A Osteogenesis Distraction Device Enabling Control of Vertical Direction for Syndromic Craniosynostosis

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Background: We have developed a hybrid facial osteogenesis distraction system that combines the advantages of external and internal distraction devices to enable control of both the distraction distance and vector. However, when the advanced maxilla has excessive clockwise rotation and shifts more downward vertically than planned, it might be impossible to pull it up to correct it. We invented devices attached to external distraction systems that can control the vertical vector of distraction to resolve this problem. The purpose of this article is to describe the result of utilizing the distraction system for syndromic craniosynostosis.

Methods: In addition to a previously reported hybrid facial distraction system, the devices for controlling the vertical direction of the advanced maxilla were attached to the external distraction device. The vertical direction of the advanced maxilla can be controlled by adjustment of the spindle units. This system was used for 2 patients with Crouzon and Apert syndrome.

Results: The system enabled control of the vertical distance, with no complications during the procedures. As a result, the maxilla could be advanced into the planned position including overcorrection without excessive clockwise rotation of distraction.

Conclusion: Our system can alter the cases and bring them into the planned position, by controlling the vertical vector of distraction. We believe that this system might be effective in infants with syndromic craniosynostosis as it involves 2 osteotomies and horizontal and vertical direction of elongation can be controlled. *(Plast Reconstr Surg Glob Open 2014;2:e113; doi: 10.1097/GOX.0000000000000060; Published online 26 February 2014)*

Distraction techniques with a variety of external and internal distraction systems have been successfully used to advance the midface.[1–6] However, external distraction devices generally cannot deliver adequate distraction forces for lengthening; as a result, the distraction achieved is limited.[7,8] On the other hand, internal distraction devices do not allow control of the post-fixation distraction vector.[9] We have developed a hybrid facial distraction system (HFDS) leveraging the advantages of external and internal distraction devices.[10] However, when the advanced maxilla undergoes excessive clockwise rotation and is shifted more downward vertically than the planned position, it might be impossible to pull it back into the planned position. To resolve this problem, we invented devices that can be attached to HFDS and

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control the vertical vector of distraction. The purpose of this article is to describe the result of application of the distraction system for syndromic craniosynostosis.

METHODS

When the advanced maxilla undergoes excessive clockwise rotation and is shifted more downward vertically than the planned position, it might be possible to pull it back into the planned position by vertical distraction device. We invented devices that can control the vertical vector of distraction (Figs. 1A–C). First, a previously reported HFDS, composed of a conventional halo-type external distraction device (KLS-Martin)[4] and an internal distraction device with a 3-dimensional adjustable angle to interlock with the halo-type distraction device, was attached.[10] Then, the new devices, which can control the vertical direction of the advanced maxilla, were attached to the external distraction device. For this, screws were fixed above the bilateral upper primary molars or first molars and spindle units were attached on contact with attachment on the side of external distraction device (Figs. 1D, E). Then, surgical wires were attached to both the screws and spindle units after they were penetrated through the scalp. The vertical direction of the advanced maxilla can be controlled by the adjustment of spindle units.

