ORIGINAL ARTICLES

Evaluation of Maxillary Permanent Molars in Patients With Syndromic Craniosynostosis After Monobloc Osteotomy and Midface Advancement With Rigid External Distraction (RED)

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Objective: **This retrospective study was conducted to analyze changes in the maxillary permanent molars after monobloc advancement with rigid external distraction (RED).**

Setting: **University hospital–based craniofacial center.**

Materials and Methods: **Fourteen patients, three in primary, eight in mixed, and three in permanent dentition underwent monobloc advancement with RED. After a latency period of 6 days, distraction was carried out for 18 days. Lateral cephalometric radiographs were taken before surgery (T1) and an average of 3.72 months after the removal of the distractor (T2). Panoramic radiographs were taken at T1, T2, and T3 (an average of 14.87 months after RED removal), to search for surgical tooth trauma, arrested crown/root development, impaction, tooth germ displacement, dilacerations, and other possible dental abnormalities. Vertical and horizontal displacement and angulations of the permanent maxillary molars were evaluated before and after surgery.**

Statistics: **A paired *t* test was used to analyze significant changes in molar position after distraction.**

Results and Conclusions: **Distraction created posterior arch length with significant horizontal forward movement of the first and second molars ($p < .05$) and minimal vertical displacement ($p > .05$). The procedure disrupted the development of one of the first molars, three of the second molars, and two of the third molars. Incidence of molar damage was increased in patients operated on during primary dentition. Careful surgical technique during pterygomaxillary disjunction, especially in young children, and long-term radiographic follow-up of maxillary molars is strongly recommended.**

KEY WORDS: *midface advancement, molar development, monobloc osteotomy, rigid external distraction, syndromic craniosynostosis*

Children with craniofacial malformations present a wide spectrum of problems and should be treated by an interdisciplinary team. Among the different craniosynostosis syndromes described, the best known are Apert, Pfeiffer and Crouzon syndromes. The premature closure of craniofacial sutures in craniosynostosis syndromes may constitute a serious clinical problem warranting surgical intervention in infancy to avert pathological effects on brain and eye development and prevent the respiratory, feeding, and speech problems usually seen in patients with craniofacial synostosis.

Monobloc advancement has been used to correct midface deficiency in craniofacial syndromes (Ortiz-Monasterio et al., 1978; Ortiz-Monasterio and Fuente del Campo, 1979). This procedure and the Le Fort III osteotomy have been associated with molar tooth damage in the tuberosity region. Disruption of a normal tooth bud at any stage of development may have a significant effect on its final form and function as a component of the masticatory system (Santiago et al., 2005). Recently, monobloc advancement utilizing rigid external distraction (RED) has been successfully used for the correction of severe midface deficiency in cleft and craniofacial syndromes (Figueroa and Polley, 2007; Figueroa et al., 2001; Jensen et al., 2007; Polley and

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Figueroa, 1998; Polley et al., 1995; Satoh et al., 2003). In theory, the gradual nature of this process and the deposition of viable bone in the pterygomaxillary region could have a less deleterious effect and facilitate the eruption of the permanent molars. However, the effect of monobloc distraction on posterior maxillary teeth is unknown. Therefore, the purpose of this retrospective radiographic study is to analyze the effect of the monobloc osteotomy and the advancement with rigid external distraction (RED) on erupted and non-erupted maxillary permanent molars.

Methods

Fourteen patients who underwent monobloc advancement with a RED device were included in this study. There were 10 females and four males: 10 with Crouzon syndrome, two with Pfeiffer syndrome, and two with Apert syndrome. Eight of the 14 patients had a previous failed attempt to advance the midface by traditional monobloc advancement at other institutions.

