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Transverse Dental Arch Relationship and Occlusion in Surgically Repaired Unilateral Cleft Lip and Palate Egyptian Children

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ABSTRACT

Objective: *To assess and compare the transverse dental arch relationship and occlusion in surgically repaired unilateral cleft lip and palate Egyptian children with those of healthy comparable non-cleft children.*

Study design: *Comparative cross-sectional. Thirty-one non-syndromic children with repaired unilateral cleft lip and palate (UCLP) and mean age 7.35 ± 1.52 years together with thirty-one healthy, comparable, non-cleft children were recruited from Faculty of dentistry, Alexandria University. For each subject, sagittal molar and cuspid occlusion were measured using dental study casts. The buccolingual dental arch relationships were determined using modified Huddart/ Bodenham scoring system.*

Results: *Mesial step terminal plane and class III cuspid relation were significantly higher in UCLP children in the age group 4-5 years. Class III permanent molar and cuspid relations were significantly higher in UCLP children in both age groups 6-7 and 8-9 years. Modified Huddart/Bodenham showed a significantly more negative total arch constriction score in 6-9 year old UCLP children.*

Conclusions: *There was a predilection for most of UCLP children to have mesial step terminal plane in primary dentition, class III permanent molar relation in mixed dentition and Class III cuspid relations. Modified Huddart/ Bodenham scores revealed that UCLP children suffered from constricted maxillary arch especially in the canine region.*

Keywords
Unilateral cleft lip and palate, Terminal plane relationship, Sagittal molar occlusion, Sagittal cuspid occlusion, Modified Huddart/ Bodenham index.

Introduction
Cleft lip and cleft palate are among the most common congenital defects in the cranio-facial region. They result from incomplete fusion of maxillary and intermaxillary processes during development of the fetus [1]. The etiology of these defects is considered multifactorial through interaction of both genetic and environmental factors. Thus, it can occur as an isolated condition, or as one component of an inherited disease or syndrome [2-4].

The prevalence of cleft lip and palate worldwide is about one per 500–700 of all births. It differs with cleft type, gender and ethnic origin [5]. Most studies give a ratio varying between unilateral and bilateral cleft lips to be predominantly favoring unilateral cleft lips [6-9]. Recent data on birth defects from population-based studies originating from Middle East are lacking [10]. Meanwhile, few published articles give a rough idea about the incidence of cleft in the region. The overall incidence rate of cleft lip and palate per 1000 live births was 1.5 in Oman in 2001 and 0.9 in Sudan in 2005 [11,12].

According to Athanasiou et al. [13] and many others [14-18], maxillary arch dimensions are generally reduced in patients with clefts. In addition, the primary surgical repairs affect maxillary arch dimensions in children with clefts. Athanasiou et al. [19] stated that cross-bite is an early and common malocclusion in children with clefts due to the reduced transverse maxillary arch widths.

Different surgical protocols and infant orthopedics are used to correct unilateral cleft lip and palate (UCLP). However, there is no general consensus on the optimal method of treatment [20]. Some centers use a multistage approach, in which more than one operation is done to close the UCLP and others use one-stage repair approach [21,22]. The Oslo protocol is an example of multistage approach in which lip closure is accomplished by Millard technique [23] at 3 months of age. The posterior palate is closed at 18 months using a modified von Langenbeck technique [24]. Finally, alveolar bone grafting is done during the mixed dentition. Secondary surgery can be performed on an individual basis to repair any residual defect [21,22].

In order to assess the treatment outcome regarding arch dimensions and occlusion of these various surgical protocols, different methods have been proposed. They can score dental arch relationships in the primary, mixed and permanent dentitions. Some have taken measurements directly from dental study casts [25,26], or have used photocopies of models [27]. The Huddart and Bodenham system [28], was developed in 1969 to be applied on the study models of UCLP patients in the primary dentition. The system uses the frequency and severity of crossbites to evaluate maxillary arch constriction in the labial, greater (non-cleft) and lesser (cleft) buccal segments. A negative score represents maxillary arch constriction. Mossey et al. in 2003 [29], modified Huddart and Bodenham system to be used with the mixed dentition. Modified Huddart and Bodenham system is considered to be the most sensitive and objective index used. Nevertheless, the Great Ormond Street London, Oslo Norway (Goslon) ranking system developed by Mars et al. in 1987 [30], is considered the most commonly used index that measures the treatment outcomes of surgical repair. However, it assesses dental arch relationships from study models in only the late mixed and early permanent dentitions. In 1997, Atack et al. [31], introduced a similar index, the five-year-old index, to be applied in the primary dentition. In spite of the common use of the Goslon and the five-year-old indices, they are considered subjective and a calibration course is required for those who wish to use the indices for outcome assessment [30,31].

