Tooth Viability Following Distraction Osteogenesis in Patients with Maxillary Hypoplasia

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Abstract

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Examination of patients with severe maxillary retrusion following maxillary distraction osteogenesis (MDO) finds variable disruptions in tooth development. This study explored maxillary osteotomy type and tooth development stage at time of surgery on long-term viability of teeth.

Records of 46 patients that received MDO met inclusion criteria: 22 with Le Fort I and 24 with Le Fort III. Dermirjian's method was used to assess tooth development on pre-distraction and follow up radiographs. Tooth viability was scored by analyzing tooth position, development, and predicted functionality.

Second molars in earlier stages of development at surgery had more nonviable outcomes. Teeth in late development stages at surgery had no unfavorable outcomes. For all tooth types, teeth with more advanced development at surgery had better viability scores than those in earlier stages. This information applies to risk/benefit discussions when planning MDO. For patients without urgent indications, delaying surgery may reduce tooth development disruption.

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DEDICATION

To my always supporting family, and my beautiful country Venezuela.

1. Specific Aims

1.1. Introduction and Statement of the Problem

Maxillary Distraction Osteogenesis (MDO) is a treatment option for surgical advancement of the upper jaw and mid face for patients with severe maxillary retrusion due to cleft palate or other craniofacial syndromes. Many of these patients are children that require advancements of more than 15mm, for which MDO is the treatment of choice (Bouchard et al 2009). MDO is done to open obstructed airways, to prevent tracheostomy or allow closure of an existing tracheostomy, to protect the eyes in patients with severe exophthalmos, and to improve appearance and quality of life (Bouchard et al 2009, Bannink et al 2010). Depending upon the objective of the surgery and level of maxillary/midface retrusion, MDO can be done with a Le Fort I, II or III osteotomy. (Bouchard et al 2009)

Tooth development occurs over many years and can be disrupted by surgery, trauma (Yamamoto 2009, Ranta and Ylipaavalniemi 1973) or systemic factors such as malnutrition (Gaur and Kumar 2012, Heinrich-Weltzein et al 2012), severe febrile illness or chemotherapy (Takeda 1987, Anneroth 1985). Since multiple factors can affect developing tooth buds and MDO is generally done in growing patients, MDO could affect tooth development. Disruption of a tooth bud may manifest as altered final tooth morphology, ectopic position and/or compromised functionality due to arrested root development, impaction, tooth germ displacement, ankylosis, or tooth loss. (Sant'Anna et al, 2009). If the osteotomy is in proximity to or contacts developing tooth buds, disrupted development may, but does not always, occur.

The geometry and surgical timing of Le Fort I, II and III level MDO surgery places different unerupted permanent teeth at risk for disrupted development (Figure 1). A Le Fort III osteotomy is in proximity to developing molars, potentially affecting their development, position or eruption (Santiago et al, 2005) while a Le Fort I osteotomy includes the pre-eruptive sites of all developing maxillary canines, premolars, and molars.

Figure 1: Facial Le Fort fractures.