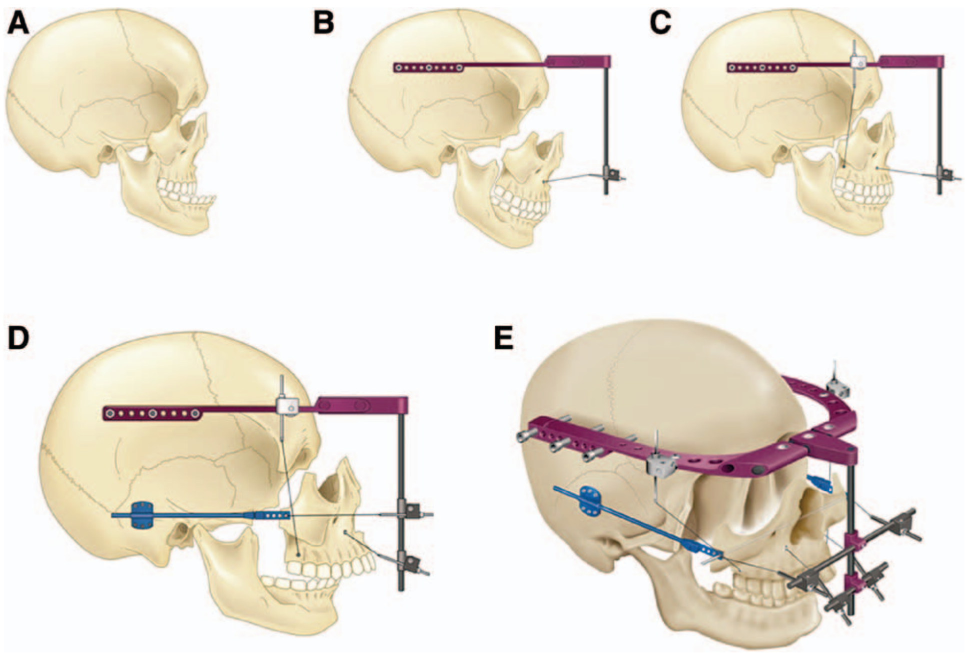

Fig. 1. External distraction devices enabling control of distraction vertical vector and maxillary distraction technique with internal and external devices. A, Before procedure. B, Orbital and facial height increased by excessive clockwise rotation of distraction. C, Adequate orbital and facial height achieved by vertical distraction device. D and E, The external distraction device enables control of the distraction vector via surgical wires. At the same time, the internal distraction device has an adjustable angle which moves up and down according to the direction of the wires. The angle of the internal distraction device's fixation position on the temporal bone can be altered by 5–15 degrees from side to side. Additionally, the device which can control the direction of the advanced maxilla vertically is attached to the external distraction device. Then, surgical wires are attached to the screws and spindle units after penetrating through the scalp and passed through the inside of the zygomatic arch. The vertical direction of the extended maxilla can be controlled by adjustment of the spindle units.

CASE REPORTS

Patient 1

A 12-year-old male patient with Crouzon syndrome (Fig. 2). The patient underwent Le Fort III osteotomy and our systems were placed. Internal and external devices for this system were attached, and a total of 6 surgical wires were attached to spindle units of the external devices. Although the maxillary position showed slight anticlockwise rotation at the distraction phase, it was corrected by the vertical wires. Immediately after the overcorrection, the external devices were removed. After an additional 3 months, all devices including the internal devices were removed. The 6-month postoperative results indicated that point Or and A were advanced by 22 and 23 mm, respectively (Fig. 3A).

Patient 2

A 10-year-old female patient with Apert syndrome (Fig. 4). She underwent an osteotomy with a bone incision similar to Le Fort II through the