The ultimate aim of UCLP treatment is to achieve a normalization of functions such as speech, growth of the naso-maxillary complex, arch dimensions and occlusion as well as facial appearance which is a multidisciplinary task [32]. The pediatric dentists have a responsibility towards the overall dental care of these children. They are often involved in the presurgical and postsurgical phase of maxillary orthopedics as numerous dental anomalies and malocclusions are encountered during the late primary and mixed dentition stage with UCLP. These malocclusions are either attributed to the congenital clefting itself or may be secondary to the surgical correction of the primary defects. Pediatric dentists can use both active and passive appliances to bring the cleft segments into a more ideal alignment and thereby promote a better initial surgical outcome [33].

In order to identify and implement the highest possible standards of care for UCLP children by the pediatric dentist, assessment of early treatment outcome after primary surgical repair of lip and palate regarding transverse dental arch relationship and occlusion is essential. The transverse dental arch relationship and occlusion of these children have been evaluated by few investigators in Egypt [34]. This presents a gap that impedes the delivery of proper dental care to these children. The present study aims at filling this gap by highlighting the main characteristics of transverse dental arch relationship and occlusion in surgically repaired UCLP children by the Oslo surgical protocol and compares them with those of healthy, comparable, non-cleft children to better meet the needs of this vulnerable group of children.

Material and Methods

The study was a comparative cross sectional study design. Thirty-one unilateral cleft lip and palate (UCLP) children (21 boys and 10 girls) with a mean age of 7.35 years together with thirty-one healthy, comparable, non-cleft children (20 boys and 11 girls) with a mean age of 7.13 years were recruited from Faculty of Dentistry, Alexandria University.

The inclusion criteria of the UCLP children were children of both genders, aging from four to nine years old, with surgically repaired UCLP according to the Oslo surgical protocol [22] (Table 1). Children with any systemic diseases, intellectual disabilities or syndromes and congenital anomalies other than UCLP were excluded from the study as well as UCLP children with previous naso-alveolar moulding, orthodontic treatment or bone graft.

Oslo surgical protocol	
PSOT*	No
3 months	Lip (Millard) and hard palate closure (single-layer vomer flap)
18 months	Soft palate closure (modified von Langenbeck)
8-12 years	Alveolar bone grafting

Table 1: Summary of the Oslo surgical protocol. * PSOT=presurgical orthopedic treatment.

As regards the non-cleft children (control group), healthy children free from any systemic diseases or syndromes, aging from four to nine-year old, free from oral habits and with limited or no crowding of teeth and no premature loss of teeth were included in this study.

Ethical approval for the study was first obtained from Dental Research Ethics Committee, Faculty of Dentistry, Alexandria University. The children's parents or guardians were asked to sign an informed consent. Participants were assured of the confidentiality of the collected data and that it was used only for research purposes.
Measurements were carried out by the researcher who was trained and calibrated to develop an acceptable intra-examiner consistency in assessing dental arch dimensions. Intra-examiner reliability was done using Kappa test which was 0.877 for all dental arch dimensions.

For each subject, demographic data were recorded and intra-oral examination was done to determine cleft side, presence or absence of palatal fistula and all erupted teeth. Upper and lower alginate impressions as well as a wax-bite under centric occlusion were taken and dental arch dimensions and arch relationships were recorded using the study casts.

As regards occlusion assessment, sagittal molar relation in primary dentition was determined as either flush terminal plane, mesial step or distal step. In mixed dentition, permanent molar relation was recorded according to angle classification as end-to end molar relation, class I, II or III. Sagittal cuspid relation was also classified according to Angle's into class I, II or III [35].

