

Dental maturation of unilateral cleft lip and palate

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ABSTRACT

Cleft lip and palate (CLP) is the most common craniofacial abnormality and the fourth most common birth defect in Singapore. Many reports suggest that CLP children have delayed dental development and asymmetrical timing of tooth-pair formation. The aim of this study was to investigate the timing of development of permanent teeth in unilateral CLP (UCLP) children and to compare the findings with non-CLP children in Singapore. A total of 60 UCLP children aged between 5 and 9 years (mean 6.64 ± 0.90 years) and a non-CLP control group matched for age, gender, and race were investigated and compared. Dental records and radiographs were studied and the dental maturation was determined using the Demirjian's method (1973). The dental maturation of UCLP children were delayed compared with non-CLP children by a mean of 0.55 ± 0.75 years and the delay was statistically significant ($p < 0.001$). The UCLP group also had significantly higher risk of asymmetrically developing tooth pairs than the control group ($p < 0.001$). The most commonly delayed tooth in development was the maxillary cleft-sided lateral incisor. In conclusion, the UCLP children in Singapore demonstrated delayed dental maturation and a higher occurrence of asymmetrical tooth-pair formation than the non-CLP children.

Keywords: Cleft lip and palate, dental age, tooth development

INTRODUCTION

Cleft lip and palate (CLP) is the most common cranio-facial abnormality^[1] and the fourth most common birth defect in Singapore.^[2,3] The incidence of CLP in Singapore was found to be 1.87 per 1000 live births.^[4] CLP patients are reported to be commonly associated with delayed dental development and asymmetrical timing of tooth formation.^[5]

Several investigators reported delayed formation of the permanent teeth in CLP patients and the delay was observed to vary from 0.3 to 0.9 year.^[6-11] Bailit *et al.* found that tooth formation in 39 children with cleft palate was significantly retarded by about 0.7 year when compared with 36 control subjects.^[6]

Ranta in his earlier study compared 258 CLP Finnish children with 1162 noncleft children and reported a delay in tooth formation of 0.5 year in the maxilla and 0.4 year in the mandible, but the difference was not statistically significant.^[8] Ranta went on further

to conduct other investigations and revealed that the delay in tooth formation increased from 0.3 to 0.7 year with increasing severity of the cleft deformity.^[12] He also found that the dental development was more delayed in the cleft subgroup with hypodontia (0.7 year) than in the subgroup without hypodontia (0.4 year), and a longer delay in tooth formation was observed with increasing number of missing teeth per child.^[13]

Harris and Hullings studied 54 CLP children and reported an overall delay in dental development of 0.9 year.^[9] They also noted that teeth formed during the early postnatal period were the most affected, while the later forming teeth were less delayed.

In a recent investigation, Lai *et al.* based their study on 231 southern Chinese CLP children from Hong Kong and compared them with a non-CLP control sample of the same size.^[11] Similarly, they found an overall delay in tooth formation (0.4 year) of Chinese CLP children with the earlier formed permanent teeth being more delayed in development than those formed later.

In accordance to the findings of Ranta,^[12] CLP children with increased severity of hypodontia also displayed a greater delay in dental maturation.

A pair of teeth was regarded as developing asymmetrically when the crown or root development of one of the teeth deviated from that of the antimeric tooth by at least one developmental stage. Ranta was one of the earliest authors to report on asymmetric tooth formation.^[8] Studies have found that children with CLP had asymmetrical tooth formation that was three^[9] to four^[11] times more common than those of the control group.

When considering individual teeth, some teeth seem to display a greater propensity for asymmetric formation. Ranta reported that asymmetric tooth development occurred most frequently in the upper central incisors followed by the upper and lower premolars, without taking into account peg-shaped teeth and third molars.^[8,14] Harris and Hullings excluded the incisors in their study and found that second premolars and third molars were more likely candidates for asymmetric formation and these teeth were also more likely to be congenitally missing.^[9]

Comparing cleft and noncleft sides of the maxilla, Ranta found that the more delayed teeth of the asymmetrically developing tooth pairs occurred four times more frequently on the cleft side than on the noncleft side.^[8] Lai *et al.* also showed similar delays in the cleft side, and the most prevalent delayed tooth was the maxillary cleft-sided lateral incisor.^[11]

Ranta's study also investigated the difference in occurrence of asymmetric tooth development between the maxilla and mandible. In the cleft palate group, asymmetry occurs with equal frequency in both jaws. However, in the cleft lip and alveolus group as well as the CLP group, asymmetry occurs more frequently in the maxilla.^[8,14]

Most of the published studies in the English language, investigating the dental development of CLP patients have been based on Caucasian populations thus far, with the exception of one study by Lai *et al.* on a southern Chinese population in Hong Kong.^[11] To date, no studies have been carried out to investigate the dental development of CLP children in Singapore. Hence, this study aims to investigate and compare the dental maturation of unilateral CLP (UCLP) children with non-CLP children in Singapore.

MATERIALS AND METHODS

Study sample

Ethics approval was obtained from the Sing Health Institutional Review Board for this research (CIRB reference code: 2010/070/D). The investigation was based on retrospective records of CLP and non-CLP patients from the Department of Orthodontics at the National Dental Centre of Singapore (NDCS).