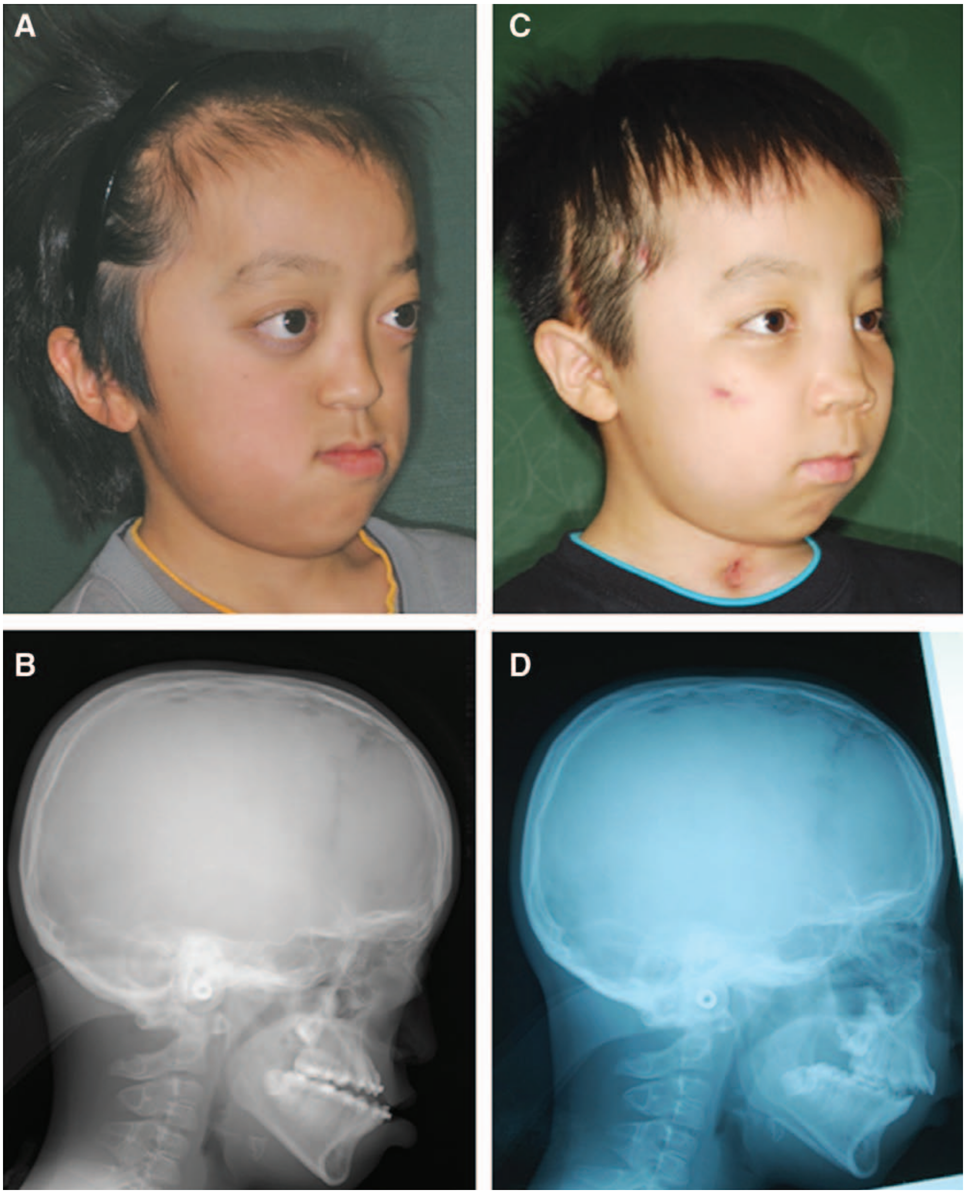

Fig. 2. Patient 1: A 12-year-old male patient with Crouzon syndrome. A, Preoperative view. He had severe maxillary retrusion and a class III soft-tissue profile. B, Preoperative lateral cephalogram. C, Six months postoperatively. After 4 months of advancement, all devices including the internal devices were removed. D, Postoperative lateral cephalogram. Point Or and A were advanced by 22 and 23 mm, respectively.

Patient 1	Movement Distance (mm)	Downward Angle (°)	Patient 2	Movement Distance (mm)	Downward Angle (°)
Point N	17	26		10	46
Or	22	0		18	14
Point A	23	13		16	40

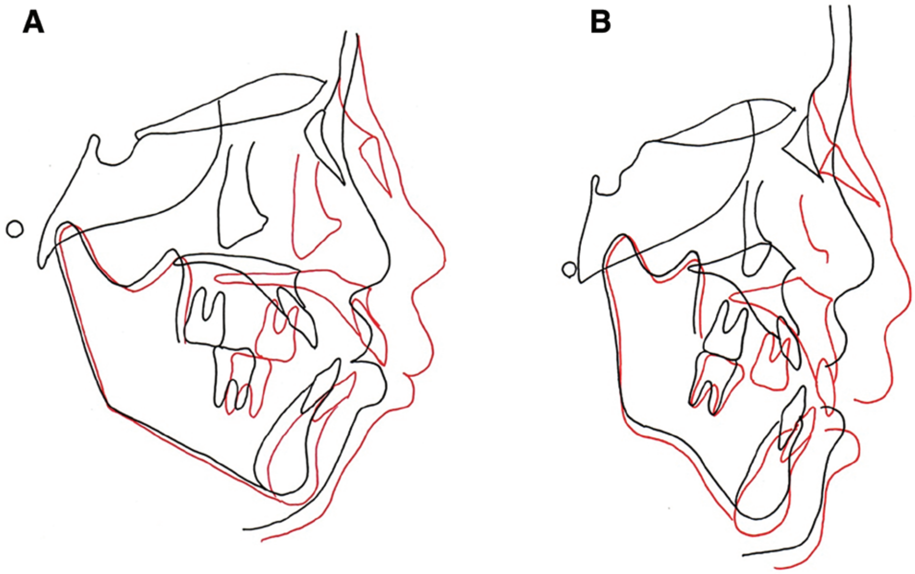

Fig. 3. Lateral cephalometric measurements. Patient 1 (A) and patient 2 (B). The predistraction (black) and postdistraction (red) lateral cephalogram tracings of a single patient superimposed on the sella and oriented along the anatomy of the anterior cranial base. The midface shows differential advancement at point N, the orbitale, and point A.

inside of infraorbital foramen in addition to Le Fort III for the correction of midfacial retrusion. Using this system, the direction of the zygomatic bones was controlled by 2 wires, which were passed through a 2-mm transmaxillary Kirschner wire, and the middle maxilla was controlled by 2 wires which were passed through a miniplate attached to the inferior border of the anterior nasal aperture and another 2 wires which were passed from the mouth through scalp. Immediately after advancement, the external devices were removed. After an additional 3 months, the rest of all devices were removed. The 1-year postoperative results indicated that point Or and A were advanced by 18 and 16 mm, respectively (Fig. 3B).