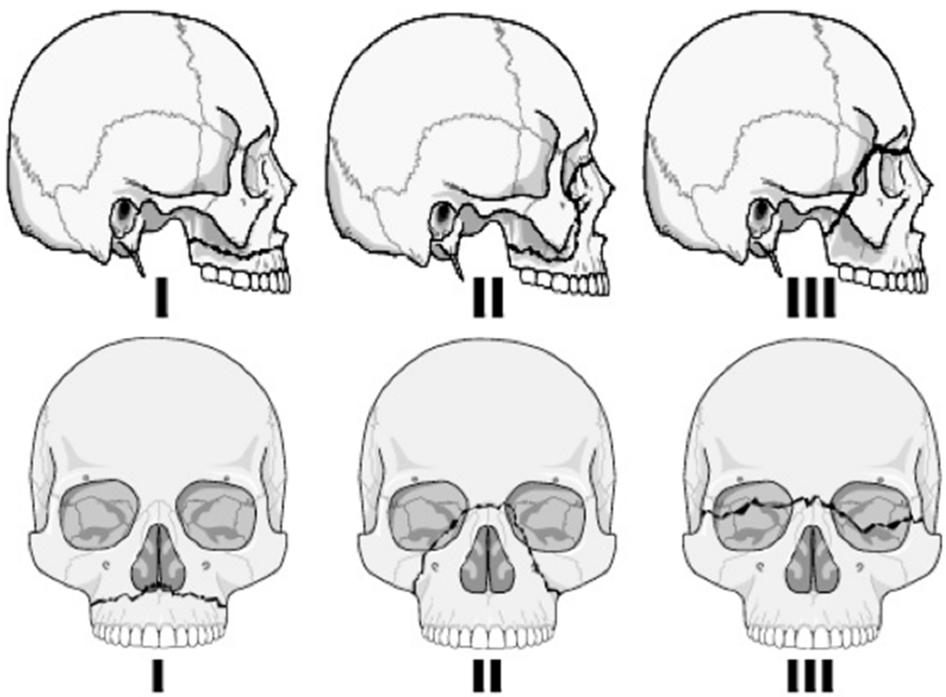

Figure 1 source: www.rad.washington.edu/mskbook/facialfx.html

Studies at other institutions assessing the effect of mandibular distraction upon tooth development have reported conflicting results: one study reported accelerated but morphologically normal tooth development and another study reported arrested and disrupted tooth development. (Keine-Hakala 2007, Kim et al 2009). Studies have examined the relationship between damage to developing teeth in young patients suffering dentofacial trauma and stage of dental development at the time of injury. Results vary; some found no relationship (Suei et al 2006,) while others found the stage of development a crucial factor in the fate of tooth buds (Ranta and Ylipaavalniemi 1973). Contrary to what dentists have traditionally been taught, a case series examining extraction of impacted maxillary midline supernumerary teeth (mesiodens) found better outcomes for maxillary permanent incisor development when mesiodens removal was done in younger patients, postulated to occur because younger teeth may have more capacity to recover from surgical insult. (Omer 2010).

Although the continued development of unerupted teeth after trauma and distraction osteogenesis has been studied, these studies haven't focused on the optimal timing for distraction surgery to avoid disruptions in tooth formation.

1.2. List of Specific Aims

a. Evaluate which stage of tooth development at time of surgery presents the least risk for disruption of normal tooth development when doing Maxillary Distraction Osteogenesis (MDO).

b. Examine the correlation between the type of osteotomy performed and the type of teeth showing disruption.

2. Background and Significance

2.1. Significance of the Problem

Maxillary Distraction Osteogenesis (MDO) is done in young children with little knowledge of predictable effects of the surgery on the development of the dentition. This study will provide more accurate information about possible outcomes that could influence the surgical design or timing of the procedure. Also, this study will help families understand more clearly the degree of risk to permanent teeth if distraction surgery is done at certain stages of tooth development. The results of this study should improve the knowledge base about the dental impacts of MDO surgery in patients with developing teeth.

2.2. Background and Review of Literature

Distraction Osteogenesis (DO) is a technique used for bone lengthening in which a controlled distraction of a healing fracture callus stimulates new bone production at the surgical site. In order to achieve this, an osteotomy is created and a mechanical device is fixed to the bone which, when activated, creates tension across the osteotomy site; this induces bone formation and histiogenesis of blood vessels, muscles, nerve, skin and mucosa (Aronson et al 1989, Bouchard et al, 2009). Codivilla in 1905 and Ilizarov in 1956 were the first to report using DO in orthopedic surgery. McCarthy et al first used DO in mandibles to treat patients with craniofacial microsomia in 1990. (McCarthy et al, 1992). Chin and Toth (1996) described DO in five patients with maxillary deformities using bilateral Le Fort III osteotomies for maxillary advancement. Since then, Maxillary Distraction Osteogenesis (MDO) has become an option for treatment of severe maxillary and/or midface hypoplasia in patients with cleft palate and craniofacial syndromes such as Apert, Crouzon and Pfeiffer.

Patients with severe midface hypoplasia, in addition to encountering esthetic and psychosocial difficulties, can have functional problems such as airway obstruction and exophthalmos which can require early intervention such as tracheotomy or midface repositioning to improve the oropharyngeal and nasopharyngeal airways and improve bony protection of the eyes (Perkins et al, 1997; Sculerati et al, 1998; Bannink et al, 2009). In these studies, tracheostomy-dependent patients required large mid face advancements to adequately improve the airway. MDO provided a method to perform larger advancements than traditional surgery with better stability, no need for grafting and better soft tissue response. (Gateno et al 2005, Figueroa and Polley 2008). Patients with severe maxillary retrusion can benefit from esthetic and other functional changes that MDO can provide, as reported by some authors. Improved occlusal relationships and enhanced facial esthetics can be achieved in patients with cleft lip and palate and/or severe maxillary hypoplasia (Molina et al 1998, Miyazaki et al 2013). Due to these potential benefits, MDO is sometimes indicated for young children, primarily to address functional problems and to improve the quality of life.

The effects of MDO surgery on the development of the dentition have not been comprehensively studied. Studies relevant to the present project have examined the effect of trauma, osteotomies and other factors on the development and eruption of teeth.

As many authors have reported, tooth development can be affected by several factors. Nutrition may influence the timing of eruption (Gaur and Kumar 2012) and the absence or presence of permanent teeth (Heinrich-Weltzien et al 2013), irradiation can alter crown and root morphology, and increases incidence of tooth agenesis (Takeda et al 1987, Nishimura et al 2013).Trauma can have different effects on tooth development. Yamamoto et al (2009) reported abnormal development of tooth buds located in proximity to mandibular fractures in three infants. Scerri et al (2010) studied 32 children and reported that trauma to primary dentition caused disturbances of enamel (primarily discoloration) but no eruption disturbances in permanent teeth.