Transverse dental arch relationships were recorded using the modified Huddart/ Bodenham scoring system of bucco-lingual

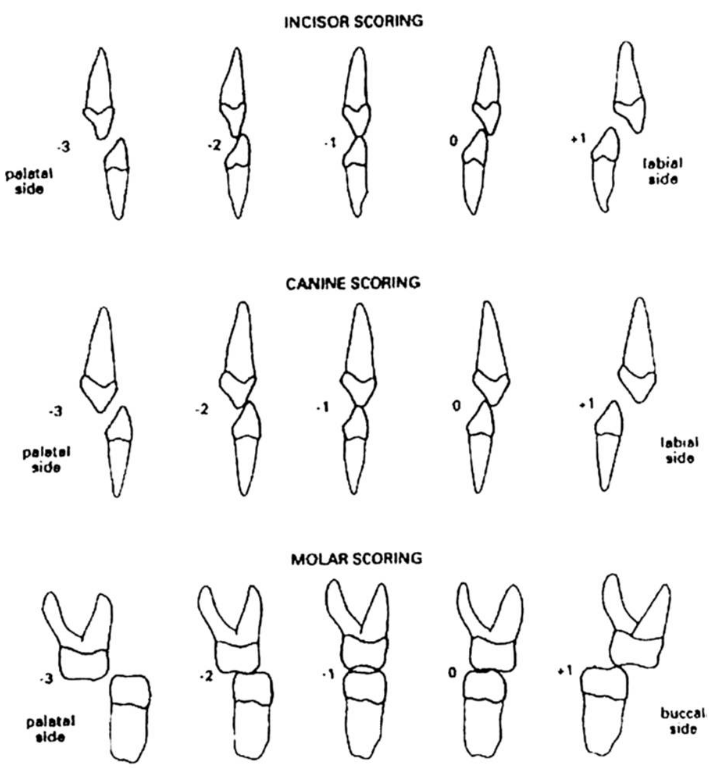

Figure 1: Modified Huddart/ Bodenham scoring instructions

dental relationships (Figure 1) [29]. The sum of the modified Huddart/Bodenham scores for a given model was described as the "Total arch constriction score".

Data were entered into an Excel file using patient identification numbers. Data were fed to the computer and analyzed using IBM SPSS software package version 20.0. Qualitative data were described using number and percent. Quantitative data were described using range (minimum and maximum), mean and standard deviation. As regards sagittal canine, primary and permanent molar relation, Fisher's exact test was conducted to compare between cleft group and non-cleft group. Independent samples t-test and Mann-Whitney test were used to compare Huddart/ Bodenham scores of both groups. Significance of obtained results was judged at the 5% level.

Results

The present study included 62 children, 31 children had unilateral cleft lip and palate (UCLP) and 31 matched non-cleft children. In an attempt to match the two groups, the controls were neighbors and school-mates (companions) of the UCLP children. In the UCLP group, males represented 67.7 % of the group, whereas 32.3% were females. In the non-cleft group, 64.5 % were males and 35.5% were females. Their age ranged between 4 and 9 years. The mean age was (7.35 ± 1.52 years) in UCLP children and (7.13 ± 1.52 years) in non-cleft children. No statistically significant differences between the two groups regarding gender (p=0.652) or age distribution (p=0.973).

Figure 2 shows the comparison between UCLP and non-cleft children regarding sagittal molar occlusion in primary dentition in age group 4-5 years. Straight terminal plane relationship was significantly higher in the non-cleft group (p=0.002), whereas mesial step was significantly higher in UCLP group (p=0.038).

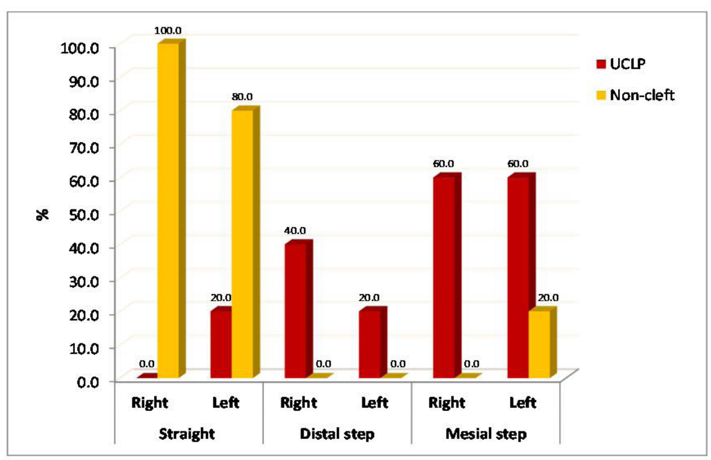

Figure 2: Comparison between UCLP and non-cleft children regarding sagittal molar occlusion in primary dentition (4-5 years)

Figure 3 shows the comparison between UCLP and non-cleft children regarding sagittal permanent molar occlusion in the mixed dentition in age group 6-7 years. Class I molar relation was significantly higher in the non-cleft group (p=0.004), whereas class III molar relation was significantly higher in the UCLP group (p=0.001).