All patients had a maxillary intraoral stainless steel wire splint with removable external traction hooks prepared before surgery and attached to the second primary molars or the first permanent molars through orthodontic bands (Figueroa and Polley 2006a, 2006b). In addition, at surgery the splint was further secured with circumdental wires around the maxillary primary or permanent canines. All patients had complete mobilization of the monobloc segment through a coronal incision and careful pterygomaxillary disjunction with an intraoral Le Fort I approach. No attempt was made to advance the osteotomized segment intraoperatively. After a latency period of 5 to 7 days, distraction was commenced at the rate of 1 to 1.5 mm per day. Subsequent to skeletal correction, the halo was left in place an average of 6 weeks. No major peri- or postoperative complications were observed in this group of patients.

The patients were subdivided into three groups according to age and dental development to allow better understanding and comparison of the data relative to the developmental stage in which the surgery was performed. Three patients were in the primary (4.15 to 5.31 years old), eight in the mixed (6.0 to 10.83 years old), and three in the permanent dentition stage (14.25 to 21.49 years old).

Panoramic Radiographic Analysis

Panoramic radiographs were taken before surgery (T1) and at 3.72 months (T2) and 14.87 months (T3) after the external distractor device was removed. The radiographs were used to search for (1) surgical tooth trauma, (2) arrested crown/root development, (3) impaction, (4) tooth germ displacement, (5) dilacerations, and (6) other possible dental abnormalities. The different stages of normal crown and root development were evaluated (Fig. 1) (Nolla, 1960) to determine developmental arrest during the follow-up period.

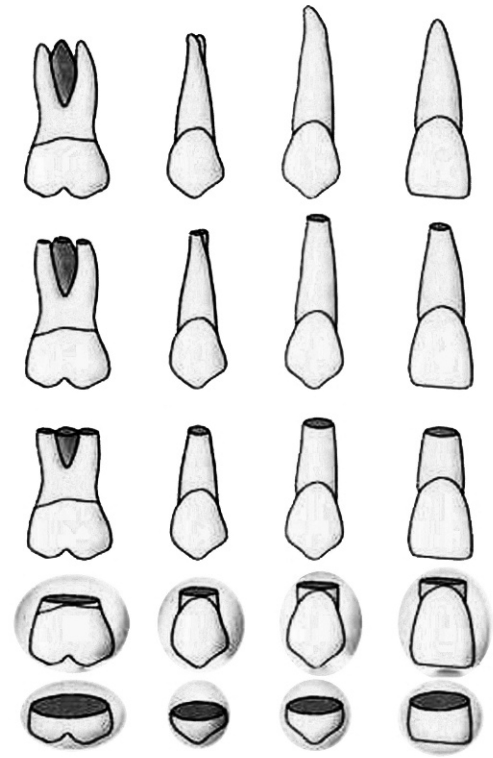

FIGURE 1 **Different stages of normal crown and root development (modified from Nolla, 1960) from partially developed crown (Stage 1, bottom) to fully developed crown and root (Stage 5, top). Stage 5 = fully developed root, Stage 4 = partially developed root (> 2/3), Stage 3 = partially developed root (2/3 a 1/2), Stage 2 = fully developed crown, Stage 1= partially developed crown.**

Lateral Cephalometric Radiographic Analysis

Lateral cephalometric analysis was performed with radiographs taken before surgery (T1) and an average of 3.72 months after distraction (T2). For each subject, two lateral cephalograms were traced (T1 and T2) by hand on acetate paper by the same examiner. Cephalometric analysis was used to measure the horizontal and vertical displacement of the first and second molars and their axial inclination before (T1) and after distraction (T2).