Tooth development anomalies have been associated with certain congenital syndromes. Reitsma et al (2014) performed a cross-sectional study of 67 patients with Crouzon and Apert syndrome and found that tooth agenesis is more commonly seen in patients with these syndromes than in control groups. Other authors report certain dental characteristics of patients with Crouzon and Apert syndromes such as higher tendency for supernumerary and ectopic teeth, delayed eruption of permanent and primary dentition, severe crowding, and crossbites. (Horbelt 2008)

Since MDO is sometimes done in children with developing dentitions, and tooth development can be affected by the factors previously reviewed, it is reasonable to postulate that MDO has the potential to affect the development of permanent teeth, especially in younger patients. As reported by Sant'Anna et al (2010) when studying 14 patients having monobloc advancements with Rigid External Distraction (RED), there was a higher risk of molar damage in patients who underwent surgery during the primary dentition, compared to those in mixed or permanent dentition, affecting mostly second molars in patients younger than six years old at the time of surgery. Santiago et al (2005) studied 31 patients with craniosynostoses and evaluated the effect of Le Fort III osteotomy on the position and eruption of the maxillary first and second molar. They reported that both first and second molars were more affected in the surgery group than in the control group, and that the first molars were more likely to erupt than the second molars. This finding raises the suspicion that the disruption of development of a tooth may be related to the stage of development of the tooth when the disruptive event occurred.

Some authors studied the effect of mandibular trauma on tooth development and eruption, reporting disparate results. In a study of 25 children, Ranta and Ylipaavalniemi (1973) suggested that the stage of tooth development is a crucial factor in the fate of tooth buds exposed to trauma such as a fracture of the bone in the area of the tooth bud. In their study examining tooth development following mandibular fracture, those teeth in which the calcification of the crown was incomplete with no root formation were more susceptible to disruption than teeth with complete crown calcification and root formation in progress. Koenig et al (1994) analyzed 30 patients with mandibular fractures and found that the majority of tooth buds directly in the line of fracture erupted normally; the authors speculated that favorable outcomes resulted due to higher elasticity of the developing follicles compared to the bone. Based on a review of 22 patients Suei et al (2006) reported that tooth buds appear capable of continued development and eruption even when on a mandibular fracture line, regardless of the developmental stage of the tooth or the degree of displacement of the fracture.

Omer et al (2010) studied the effects of removing an unerupted mesiodens on the adjacent developing permanent incisors in 126 children. He reported that surgical intervention at early tooth developmental stages resulted in less damage to permanent teeth with a cutoff time point of developmental stage “E” of Dermijian’s Classification, crown is completely formed (Figure 2-4).

Figure 2: Dermijian’s Dental Calcification Stages.

Stage A:	Calcification of single occlusal points without fusion of different calcifications.
Stage B:	Fusion of mineralization points; the contour of the occlusal surface is recognizable.
Stage C:	Enamel formation has been completed at the occlusal surface, and dentine formation has commenced. The pulp chamber is curved, and no pulp horns are visible.
Stage D:	Crown formation has been completed to the level of the cementoenamel junction. Root formation has commenced. The pulp horns are beginning to differentiate, but the walls of the pulp chamber remain curved.
Stage E:	The root length remains shorter than the crown height. The walls of the pulp chamber are straight, and the pulp horns have become more differentiated than in the previous stage. In molars the radicular bifurcation has commenced to calcify.
Stage F:	The walls of the pulp chamber now form an isosceles triangle, and the root length is equal to or greater than the crown height. In molars the bifurcation has developed sufficiently to give the roots a distinct form.
Stage G:	The walls of the root canal are now parallel, but the apical end is partially open. In molars only the distal root is rated.
Stage H:	The root apex is completely closed (distal root in molars). The periodontal membrane surrounding the root and apex is uniform in width throughout.

Figure 2 Source: Sun-Mi Cho, DDS, MSD,a and Chung-Ju Hwang, DDS, MSD, PhD. Skeletal maturation evaluation using mandibular third molar development in adolescents. Korean J Orthod. 2009 Apr;39(2):120-12.

Figure 3: Development chart of molars with radiographic images, modified from Dermijian et al by Kasper et al.

