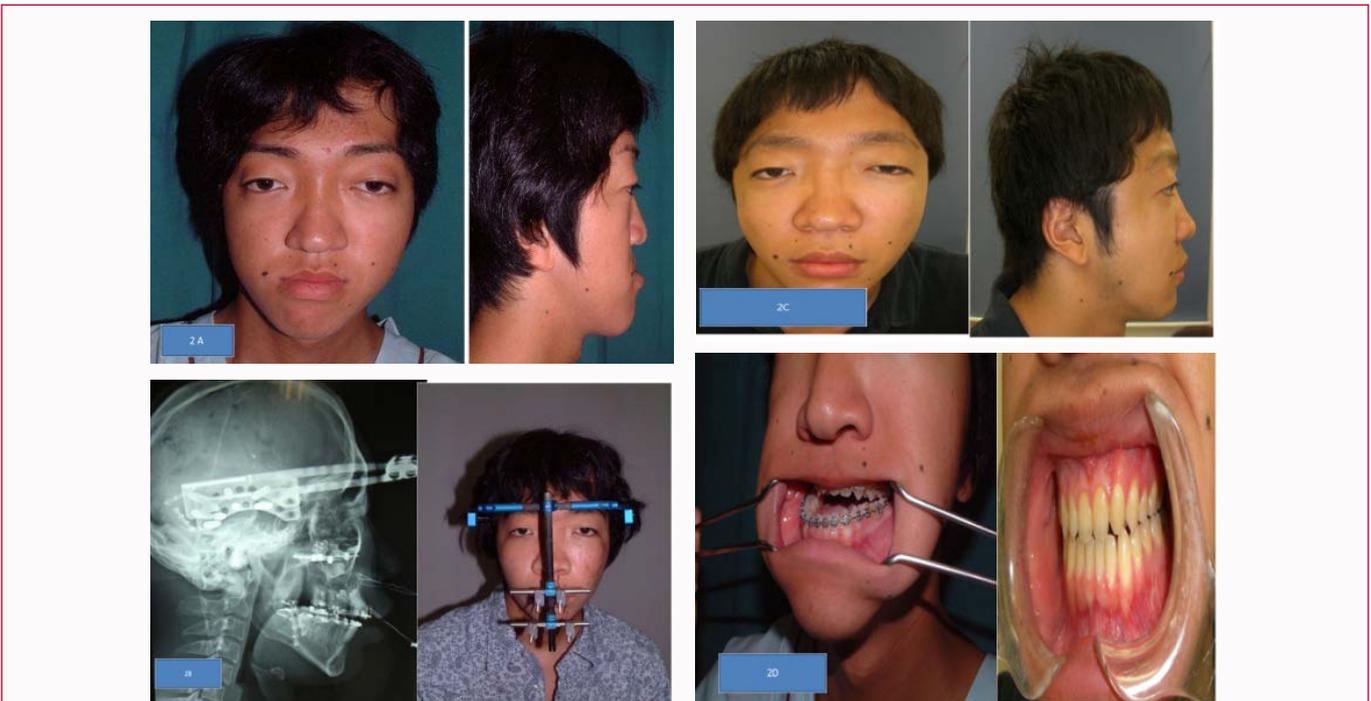
16 years of aged boy, Crouzon disease (Case 5 in Table 1).

Fronto-orbital advancement had been conducted at the age of 1 year at nearby children's hospital. The patient was referred to us to consult the facial deformity with recessed midface at the age of 16 years. Le Fort III advancement was required to repair facial deformity. He was a high school boy, and more than a month of inconvenience wearing Halo device was not tolerated. Le Fort III minus I and Le Fort I portion was planned to advance separately by distraction after the osteotomy. Internal distraction device for the Le Fort III minus I portion, and Halo distraction for Le Fort I portion is planned. At the age of 17 years, the operation was conducted described above and postoperatively 18 mm advancement in the upper portion and 14 mm advancement was obtained by distraction with 1 mm per day. Three weeks later, Halo device was taken off completely and Le Fort I portion was secured to the upper portion with preferable occlusion by two titanium mini-plates. The internal devices were hidden subcutaneously. Postoperative clinical course was uneventful. The midfacial recession was corrected and the Class III malocclusion was repaired. 10 years have passed with good result (Figure 2 A,D).

**Case 2**

9 years of aged boy, Crouzon disease (Case 7 in Table 1).

The patient was referred to us after fronto-orbital advancement



**Figure 2:** Case 1; A: Preoperative frontal view, Preoperative lateral view, midfacial retreat of Crouzon disease obviously revealed. B: During the retention period. Left; Halo device is attached to the skull Right; lateral cephalogram reveals midfacial advancement by distraction. C: 7 years postoperative views (frontal and lateral), significant improvement of the facial configuration noticed. D: Above, Preoperative occlusal view, below, 7 years postoperative occlusal view.