Seven cephalometric anatomic landmarks were used for the cephalometric analysis: opisthion (OP), defined as the most posterior point on the posterior margin of the foramen magnum (Miyashita, 1996); basion (BA), defined as the most inferior point on the anterior margin of the foramen magnum (Miyashita, 1996); sella (S), the center of the hypophyseal fossa; the upper first molar distal end (M1); upper first molar mesiobuccal cusp most inferior point (M1′); upper second molar distal end (M2); upper second molar mesiobuccal cusp most inferior point (M2′) (Fig. 2). Op, BA, and S were the landmarks used to draw

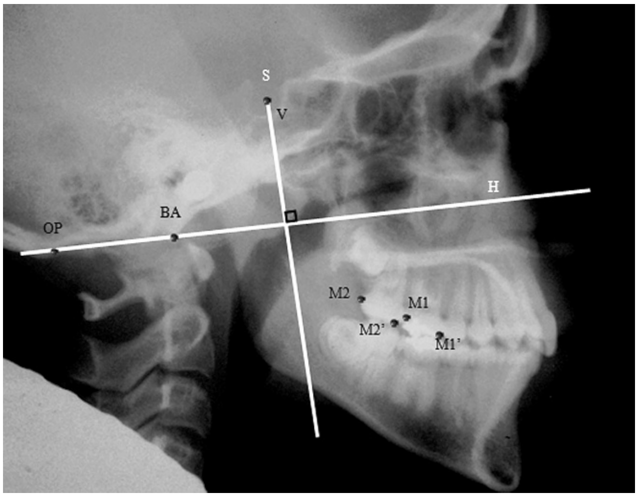

FIGURE 2 Cephalometric landmarks: OP, BA, S, M1, M1′, M2, M2′. Horizontal (H) and vertical (V) reference lines.

horizontal (H) and vertical (V) reference lines. The V line went perpendicular to the H line through S (Fig. 2).

To analyze the molar vertical displacement after monobloc advancement, lines from the most anterior and occlusal point of the mesiobuccal cusp on the upper first and second (M1′ and M2′) molars perpendicular to the H reference line were traced to measure the vertical change in millimeters before (T1) and after distraction (T2) (Fig. 3A and 3B).

To analyze the molar horizontal displacement, lines from the distal end of the first and second (M1 and M2) molars perpendicular to the V reference line were traced to measure the horizontal change in millimeters before (T1) and after distraction (T2) (Fig. 3A and 3B).

The molar angular axial inclination was also evaluated by tracing a line through the long axis of first and second molars in relation to the horizontal reference line (Fig. 4A and 4B). Descriptive statistical analyses with paired *t* test ($p < .05$) were used to analyze the data. This study was approved by the Institutional Review Board office of Rush University.

Results

Panoramic Radiographic Analysis

Of all the first permanent molars evaluated in patients operated in the primary, mixed or permanent dentition (n = 24), 14 advanced one developmental stage during follow-up. It could be observed during the period of this study that one first permanent molar had arrest of crown/root development. Of all the first molars available in the sample (n = 24), one (4.1%) became impacted and was not erupting into its expected position (Fig. 5).

Of the second permanent molars on patients operated in the primary or mixed dentition (n = 17), 13 advanced one developmental stage (Fig. 6A and 6B), and one may have arrested crown/root development (Fig. 7A through 7C), but longer follow-up is required. Three (23.5%) unerupted second permanent molars were disturbed after surgery (T2). These occurred in two patients (average age = 4.58 years) who were in primary dentition at the time of surgery. After distraction, interdental spaces could be seen between the maxillary permanent molars in most patients operated in the primary and mixed dentition (Fig. 8A and 8B).