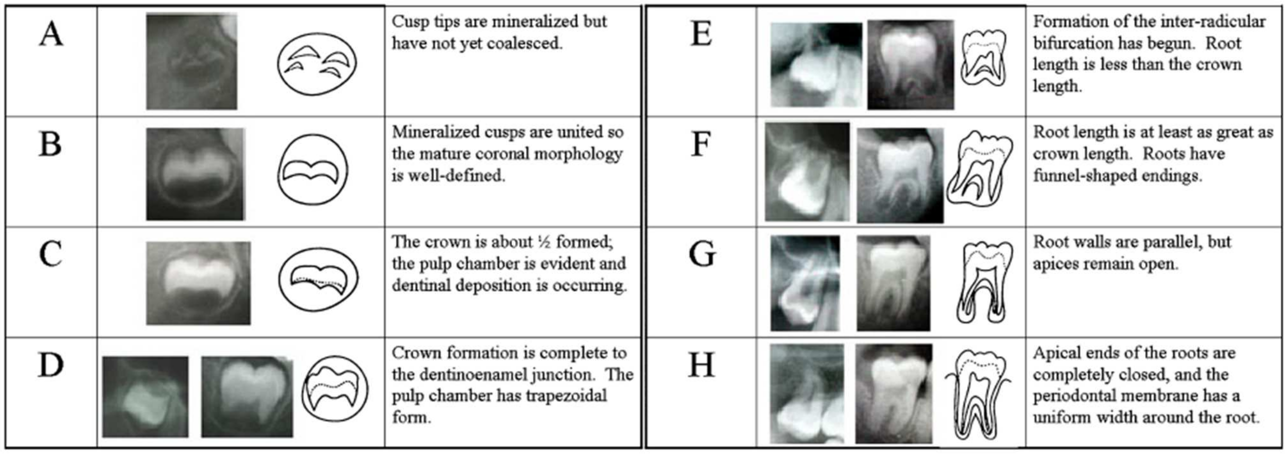

Figure 3 source: Kasper KA, Austin D, Kvanli AH, Rios TR, Senn DR. Reliability of third molar development for age estimation in a Texas Hispanic population: a comparison study. J Forensic Sci. 2009 May;54(3):651-7.

Figure 4: Dental Stages of monoradicular teeth by Dermijian

Dental stage	Description
C 4-5 ys*	The enamel formation is completed at the occlusal surface, with extension and convergence toward the cervical region. Dentinal deposits started to form, with curved shape outline of pulp chamber at the occlusal surface.
D 5-6 ys	The crown formation is completed down to the cementoenamel junction. The superior border of the pulp chamber has a definite curved form, being concave toward the cervical region. Pulpal horns projection may be present. The beginning of root formation is seen in the form of a spicule.
E 6-7 ys	The crown is completely formed and the pulp chamber has formed straight lines with continuity broken by the presence of a large pulp horn. The root length is less than the crown height.
F 7-9 ys	The pulp chamber roughly forms an isosceles triangle. The apex ends in a funnel shape. The root length is equal to or greater than the crown height.
G 9-10 ys	The root length is greater than that in stage F. The root canal walls are parallel. The apex is still or partially open.
H >10 ys	The apex is completely closed. The periodontal membrane is of uniform width around the apex.

Figure 4 source: Omer RS, Anthonappa RP, King NM. Determination of the optimum time for surgical removal of unerupted anterior supernumerary teeth. Pediatr Dent. 2010;32(1):14-20

Since the surgical intervention and osteotomies inherent to MDO may be considered as a source of trauma to developing tooth buds, the studies cited provide raise the question: is there an optimal time for MDO that minimizes the chance of damage to the developing teeth?

The bone cuts for MDO are analogous to the maxillary fracture lines described by Le Fort. Depending upon the degree of maxillary hypoplasia, osteotomies at different levels are selected to advance the maxilla. In Le Fort I, the bone incision is above the floor of the nasal cavity through the lower nasal septum, lower walls of maxillary sinuses and lower pterygoid plates, separating the entire palate and alveolar process from the maxilla. A Le Fort II osteotomy separates the entire maxilla by fracturing through the roof of the nose, lacrimal bones, the floor of the orbits and the zygomaticomaxillary sutures, continuing posteriorly through the infratemporal surfaces of the maxilla and lower pterygoid plates. For Le Fort III osteotomies, a fracture is done on the roof of the nose, lacrimal bones, medial orbital walls, floor of the orbits, lower pterygoid plates and zygomatic arches to separate the viscerocranium from the skull base (Larheim 2008). Le Fort II osteotomy is not commonly used for MDO. (Figure 5)

Figure 5: Lateral cephalogram pre, during, and post maxillary Le Fort I distraction