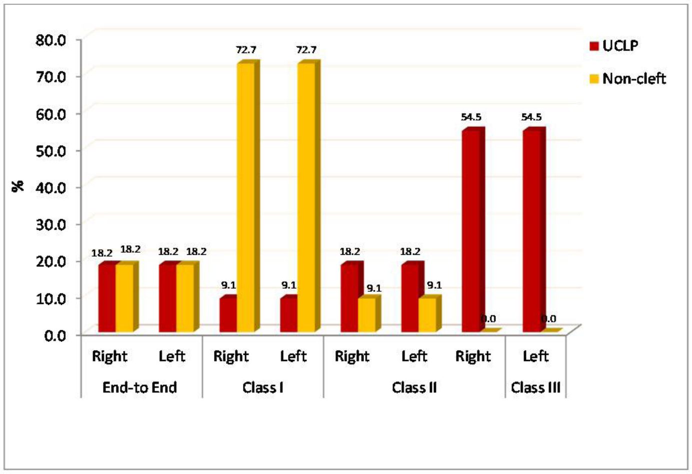

Figure 3: Comparison between UCLP and non-cleft children regarding sagittal molar occlusion in mixed dentition (6-7 years)

Figure 4 shows the comparison between UCLP and non-cleft children regarding sagittal permanent molar occlusion in the mixed dentition in age group 8-9 years. Among group comparisons showed that class I molar relation was significantly higher in non-cleft group (p=0.007), whereas class III molar relation and class II molar relation were significantly higher in UCLP group (p=0.001 and 0.046 respectively).

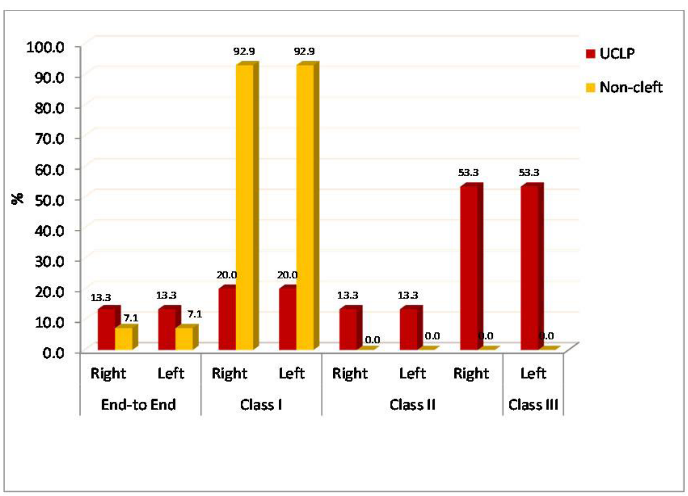

Figure 4: Comparison between UCLP and non-cleft children regarding sagittal molar occlusion in mixed dentition (8-9 years)

Regarding sagittal canine occlusion in primary dentition in age group 4-5 years; among group comparisons showed that class I cuspid relation was significantly higher in non-cleft group (p=0.002), whereas class III cuspid relation significantly predominated in the UCLP group (p=0.002). As for sagittal canine occlusion in the mixed dentition in age group 6-7 and 8-9 years; among group comparisons showed that class I cuspid relation was significantly higher in non-cleft group (p=0.001), whereas class III cuspid relation was significantly higher in UCLP group (p=0.001).

Table 2 shows the comparisons between the UCLP and non-cleft groups with respect to modified Huddart/Bodenham scores for central incisors, canines and molars in the age group of 4-5 years. In this age group, the mean total arch constriction score was -6.40 ± 9.45 in UCLP children which was not statistically significant from that of non-cleft group (p=0.216).

Age (4-5 years) Teeth		UCLP		Non-cleft		T	P
		Mean	SD	Mean	SD		
Central incisor	Right	-1.80	1.64	0.24	0.43	2.7	0.048*
	Left	-2.00	1.41	0.02	0.18	3.2	0.032*
Canine	Right	-2.80	0.45	-0.16	0.23	11.7	0.001*
	Left	-2.40	0.89	-0.10	0.22	5.6	0.004*
First primary molar	Right	-1.00	1.41	0.04	0.09	1.6	0.139
	Left	-0.40	0.55	-0.02	0.04	1.5	0.196
Second primary molar	Right	-0.80	1.10	0.06	0.59	1.5	0.172
	Left	-0.60	0.89	0.02	0.04	1.5	0.169
Total arch constriction score		-6.40	9.45	-0.20	0.45	1.5	0.216#

Table 2: Comparison between UCLP and non-cleft children with respect to modified Huddart/Bodenham scores for central incisors, canines and molars in the age group of 4-5 years.

t: independent samples t-test; **#:** Mann-Whitney test; * P<0.05 (significant).