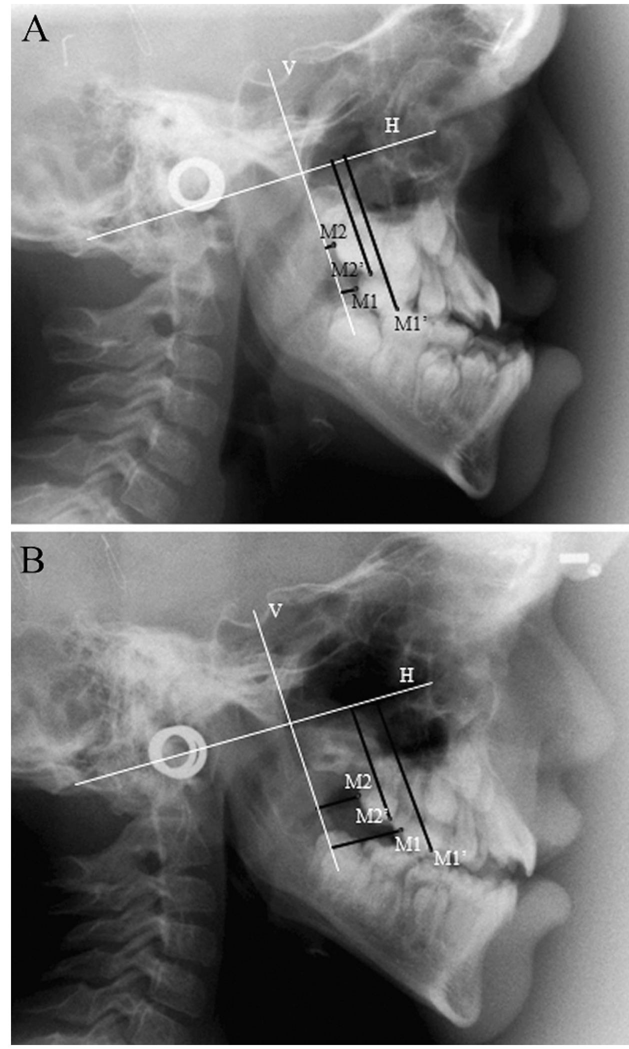

FIGURE 3 Measurement of vertical and horizontal displacement of the first and second permanent molar. A: Before distraction. B: After distraction.

It was possible to evaluate only five third molars. Two had extreme tooth germ displacement. One had anterior and one had posterior or distal movement (Fig. 8A and 8B). Three third molar tooth germs were completely disrupted.

Cephalometric Radiographic Analysis

Molar Horizontal and Vertical Displacements

Changes in molar horizontal and vertical position are shown in Table 1 and Figure 3.

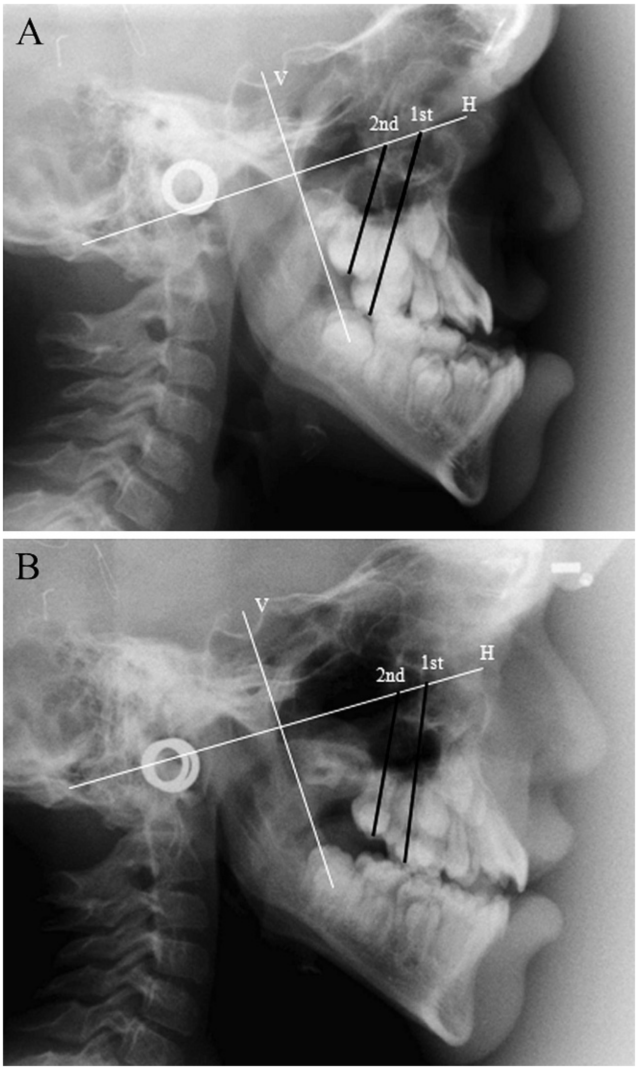

FIGURE 4 Measurement of first and second permanent molar axial inclination. A: Before distraction. B: After distraction.