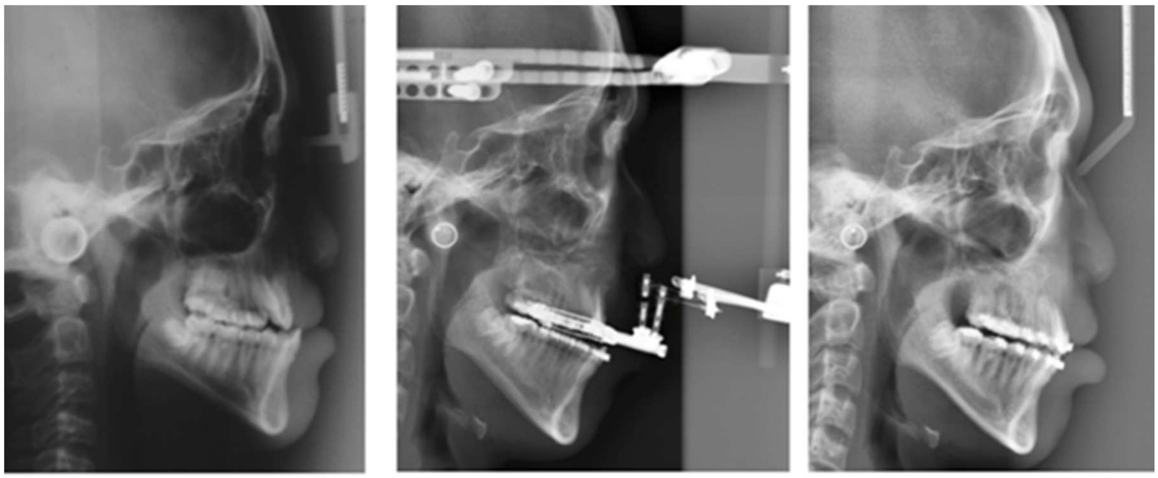

Figure 5 source: intechopen.com

To date, few patients receiving MDO have been specifically studied regarding tooth development outcomes. The effect of Lefort I and Le Fort III MDO on the development of tooth buds is an important question and merits further study.

2.3. Purpose of the Study

The purposes of the study were to explore the relationship between the stage of tooth development at time of MDO surgery with the subsequent development of the permanent teeth, and to examine the effects of Le Fort I versus Le Fort III MDO on subsequent development of the permanent teeth

3. Research design and Methods

3.1. Research Question & Hypotheses

Question #1

Does the stage of tooth development at the time of MDO affect long term tooth viability?

Null Hypothesis #1: No significant correlation exists between the stage of tooth development at the time of MDO surgery and long-term tooth viability.

Expectation: When comparing teeth with normal development and teeth with disrupted development, data analyses will show a significant correlation with surgical timing at different stages of tooth development. It is expected that completion of no more than crown development at the time of surgery will be a key factor in susceptibility to disrupted development.

Question #2

Does the type of the osteotomy performed have an effect on which teeth show disrupted development?

Null Hypothesis: No significant correlation exists between the type of osteotomy performed and the type of teeth showing disrupted development.

Expectation: Teeth developing in close proximity to the osteotomy site will be more affected than those located further away. Permanent Canines will be more affected by Le Fort I osteotomies than by Le Fort III osteotomies.

3.2. Research Design to Accomplish Specific Aims

This descriptive study was a retrospective analysis of data from patient charts and images at stage T1 (pre-distraction osteogenesis), T2 (post-distraction), T3 (last diagnostic image available).

This study was approved by the Institutional Review Board (IRB) of the Human Resources division of Seattle Children's Hospital (IRB #14905).

3.3. Methods

1. Patient Sample

This study used all qualifying data from medical records at Seattle Children's Hospital for patients that received Maxillary Distraction Osteogenesis between January 1, 2002 and December 31, 2011.

Inclusion Criteria:

- Availability of diagnostic quality imaging at all time points.
- Patient received Maxillary Distraction Osteogenesis at Seattle Children's Hospital.
- Patients with Le Fort I or Le Fort III osteotomies

Exclusion Criteria

- Patients who received other craniofacial surgeries during the study period.

2. Patient Data Collection

For each patient that met the inclusion criteria the following data were collected:

- ID: number used to identify the chart number
- Birthdate: month/day/year
- Sex: Male or Female
- Medical Diagnosis: craniofacial syndrome or condition according to the medical record. These include Crouzon Syndrome, Pfeiffer, Otopalatal

Digital Syndrome, cleft palate, Saethre Chotzen and Apert Syndrome. If more than one diagnosis was applicable, they were recorded and the patient was categorized as multisyndromic.

- Surgery type: Osteotomy performed, Le Fort I or Le Fort III
- Pre-treatment date (T1): month/day/year in which the latest pre-surgical image was taken.
- Type of image: Panoramic (Pan) or Computed Tomography (CT). If both were available, the panoramic image was the image analyzed.
- Date of Surgery: month/day/ year in which MDO was done.
- Time between T1 image and surgery.
- Stage of tooth development: the stages of maxillary canines, first and second molars, as well as the stages of mandibular canines, first and second molars were collected at every time point. The tooth development stages were classified according to the method of Dermirjian et al (figures #3-5). In cases where a tooth was not visible due to image quality or was not calcified enough to visualize, it was considered not visible (N/V). If the tooth was never seen in any collected image it was considered missing (MISS).
- Crown calcification: according to the stage of calcification and Dermijian et al method, the tooth was classified as not having complete calcification of crown (Early Stage) including stages A, B, and C; having the calcification of the crown completed but less than half of the root

(Middle Stage) including stage D and E, or as having more than half of the root formed including stages (Late Stage) F, G, and H of the Dermijian classification.