DISCUSSION

We developed a flexible facial distraction system in which new devices that control the vertical direction of the advanced maxilla can be added to a previously reported HFDS. We believe that this system would allow easy and reproducible overcorrection in distraction osteogenesis and the main indications of this system would be the cases that need a distraction distance of 20 mm over or multiple osteotomy.

External distraction allows for control and adjustment of horizontal and vertical movements of the midface after the distraction process has been initiated and favors skeletal movements with minimal dental changes.

Although the external distraction devices help to control the distraction vector, the possible distraction distance may be generally limited.[11] Our system can help to bring the cases into the planned position, because the vertical vector of distraction can be controlled for an adjuvant of the external distraction. And also, it is possible for this system to have early removal of external devices because the internal devices can prevent relapse. If the wires of external devices were used to change the vector of more than 10 degrees during advancement, the devices should not be removed immediately after advancement.

The Or and point A are evaluated by cephalometric analysis to determine different distraction vector positions for infant syndromic craniosynostosis. The vectors of Or and point A in Apert syndrome are commonly planned different from those of Crouzon

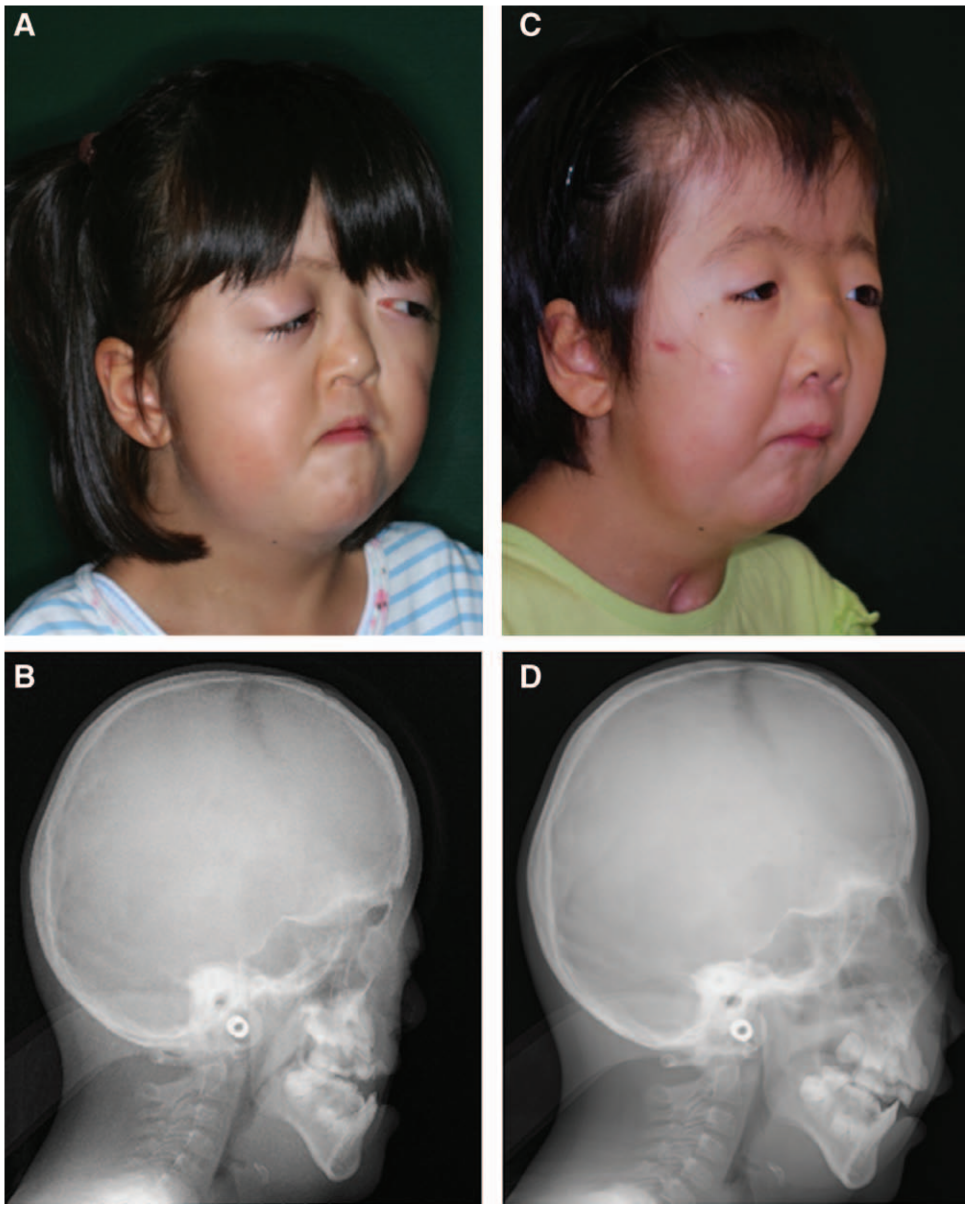

Fig. 4. Patient 2: A 10-year-old female patient with Apert syndrome. A, Preoperative view. She had severe maxillary retrusion and a class III soft-tissue profile. B, Preoperative lateral cephalogram. C, One year postoperatively after osteotomy with a bone incision similar to Le Fort II but through the inside of the infraorbital foramen with Le Fort III. D, Postoperative lateral cephalogram. Point Or and A were advanced by 18 and 16 mm, respectively.

syndrome before operation because of short facial height. That is, the vector of point A is set more downward than the vector of Or.[11] But, it is difficult to advance the maxilla to the planned position. The advancement to the planned vector of Or and point A differs when the distraction distance is greater. Correction of these cases may require additional procedures, such as Le Fort II osteotomy in addition to Le Fort III surgery.[12–14] Advancement of the maxilla to the planned position could be controlled by combining the 2 osteotomies and this system in Apert syndrome.

CONCLUSIONS

Our system can alter the cases and bring them into the planned position by controlling the vertical vector of distraction. We believe that this system might be effective in infants with syndromic craniosynostosis as it involves 2 osteotomies, and horizontal and vertical direction of elongation can be controlled.