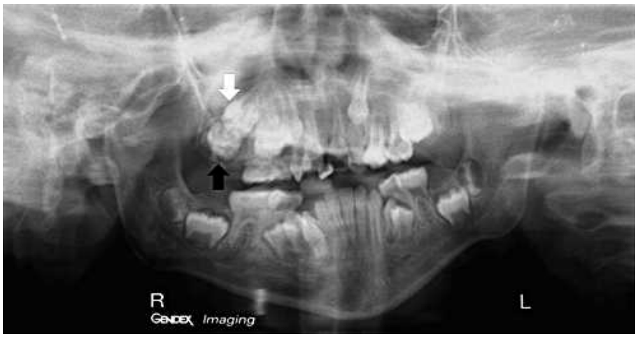

FIGURE 5 Panoramic radiograph on a patient in the earl transitional dentition stage after monobloc distraction with RED. Note impacted first molar (white arrow) and a second molar with extreme displacement (black arrow).

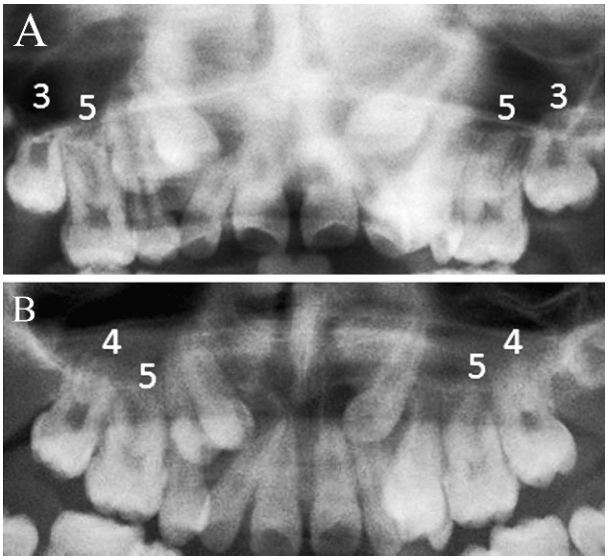

FIGURE 6 Follow-up of crown and root development. Advancement in one developmental stage of a second permanent molar in a 10-year-old patient. A: Before surgery. B: 7 months after surgery.

First and Second Molars Axial Inclination

The axial inclination of the first and second molars relative to the horizontal reference plane increased by 3 and 18 degrees, respectively ($p > .05$) (Figures 4A and 4B). Third molars were not cephalometrically evaluated due to small sample size (n = 5).

Discussion

Before distraction osteogenesis was introduced, most patients with severe craniofacial synostosis were treated with a monobloc or a Le Fort III advancement. The potential disadvantage of this approach is that it requires an acute advancement of the bone segment with increased risk for infection and morbidity and the need for bone grafting and rigid fixation for stabilization. The acute advancement, the placement of bone grafts, and rigid fixation could increase the risk for tissue disruption around the pterygomaxillary region that could lead to dental damage, especially in those molars that are unerupted and in close proximity to the area of the required pterygomaxillary disjunction. Midface distraction osteogenesis after complete monobloc osteotomy allows for gradual and controlled advancement with less disruption or need for secondary surgical manipulation of the osteotomized segment. Advancement and extreme mobilization of the bone segment is not required in the operating room, eliminating the need to use disimpaction forceps, which may have a traumatic effect on the maxillary tuberosity and the developing molars contained within it.