- Post-Distraction date (T2): month/day/year in which the distractor was removed. Many Subjects had an image taken at the time of hardware removal. At this time point, it was noted if there was any obvious damage to the tooth bud during the surgery such as incision directly through the bud, or screws /plates placed into the bud.
- Follow up (T3): Latest month/day/year in which an image was taken. At this point the following additional data was collected:
- Viability: defined as the projected capability of the tooth to function effectively in the future. The following factors were considered to determine tooth viability.

    1. Acceptable Position: Any tooth in a position that allows eruption in a reasonable proximity to arch.
    2. Orthodontically retrievable: Any tooth with an abnormal path of eruption was was categorized by the potential to be moved orthodontically into a functional position.
    3. Normal Development: Any tooth where crowns and roots are developing normally for patient’s age and stage of development.
    4. Potentially Functional: Any tooth with structural and

positional parameters, which should result in acceptable function- in the occlusion.

5. Not Possibly Functional: Any tooth not in good position and either not retrievable with orthodontics or with grossly dysmorphic crown development.

The researcher used the collected data to judge the viability of the tooth and rank it according to the following scale: (Table #1)

Table 1. Viability Ranking Scale

Acceptable Position	**Ortho Retrievable**	**Normal Development**	**Potentially Functional**	**Viability ranking**
Y	N/A	Y	Y	**1**
N	Y	Y	Y	**2**
Y	N/A	N	Y	**3**
N	N	Y	N	**4**
N	N	N	N	**5**

Some teeth were extracted during the patient's treatment and clinical notes were reviewed to evaluate the reasons. If teeth were extracted due to non-viability they were ranked as 5 on our scale. Teeth extracted due to causes independent of the surgery such as caries or crowding were excluded from the sample for statistic analysis

Mandibular tooth development served as a control for differentiating between the disruption of tooth development due to the systemic challenges of general anesthesia, the surgery, and post-surgical course and the inherent tooth development pattern of the subject. Questions on viability were resolved by consensus after review of radiographs by three orthodontists (BS, BW, GG).

At completion of data collection, data was examined for consistency with a sample set of 10% of all included patient records. Images of 6 randomly selected patients were reexamined and data extracted a second time by the examiner in four categories

Table 2. Intra examiner accuracy table

Intra Examiner Accuracy

	Stage at T1	Obvious damage at T2	Viability Ranking	Stage at T3
Sample	33/36	36/36	35/36	34/36
Accuracy	92%	100%	97%	94%

3. Patient Sample Size

The study sample was collected from patients treated with distraction osteogenesis after Le Fort I or Le Fort III ostetomies at Seattle Children's Hospital from January 1 2002 to December 31 2011.

A total of 61 consecutive potential patients were screened of which 46 (75%) met the inclusion criteria. The sample was divided into two groups according to osteotomy type: Le Fort I or Le Fort III.

3.4. Data Analysis

A descriptive analysis was performed to evaluate the distribution of patient factors in the sample. Fisher's exact test and a Wilcoxon signed-ranked test were used to examine the difference between gender and final viability ranking between both groups. Test results with considered significant at $p<0.05$. Logistic regression was used to assess the relationship of tooth viability to multiple factors in the Le Fort III patients. The statistical regression models were adjusted with the general estimating equation (GEE) to account for the non-independence of multiple teeth from a same individual. The unit of analysis was based on a single tooth.

4. Results

The Le Fort I group consisted of 28 consecutive treated patients of which 22 (78%) met the inclusion criteria. Five potential patients were excluded due to a previous Le Fort I for expansion or later Le Fort osteotomy for advancement. One potential patient didn't have enough quality images.

From the 33 patients with Le Fort III MDO, 24 (72%) met the inclusion criteria. Reasons for exclusion were: previous Le Fort 1 or Lefort III surgery (5 patients), missing images (3), and unusual modification of Le Fort III surgery (1).

Table 2 displays the distribution of patients between groups by descriptors

Table 2. Distribution of patients between groups

	LE FORT I N= 22	LE FORT III N=24
Medical Diagnosis 1		
Crouzon	0	12
Pfeiffer	0	1
Apert	0	3
Cleft Lip and Palate	20	0
Other	2	8
Gender		
Male	13	12
Female	9	12
Age at Surgery: Mean (Median)		
	15.3 (15.3)	8.1 (7.9)
Time between Sx & T1 Image in Days: Mean (Median)		
	249 (37)	61 (31)
Time between Sx & T3 Image in Years: Mean (Median)		
	1.85 (2.3)	5.1 (5.2)
Maxillary Tooth Type		
Canines	43	47
First molars	42	48
Second Molars	43	43
Stage at T1		
Primary	0	30
Middle	2	56
Final	125	51
Not visible due to image	3	0
Missing	2	6
Unable to classify (image or others)	0	1
Obvious Damage at T2		
Yes	0	14
No	33	125
Not available	99	5
Viability Ranking		
1	127	106
2	1	3
3	0	1
4	0	1
5	0	24
Missing	2	6
Extracted due to cavities or crowding	0	3
Not visible due to image	2	0

Patients that had Le Fort I osteotomy had mainly Cleft and Lip Palate as the primary medical diagnosis. Craniofacial Syndromes were the most common diagnoses in the Le Fort III group. The Le Fort I patients did not always have available images at T2 since is not routine to take images on this group of patients at the time of distractor removal. The Le Fort I group showed only favorable outcomes on the viability ranking scale so further statistical analyses were restricted to the Le Fort III subjects.