PATIENT CONSENT

Parents or guardians provided written consent for the use of the patients' image.

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REFERENCES

1. Britto JA, Evans RD, Hayward RD, et al. Maxillary distraction osteogenesis in Pfeiffer's syndrome: urgent ocular protection by gradual midfacial skeletal advancement. *Br J Plast Surg.* 1998;51:343–349.
2. Chin M, Toth BA. Le Fort III advancement with gradual distraction using internal devices. *Plast Reconstr Surg.* 1997;100:819–830; discussion 831.
3. Molina F, Ortiz Monasterio F, de la Paz Aguilar M, et al. Maxillary distraction: aesthetic and functional benefits in cleft lip-palate and prognathic patients during mixed dentition. *Plast Reconstr Surg.* 1998;101:951–963.
4. Polley JW, Figueroa AA. Management of severe maxillary deficiency in childhood and adolescence through distraction osteogenesis with an external, adjustable, rigid distraction device. *J Craniofac Surg.* 1997;8:181–185; discussion 186.
5. Swennen G, Colle F, De May A, et al. Maxillary distraction in cleft lip palate patients: a review of six cases. *J Craniofac Surg.* 1999;10:117–122.
6. Cohen SR. Craniofacial distraction with a modular internal distraction system: evolution of design and surgical techniques. *Plast Reconstr Surg.* 1999;103:1592–1607.
7. Gosain AK, Santoro TD, Havlik RJ, et al. Midface distraction following Le Fort III and monobloc osteotomies: problems and solutions. *Plast Reconstr Surg.* 2002;109:1797–1808.
8. Cohen SR. Midface distraction. *Semin Orthod.* 1999;5:52–58.
9. Polley JW, Figueroa AA. Rigid external distraction: its application in cleft maxillary deformities. *Plast Reconstr Surg.* 1998;102:1360–1372; discussion 1373.
10. Kobayashi S, Nishiouri T, Maegawa J, et al. A novel craniofacial osteogenesis distraction system enabling control of distraction distance and vector for the treatment of syndromic craniosynostosis. *J Craniofac Surg.* 2012;23:422–425.
11. Shetye PR, Boutros S, Grayson BH, et al. Midterm follow-up of midface distraction for syndromic craniosynostosis: a clinical and cephalometric study. *Plast Reconstr Surg.* 2007;120:1621–1632.
12. Hopper RA, Prucz RB, Iamphongsai S. Achieving differential facial changes with Le Fort III distraction osteogenesis: the use of nasal passenger grafts, cerclage hinges, and segmental movements. *Plast Reconstr Surg.* 2012;130:1281–1288.
13. Dai J, Wang X, Yu H, et al. Simultaneous Le Fort I, II, and III osteotomies for correction of midface deficiency in Apert disease. *J Craniofac Surg.* 2012;23:1391–1395.
14. Paliga JT, Goldstein JA, Storm PB, et al. Monobloc minus Le Fort II for single-stage treatment of the Apert phenotype. *J Craniofac Surg.* 2013;24:596–598.