In this study, changes in the maxillary permanent molars after monobloc advancement with RED were analyzed. Our goal was to investigate whether monobloc advance-

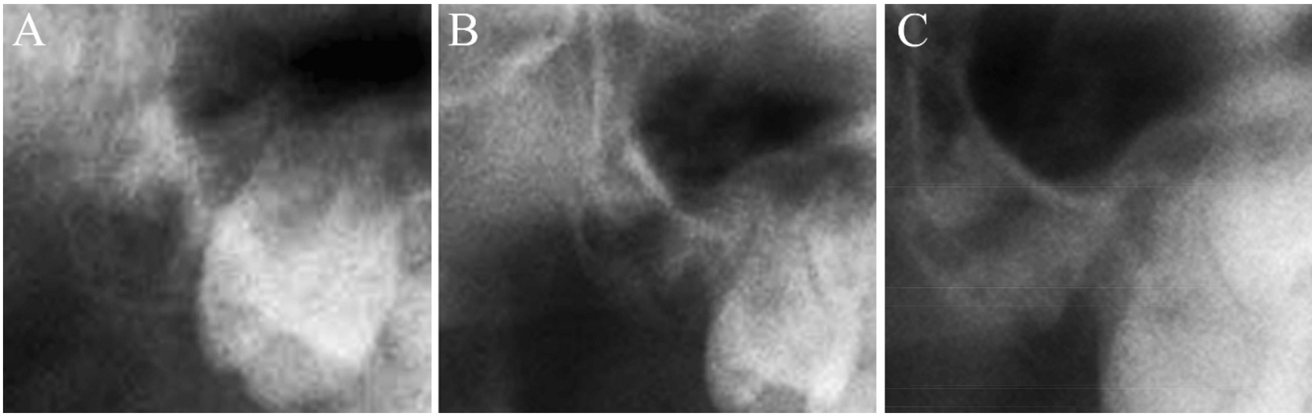

FIGURE 7 Example of a second permanent molar crown with development arrest during the postsurgical follow-up period. A: 2 months. B: 4 months. C: 8 months.

ment with RED was a safer surgical protocol in regards to preservation and integrity of the maxillary molars than conventional Le Fort III and monobloc advancements (Jensen et al., 2007; Santiago et al., 2005).

Of the 14 patients evaluated in this study, eight had experienced a previous conventional attempt at midface advancement elsewhere before distraction osteogenesis. Therefore, some of these patients were already missing teeth or had some form of surgical dental injury. In this investigation, only viable teeth present before the distraction process were analyzed (24 first permanent molars, 17 second permanent molars, and five third molars). Therefore, it should be recognized that the amount of teeth does not correspond to the number of teeth that would be expected if some of the patients had not been previously operated. In addition, third molars buds were not yet developed or visible in six patients in primary and mixed dentition at the time of the first evaluation before surgery.

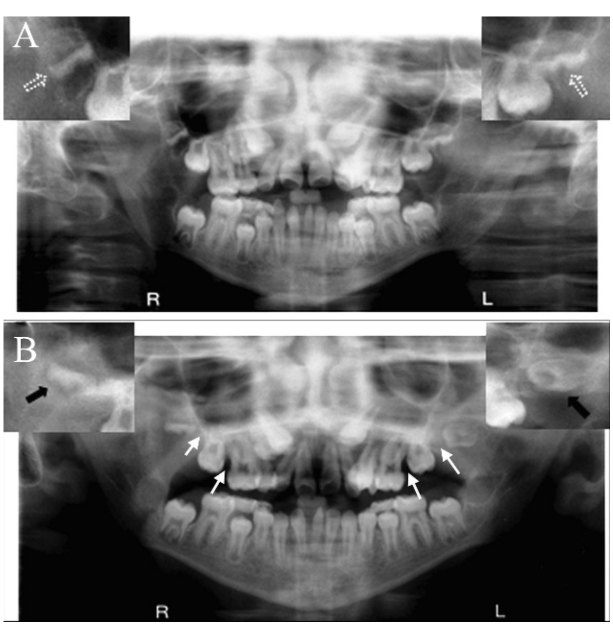

FIGURE 8 Panoramic radiographs of a patient in the late transitional dentition stage. A: Before distraction. B: After distraction. Note creation of space between the maxillary molars after distraction (white arrows). The extreme rotation of the maxillary third molars is also noted. Compare presurgical position (dotted arrows) to postoperative position (black arrows).