For further analysis the final viability ranking was divided into two groups according to its final functionality: favorable outcome (1-3) and unfavorable outcome (4-5)

Table 3. Represents logistic regression analysis for outcome by risk factor for unfavorable outcomes on the Le fort III group.

		Favorable		Unfavorable		
		# Teeth	%	# Teeth	%	p-value*
Total		110	81%	25	19%	
Gender						
	Female	57	83%	12	17%	0.72
	Male	53	80%	13	20%	
Tooth						
	Canines	46	98%	1	2%	< 0.001 *
	1st Molars	43	93%	3	7%	
	2nd Molars	21	50%	21	50%	
Side						
	Left	58	85%	10	15%	0.09
	Right	52	78%	15	22%	
T1 stage						
	Early	12	41%	17	59%	< 0.001*
	Middle (D,E)	49	88%	7	13%	
	Late (F-H)	49	100%			
Obvious disruption at T2						
	Yes	1	7%	13	93%	< 0.001 *
	No	109	90%	12	10%	

Neither gender nor side of the dental arch had statistical significance on the outcome/viability of the teeth. The type of tooth was statistically significant for unfavorable outcome; second molars were more affected than first molars or canines. The stage of tooth development was also significant–teeth at earlier stages of development at the time of surgery were more affected compared to those in middle or late stages.

Table 4. Unfavorable outcomes by tooth and stage of development for Le Fort III group

	T1 stage							
	Early		Middle		Late		Total	
	n	% Unfavorable	n	% Unfavorable	n	% Unfavorable	n	% Unfavorable
Canine	8	0% (0)	23	4% (1)	16	0% (0)	47	2% (1)
1st Molar	2	100% (2)	18	5% (1)	26	0% (0)	46	6% (3)
2nd Molar	19	78% (15)	15	33% (15)	7	0% (0)	41	48% (20)
Total	29	59% (17)	56	13% (7)	49	0% (0)	134	17% (24)

Second molars in the earlier stages of tooth development at the time of surgery showed more unfavorable outcomes. Teeth in the late stages of development at the time of surgery had no unfavorable outcomes. Canines had the least unfavorable outcomes overall.

Dental development of mandibular canines and molars were used to evaluate each patient's tooth development trajectory and to explore the possibility that disrupted maxillary tooth development could have been caused by other factors other than the MDO procedure such as systemic effects of anesthesia, medications, compromised nutrition, or an atypical tooth development pattern in one or more of the subjects.

Table 5. Viability outcome of mandibular teeth at T3 by tooth type and stage of development at time of Le Fort III or Le Fort I Maxillary Distraction Osteotomy

LE FORT III PATIENTS

	T1 stage						Total	
	Early		Middle		Late			
	n	% Unfavorable	n	% Unfavorable	n	% Unfavorable	n	% Unfavorable
Canine	10	0% (0)	20	0% (0)	18	0% (0)	48	0% (0)
1st Molar	2	0% (0)	14	0% (0)	32	0% (0)	48	0% (0)
2nd Molar	26	0% (0)	12	0% (0)	10	0% (0)	48	0% (0)
Total	38	0% (0)	46	0% (0)	60	0% (0)	144	0% (0)

LE FORT I PATIENTS

	T1 stage						Total	
	Early		Middle		Late			
	n	% Unfavorable	n	% Unfavorable	n	% Unfavorable	n	% Unfavorable
Canine	0	0% (0)	0	0% (0)	44	0% (0)	44	0% (0)
1st Molar	0	0% (0)	0	0% (0)	44	0% (0)	44	0% (0)
2nd Molar	0	0% (0)	0	0% (0)	41	0% (0)	41	0% (0)
Total	0	0% (0)	0	0% (0)	129	0% (0)	129	0% (0)

Mandibular teeth had 100% favorable outcomes in both groups, regardless of the stage of tooth development at time of MDO surgery. Normal development continued and tooth viability was not adversely affected.

5. Discussion

Maxillary Distraction Osteogenesis (MDO) has been a treatment alternative to monobloc advancement in patients with severe maxillary retrusion for many years. It allows progressive bone formation, avoids the need of autogenous iliac crest grafting, and permits gradual soft tissue adaptation that may improve the stability of the surgery and esthetic outcomes. Nevertheless, the effects of MDO on the dentition are not well described.

Some studies have shown that tooth development can be easily affected and clinicians have reported disruption of teeth on patients undergoing distraction osteogenesis. This disruption can sometimes severely affect the final occlusion of the patient since multiple posterior teeth can be missing or are not functional because of abnormal development.