Table 3 shows the comparisons between UCLP and non-cleft groups with respect to modified Huddart/Bodenham scores for central incisors, canines and molars in the age group of 6-7 years. The modified Huddart/Bodenham scores were noticeably more negative in the UCLP group. The mean total arch constriction score was -14.91±6.28 for the 6-7 years UCLP group and had distinctively more negative value than that of non-cleft group. The difference between groups was highly significant (p=0.001).

Age (6-7 years) Teeth		UCLP		Non-cleft		T	P
		Mean	SD	Mean	SD		
Central incisor	Right	-2.27	1.19	0.15	0.42	6.4	0.001*
	Left	-2.55	1.04	0.04	0.13	8.2	0.001*
Canine	Right	-2.45	0.52	-0.06	0.21	14.2	0.001*
	Left	-2.55	0.93	-0.25	0.30	7.8	0.001*
First permanent molar	Right	-0.50	0.85	0.34	0.61	2.6	0.019*
	Left	-0.70	0.82	0.21	0.29	3.3	0.007*
Second primary molar	Right	-1.27	1.19	-0.33	0.56	2.4	0.031*
	Left	-1.09	1.22	-0.04	0.10	2.8	0.017*
First primary molar	Right	-1.00	1.26	-0.05	0.12	2.5	0.032*
	Left	-1.18	1.17	-0.01	0.05	3.3	0.008*
Total arch constriction score		-14.91	6.28	-0.67	1.72	7.3	0.001*

Table 3: Comparison between UCLP and non-cleft children with respect to modified Huddart/Bodenham scores for central incisors, canines and molars in the age group of 6-7 years.

t: independent samples t-test; * P<0.05 (significant).

Table 4 shows the comparisons between UCLP and non-cleft groups with respect to modified Huddart/Bodenham scores for central incisors, canines and molars in the age group of 8-9 years. The mean total arch constriction score was -10.64 ± 8.01 for 8-9 years UCLP group, and had distinctively more negative values than that of non-cleft group. The difference between groups was highly significant (p=0.001).

Age (8-9 years) Teeth		UCLP		Non-cleft		T	P
		Mean	SD	Mean	SD		
Central incisor	Right	-1.93	1.33	0.01	0.08	5.6	0.001*
	Left	-1.73	1.28	0.01	0.07	5.3	0.001*
Canine	Right	-2.07	1.33	0.05	0.10	6.1	0.001*
	Left	-2.33	1.23	-0.04	0.22	7.1	0.001*
First permanent molar	Right	-0.53	1.64	0.11	0.18	1.5	0.151
	Left	-0.40	0.83	0.09	0.27	2.2	0.043*
Second primary molar	Right	-0.67	0.98	-0.03	0.17	2.5	0.025*
	Left	-0.87	0.92	0.01	0.34	3.5	0.003*
First primary molar	Right	-0.87	0.92	0.02	0.16	3.7	0.002*
	Left	-1.20	1.08	0.04	0.11	4.4	0.001*
Total arch constriction score		-10.64	8.01	-0.43	0.94	4.7	0.001*

Table 4: Comparison between UCLP and non-cleft children with respect to modified Huddart/Bodenham score for central incisors, canines and molars in age group 8-9 years.

t: independent samples t-test; * P < 0.05 (significant).

Discussion

Children with unilateral cleft lip and palate (UCLP) usually suffer from deficiency in growth of the naso-maxillary complex due to tissue deficiency, scar tissue formation after early reconstructive surgery and inherent growth retardation. Maxillary dental arch development is also retarded due to the naso-maxillary complex deficiency [13].

The surgical outcomes for the early repair of UCLP are highly variable and this can be attributed to several factors [36]. This fact, in addition to scarcity of data available in Egypt regarding early outcome assessment initiated this descriptive study to establish baseline information for the pediatric dentists to improve the standards of care available to this vulnerable group of children.