Follow-up panoramic radiographs (T2 and T3) ranged from 3.72 (T2) to 14.87 months. The dental development in deciduous, mixed and permanent dentition was studied for any disruptions in development. It was observed that 14 of 24 first permanent molars and 13 of 17 permanent second molars advanced at least one developmental stage during the study period. It should be noted that most of the first molars were fully developed at the time of surgery (Fig. 6A and 6B) and kept their integrity during the follow-up period. Only 1 of 24 first molars had arrested crown and root development, which corresponds to 4.1% of 24 teeth. From the 17 second molars analyzed, three had developmental disruption after the monobloc advancement with RED. One had signs of arrested crown and root development (Fig. 7A through 7C). If damage of this unerupted tooth is considered, 23.5% of the viable permanent second molars were affected by surgery or distraction. These data should be carefully interpreted as 11 second molars were missing before the monobloc distraction surgery. Santiago et al. (2005) found that an early Le Fort III procedure (around 5 years old) did not affect first molar eruption in most cases; however, in 77% of patients the eruption pattern of the second permanent molars was severely compromised or disrupted. An obvious clinical improvement was noted when distraction was used rather than traditional acute advancement. In our sample, only 24% of the second molars had postoperative eruption and/or developmental alterations. Although the surgery protocol improved with distraction, refinements are still needed to preserve the integrity of the dentition, especially when surgery is performed in deciduous or early mixed dentition. Surgeons need to carefully complete the pterygomaxillary disjunction and, in certain instances, help themselves with endoscopic techniques.

TABLE 1 Molar Horizontal and Vertical Displacement

Molars	*Horizontal (mm)*	*Vertical (mm)*
First (M1)	10.37**	2.87*
Second (M2)	11.81**	4.38*

* $p > .05$.
** $p < .05$.

This study is in agreement with that of Santiago et al. (2005), in which surgical disruption of the teeth occurred mainly in patients under 5 years old. On the other hand, the observed changes in the position of the second and third molars may not be totally related to surgical trauma but to the rapid creation of space. Developing teeth, especially those in stage 2 of development (Fig. 1), in which there is minimal root formation, are susceptible to rotation inside the bony dental crypt after new and rapid arch length development. In addition, the observed alterations in second molar development might have been less if all of the patients had not undergone a previous attempt at midface advancement with traditional approaches. In this sample, 8 of the 14 patients had previous failed traditional midface advancement surgery. The chances for unaffected second molar development after midface advancement with distraction are likely to be better as it is a less traumatic procedure than traditional acute midface advancement.

The impaction observed in one first permanent molar (Fig. 5) could be interpreted as a condition exacerbated by surgery and distraction and not the only cause of it. Dental development in children with craniosynostosis is nearly always abnormal. The constricted arch makes crowding of maxillary teeth very common, and ectopic eruption of maxillary first molars can occur in up to 47% of patients with Crouzon syndrome (Gorlin et al., 2001). Most patients analyzed had no third molars either because of their young age or because of previous surgery. Of the third molars, evaluated one had an extreme superior and posterior displacement, and the other had a severe anterior rotation (Fig. 8). Three had surgical tooth trauma and complete disruption of the dental germ. Damage of the third molars will continue to be common after monobloc osteotomy because of its crowded position in the most posterior region of the maxillary tuberosity. Although reported in this study, damage to the maxillary third molars is not considered clinically relevant as most of these teeth need to be extracted at the time or after orthodontic treatment.