**Figure 3:** Case 2; A: preoperative views (frontal and lateral view), Typical midfacial retreat is noticed. B: left; 3D-CT reveals the midfacial recession is revealed. Right; in the orthopan-tomogram, low positioned dental radix is noticed at the age of 10 years. Le Fort I osteotomy should be carried out to avoid the root injury. C: 1 year postoperative views (frontal and lateral), D: left; 1 year postoperative occlusal view, right; 1 year postoperative lateral 3D-CT view, significant skeletal change is obviously noticed.



**Figure 4:** Case 3; A: preoperative view (frontal and lateral view), typical change of the facial contour is revealed. B: 2 years-postoperative view (frontal and lateral), C: left preoperative lateral 3D-CT, right; 2-years postoperative 3D-CT, preoperative and 2 years postoperative change of the midfacial configuration is obviously noticed.

by distraction in a nearby children’s hospital to repair the craniofacial deformity due to Crouzon disease. The patient was referred to us to consult. The midface revealed to be retarded much with severe exophthalmos and class III malocclusion. Improvement of the midfacial deformity and Class III mal-occlusion was required. At the age of 10 years, Le Fort III minus I and Le Fort I distraction separately

was planned to perform. The orthopan-tomography revealed the low setting of the teeth. Le Fort I osteotomy was assumed to be conducted carefully to avoid the tooth germ injury. The internal device was attached to the upper portion and Halo device was attached to the lower portion, and on the 5<sup>th</sup> day after the surgery, distraction with 1 mm per day was started and finalized to obtain 20 mm in the upper and

15 mm in the lower. On the 28<sup>th</sup> day after the surgery, Halo device was taken off completely and the lower part was secured to the upper part in a preferable occlusion with mini-plates. The midfacial deformity was corrected considerably and the occlusion was improved very well. Five years have passed with good result (Figure 3 A,D).

### Case 3

12 years of aged boy, Crouzon disease (Case 9 in Table 1).

The patient was referred to us to repair the facial recession due to Crouzon disease at the age of 12 years. At the 1 and half years of age, fronto-orbital advancement was conducted in a nearby children's hospital to repair the syndromic craniosynostosis due to Crouzon disease. The midface was retarded much with severe exophthalmos and Class III malocclusion. The patient was required to improve the midfacial deformity and the occlusion. Le Fort III minus I and Le Fort I distraction was planned separately to perform. Le Fort I osteotomy was conducted carefully to avoid the tooth germ injury. The internal device was attached to the upper portion and Halo device was attached to the lower portion, and on the 5<sup>th</sup> day after the surgery, distraction with 1 mm per day was started and finalized to obtain 18 mm in the upper and 13 mm in the lower part. On the 28<sup>th</sup> day after the surgery, Halo device was taken off completely and the lower part was secured to the upper part in a preferable occlusion with mini-plates. The midfacial deformity was corrected very much and the occlusion was improved very well. Four years have passed with remained operative result (Figure 4 A,B).

## Discussion

In syndromic craniosynostosis, midfacial advancement is indispensable. Le Fort III or Monobloc distraction is routinely performed worldwide. However, the surgical timing of these differs depending on the patients' severity and the age of consultation. In children Le Fort III distraction can be planned without strict consideration of the occlusion. Conversely, in adolescence or adults, Le Fort III should be conducted with strict consideration of the occlusion. Hence the first author insisted on the importance of separate distraction of the upper and lower part of Le Fort III distraction for adolescence or adults patients and reported the surgical technique in 2004 [3]. Le Fort III advancement guarantees improvement of the typical midfacial depression revealing dish face. However, occlusal function is extremely important for adolescence and adults. The upper and lower part of Le Fort III portion should be advanced separately in either one-staged advancement or gradual distraction. Halo distraction can induce large amount of advancement by distraction, but long period of consolidation for 2-3 months

results physical and mental burden to patients. As early as possible detachment of Halo can reduce the mental load to the patients. Halo device can be limited to Le Fort I lower part, and consolidation of Halo can be reduced 3 weeks. From this point of view, the upper part of Le Fort III minus I by distraction is better secured with internal devices.

Since all our 14 patients visited us to repair the facial unbalance, they didn't complain of the airway obstruction. They came to us rather late in childhood, adolescence, and adults. No particular evaluation of apnea-hypopnea index was assessed for any of them. In this series, mandibular set-back was associated with in 2 cases. Large maxillo-mandibular discrepancy is sometimes encountered in syndromic craniosynostosis, and mandibular set-back can be conducted simultaneously to correct the facial unbalance. Although airway problem is often recognized in syndromic craniosynostosis, it is basically induced by midfacial retrusion. Mandibular set-back left no airway problems postoperatively in 2 cases.

Although the precise assessment of occlusion may not be feasible, post-distraction occlusion is extremely important for 9-10 years of aged children. Le Fort I distraction to obtain more preferable occlusion makes sense. Long term orthodontic care is required for postoperative period to observe better occlusion. Complete exchange from baby to permanent teeth depends on the patients. Surgical avoidance to injure teeth germ can be accomplished by careful osteotomy in early adolescence. In our case series, the surgical timing of Le Fort I osteotomy can be lowered down to 10 years of age case by case, although careful follow up is required. No particular impairment of tooth growth was noticed up to 3-7 years postoperatively.

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