In the present study, UCLP children were divided into three critical age periods; namely primary dentition (4-5 years), early mixed dentition (6-7 years) and late mixed dentition (8-9 years). Among these age groups, different preventive and interceptive orthodontic interventions have been reported to improve the dental arch dimensions and occlusion for UCLP children [36].

A total of thirty one UCLP children (21 males and 10 females) with an age range 4-9 years were included. The mean age scores were 7.35 ± 1.52 years in UCLP children and 7.13 ± 1.52 years in non-cleft children. Males and females were pooled together the same way as in the normative data.

Since there are racial differences in development of dental arch and growth patterns, as stated by Lavelle 1975 [37], selection of matching control population to neutralize this variable is needed. Accordingly, the current study included a comparable sample of healthy matching non-cleft children. This could be considered a point of strength that distinguishes the present study from other studies, such as Sandy et al. in 1998 [38], Alam et al. in 2008 [39], and Fudalej P et al. in 2009 [22]. The mentioned studies [22,37-39], used the Five-Year-Old and Great Ormond Street, London and Oslo (Goslon) indices of dental arch deformity in examining early surgical outcomes in UCLP children. However, neither of these qualitative indices compares dental arch relationship with non-cleft data.

In the current study, dental arch relationship was scored using modified Huddart/Bodenham index because it is more objective, reliable, and more sensitive to inter-arch discrepancies than the Five-Year-Old and Goslon indices as was reported by Mossey et al. in 2003 [29]. Furthermore, the severity of the cross-bite which is also taken into account is easier to assess statistically [29,40].

As regards the sagittal molar and canine occlusion, the results revealed a predilection for most of UCLP children in all age groups to have mesial step terminal plane in primary dentition, class III permanent molar relation in mixed dentition and class III cuspid relations. This finding was statistically significant compared to non-cleft children who mostly exhibited flush terminal plane in primary dentition, class I permanent molar relation in mixed dentition and class I cuspid relation. This is probably due to the deficiency in maxillary dental arch in UCLP children. However, there were few exceptions in each age that happened to have distal step terminal plane or Class II permanent molar and cuspid relations. Skeletally developing class II which is genetically inherited or mesial migration of upper permanent molars due to congenitally missing teeth could be the main causes of these exceptions.

In the present study, the Modified Huddart/Bodenham index analyzed the transverse dental occlusion and the results were in accordance with transverse distance measurements between canines and molars. In the age group 4-5 years, the modified Huddart/Bodenham score was only significantly different for the central incisors and canines. The mean total arch constriction score was -6.40; however it was not significantly different from that of non-cleft group. In the older age groups, 6-7 years and 8-9 years, the modified Huddart/Bodenham score was significantly different for all measured teeth and the mean total arch constriction scores were more negative and significantly higher than in the non-cleft groups. These findings agree with the results reported in earlier studies performed by Garrahy et al. in 2005 [17], and Stein et al. in 2007 [41].

Considering the finding that modified Huddart/Bodenham scores were more negative, the transverse correction using rapid palatal expanders is needed and should be completed before performing secondary bone graft in order to guarantee stabilization of the dental arch at a correct width. In addition, more comprehensive orthodontic measures are necessary in the presence of pronounced cross-bite.

Conclusion

Based on the results of present study, it can be concluded that:

- There was a tendency towards mesio-occlusion in UCLP children (mesial step terminal plane, class III permanent molar and cuspid relation).
- Modified Huddart/Bodenham scores revealed that UCLP children suffered from constricted maxillary arch in all age groups especially in the canine region.

Recommendations

Further research should be initiated to assess arch dimensions and occlusion following different interceptive orthodontic measures and alveolar bone grafting in attempt to improve surgical outcomes. In addition, characteristics of arch dimensions and occlusion in children with different types of cleft lip and palate in Egyptian children should be also investigated.

Why this paper is important to pediatric dentists?

- This paper has shown that unilateral cleft lip and palate Egyptian children show arch constriction varying in severity and malocclusion that pediatric dentist should be acquainted with as they are part of the multidisciplinary team responsible for management of this group of children.
- Different dental arch features have been shown through out this study that pediatric dentist should be oriented with in order to provide best treatment outcome for this vulnerable group of children.
- According to the results of this pediatric dentist can provide both active and passive appliances to bring the cleft segments into a more ideal alignment promoting better initial surgical outcome.
- Future studies can compare the outcome after orthodontic treatment of this group of Egyptian children using different appliances with the results of this study to reach the best offered line of treatment.

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