Patients with craniofacial synostosis have severe maxillary hypoplasia and lack of space for the eruption of most posterior teeth. Monobloc distraction results in gradual maxillary bone lengthening. In this study, bone formed in the posterior aspect of the tuberosity, and sufficient space was created for the eruption of the first and second molars. The space created could be confirmed by the horizontal average displacement of 10.37 mm ($p < .05$) for first permanent molars and 11.81 mm ($p < .05$) for second permanent molars. The vertical displacement of 2.87 and 4.38 mm of the first and the second permanent molars, respectively, was not significant ($p > .05$), and it is a reflection of vertical control of the monobloc segment with the RED technique (Fig. 3A and 3B). Previously, we reported on the controlled forward and downward advancement of the whole monobloc segment without creation of an open bite (Figueroa et al., 2004; Polley and Figueroa, 2003). Jensen et al. (2007) reported a counterclockwise rotation of the Le Fort III midface segment. The reason for this unfavorable rotation was attributed to the pull force applied along the inferior aspect of the Le Fort III segment with the nasofrontal junction acting as a pivot for the skeletal rotation (Jensen et al., 2007) This resulted in a lack of vector control and creation of iatrogenic open bite. Other researchers (Fearon, 2001; Havlik et al., 2004) have reported on this problem and developed various techniques to control this unfavorable rotation. The monobloc advancement with RED using four external points of pull, two superior (supraorbital) and two inferior (attached to a toothborne intraoral splint with external traction hooks), as developed by the authors (Figueroa and Polley, 2006a, 2006b; Figueroa and Polley, 2007; Figueroa et al., 2001; Polley and Figueroa 1998; Polley et al., 1995), permitting excellent horizontal and vertical control throughout the distraction process. This horizontal and vertical control over the mobilized large skeletal segment is one of the main reasons why the authors prefer the monobloc osteotomy to the Le Fort III osteotomy.

Although changes in the axial inclinations of the permanent molars were not statistically significant, the average 3 and 18 degree changes in the axial inclination of the first and second molars, respectively, were favorable and clinically relevant. As observed in the cephalometric radiographs (Fig. 4A and 4B), those teeth developed in a more upright and normal position. Spaces created between the molars after distraction could also be observed in the panoramic radiographs, which indicated creation of arch length necessary for a normal eruption pattern of the permanent molars (Fig. 8). Furthermore, the eruption process of the permanent molars contributes to the creation of posterior bone and may further enhance the stability of the monobloc procedure (Figueroa et al., 2004).

Although the spaces detected may benefit the eruption of the permanent molars, future investigations could answer whether those spaces could benefit the teeth in the middle segment (premolars and permanent canines) where crowding is severe and premolar extraction and/or transverse expansion is frequently required to relieve middle and anterior arch crowding. The long-term effects of monobloc distraction on the maxillary molars needs further investigation with longer follow-up and a larger sample of patients, especially those in whom only distraction surgery is performed in infancy or in the primary or mixed dentition stages. As distraction has become the treatment of choice over traditional monobloc and Le Fort III advancements in most centers around the world, obtaining larger patient samples without previous surgical damage might be quite feasible in the near future.

Conclusions

Monobloc advancement with RED did not damage the development of the first permanent molars and affected in various ways 23.5% of the developing and unerupted second permanent molars in the tuberosity region. Severe tooth damage usually occurred in younger patients (under 6 years old) in the primary dentition. Radiographic follow-up of maxillary molars is recommended after monobloc distraction.

Distraction created posterior spaces with significant horizontal forward displacement of the second and first permanent molars, which allowed these teeth to clinically improve their axial inclination and eruption. In addition, the maxillary molars were carried forward within the advanced monobloc segment with minimal vertical change. The observations from this study indicate that monobloc advancement with RED causes significantly less damage to the unerupted first and second maxillary molars than does conventional surgery. Surgeons must exercise extreme caution when performing pterygomaxillary disjunction to avoid direct damage to the developing maxillary molars, especially in younger patients.

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