This study evaluated the effects of MDO on developing teeth classified using a standardized reliable methodology as well as the effects of two osteotomy types, Le Fort I and Le Fort III, on the developing maxillary dentition.

When analyzing the descriptive results of both groups (Table 2) it is notable that the primary medical diagnosis differed between the Le Fort I and Le Fort III groups; most Le Fort I patients had cleft and lip palate while the majority of Le Fort III patients had craniofacial syndromes. Patients with cleft lip and palate commonly have lip and palate surgical repair(s) at young ages, these surgeries generate scar tissue that interferes with maxillary development, which can result in a severely retrusive maxilla/mid face in some patients. Such patients benefit from Le Fort I

MDO to advance the area of greatest retrusion. Le Fort III is indicated for patients with craniosynostosis and severe retrusion of the entire midface involving the orbits, upper airway, or both.

Significant differences were found between groups for both mean age at surgery and stage of tooth development at time of surgery. Le Fort I patients had MDO at an a mean age of 15 years old while Le Fort III patients underwent surgery at younger ages, averaging 8.1 years old. This correlates with most teeth in the Le Fort I group being in the final stage of development whereas those of the Le Fort III group were more evenly distributed between the three stages of development in this study. Earlier timing of MDO in children with craniofacial syndromes is generally recommended to avoid or ameliorate persisting complications from severe midface retrusion during early facial growth. Patients with cleft palate usually receive MDO after much of the growth has been completed to improve airway patency and occlusion.

The differences in age and tooth developmental stage explain the difference in the viability outcomes between both groups. The Le Fort I group had only favorable outcomes, while the Le Fort III group had some teeth with unfavorable scores. Our results do not imply that Le Fort I osteotomy carries less risk to teeth than the Le Fort III osteotomy. At Seattle Children's Hospital Craniofacial Center, Le Fort I distraction surgery is done for patients at ages when teeth are in more advanced stages of development, which impacts the favorable outcomes for long-term tooth viability The stage of development of teeth at time of surgery, rather than type of osteotomy, is most likely to influence the viability of teeth near the surgical sites.

Since tooth viability outcomes were more diverse for the Le Fort III group, further analysis was done to evaluate the possible risk factors. Table 3 notes that 19 percent of the teeth in this group had an unfavorable outcome. Second molars were more affected than canines and first molars, and those teeth in early stages at time of surgery were at higher risk compared to those in middle or late development stages. As expected, when an obvious disruption of the tooth was visible at T2 there was a greater than 90 percent chance that the disrupted teeth had an unfavorable outcome.

Table 4 demonstrates in more detail the relationship between stage of tooth development, type of tooth, and unfavorable outcomes. The second molars and teeth at earlier stages of development are the most affected. 60% of teeth in earlier stages are affected compared to 0% in the late stages. 48% of second molars had unfavorable outcomes in contrast with less than 8% of first molars and canines. These results are in agreement with those from Sant'Anna (2010), who reported second molars were more affected during MDO than first molars, and usually occurred in patients in the primary dentition. It also parallels the results found by Santiago *et al* (2005), where second molars were more commonly damaged compared to first molars in patients undergoing Le Fort III advancement due to craniosynostosis.

To examine the potential for tooth disruption caused by other factors such as general anesthesia, medications, compromised nutrition, or other peri-surgical factors, development of the mandibular molars and canines was analyzed for all subjects. All developing mandibular teeth continued on a normal developmental trajectory following MDO surgery of both types. This finding provides evidence that systemic factors didn’t affect normal development of teeth in the subject and that MDO surgery places only developing maxillary teeth at risk and has no impact on the mandibular teeth.

Potential contributing factors such as degree of maxillary hypoplasia, millimetric amount of distraction, the duration of distraction, and proximity of osteotomy site to the tooth could not be measured in this study. These data were not available and the osteotomy cut could not be consistently visualized on the radiographs. Our results do not discount that these factors affect tooth viability. Even though the time between the T1 image and MDO surgery differed, this variability was not considered clinically significant because teeth take months to years to progress within developmental stages and the stages were grouped into three categories: early, middle, and late stages.

This study provides a better understanding of the effects that maxillary distraction osteogenesis may have on the developing dentition. More complete information about potential risks and benefits to the dentition is important to patients, families, and clinicians. Each patient considered for MDO is unique and may present with factors that influence a craniofacial team to recommend early surgery (eg. to avoid a tracheostomy). For patients without urgent indications for MDO, delaying surgery may offer the benefit of reduced disruption in tooth development and increased numbers of viable teeth over the long term.

Given the findings in this study t would be of interest to compare results from the Le Fort III subjects having MDO in the present study with patients having Le Fort III monobloc advancement to determine any differences in long term tooth viability.

6. Conclusion

This study found that the stage of tooth development at the time of surgery is the primary risk factor for unfavorable long term tooth viability in patients treated with maxillary distraction osteogenesis.

7. References

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