The inclusion criteria of the CLP study sample were:

1. Patients with repaired UCLP
2. Aged between 5 and 9 years
3. Born and living in Singapore
4. Complete dental records consisting of treatment record notes and orthopantomograms (OPGs)

The exclusion criteria of the CLP subjects were:

1. Inadequate dental records and missing radiographs
2. Bilateral clefts
3. Clefts presenting as part of a syndrome, or medical conditions suggestive of a syndrome
4. Bilaterally missing mandibular teeth

The subjects in the non-CLP control group were selected by matching with the UCLP subjects for:

1. Age and date (within 60 days) of OPGs taken
2. Gender
3. Ethnic group

Similarly, the subjects in the control group were excluded if they had any of the following:

1. Documented medical syndromes
2. Bilaterally missing mandibular teeth

A total of 60 UCLP and non-CLP patients were obtained for the study using the inclusion and exclusion criteria.

All the records were studied by a single operator, the first author of this study. Every subject's clinical records were examined for chronological age; gender; medical, dental, and social histories; details of diagnosis; treatment planning and treatment procedures rendered, such as tooth extraction. The OPGs were viewed from an illuminated viewing box in a darkened room.

Dental maturation

The developing stages of the permanent teeth were determined from the OPGs using the method described by Demirjian *et al.*^[15] Prior to the conduct of the study, the operator first underwent a pre-experimental calibration module using the Demirjian's Dental Development CD-Rom (1993–1994).

To determine the maturity of the seven mandibular teeth on the left side (excluding the third molar), Demirjian *et al.*^[15] established a set of criteria to compare their radiographic appearances with a sequence of reference radiographs, diagrams and description of formation stages. Each tooth was divided into eight formative stages (A to H), and each stage was allocated a score depending on the gender. A maturity score is then derived from the addition of the scores of all the seven teeth. This maturity score can be converted directly into a dental age by reading off a percentile curve the age at which the 50th percentile attains the maturity score value, or by using a pre-constructed table.

The mean chronological age and mean dental age of the UCLP and control group were determined. The mean dental age difference was calculated by subtracting the mean dental age from the mean chronological age (mean dental age difference = mean chronological age – mean dental age). The mean dental age difference between the UCLP and control group were then compared to determine if any significant delay in dental development occurred in the UCLP group (mean dental age difference [UCLP] – mean dental age difference [non-CLP]).

Asymmetric tooth formation

The developmental stages of every individual tooth, with the exception of the third molars were determined using the Demirjian’s method.^[15] The dental maturation status within each tooth pair was compared to identify presence of asymmetrical tooth development, and the delayed tooth in each pair was noted.

Intra-examiner reliability

One month after the study was completed, 15 OPGs were randomly selected from both the UCLP and control group. The maturity stages of the seven mandibular left teeth were re-determined using the Demirjian’s method^[15] to establish intra-examiner reliability.

Statistical analysis

The statistical analyses were performed using SAS 9.2 (SAS Institute Inc., Cary, NC). Intra-examiner reliability for determining the teeth formation stages using the Demirjian’s method^[15] was analyzed using the Kappa coefficient (k). Paired t-test was used to evaluate the comparison in the mean dental age delay between UCLP and non-CLP groups.

The dental age delay of UCLP subjects with and without hypodontia was compared using the Mann–Whitney U test to determine if the presence of hypodontia affected dental development. In addition, Spearman’s correlation coefficient was also performed to evaluate the relationship between the severity of hypodontia and dental age delay.

Poisson regression analysis with corrected multiplicative dispersion factor was used to compare the risk of asymmetric tooth pairs between the UCLP and non-CLP groups and to evaluate the risk of delayed tooth on tooth number, site (maxilla/mandible), and side (cleft/noncleft). $p \leq 0.05$ was taken as significant.

RESULTS

Demographics

A total of 60 UCLP subjects in the experimental group consisted of 24 right UCLP and 36 left UCLP. There were 36 boys and 24 girls, 51 Chinese, 7 Malays, and 2 Indians with a mean chronological age of 6.64 ± 0.90 years. The control group consisted of the same number of non-CLP subjects, matched for gender, race, and age.

Intra-examiner reliability

The Kappa coefficients for the seven mandibular left teeth ranged from 0.70 to 1.00 [Table 1], showing substantial agreement to almost perfect agreement.

Table 1: Kappa coefficients for seven mandibular left teeth	
Tooth	Kappa coefficient with 95% CI
# 31	0.91 (0.75–1.00)
# 32	0.90 (0.72–1.00)
# 33	0.91 (0.74–1.00)
# 34	0.91 (0.75–1.00)
# 35	0.91 (0.74–1.00)
# 36	1.00
# 37	0.70 (0.40–1.00)

The levels of agreement between the first and second measurements of the tooth developmental stages for the individual seven mandibular left teeth are as follows:

1. For teeth #31–#35: Substantial agreement to perfect agreement
2. For tooth #36: Perfect agreement
3. For tooth #37: Fair agreement to perfect agreement.

Dental maturation of UCLP and non-CLP children

The mean chronological age for both the UCLP and non-CLP children was $6.64 + 0.90$ years. The mean dental ages for UCLP and non-CLP children were $6.97 + 0.83$ years and $7.52 + 0.92$ years, respectively. Both the UCLP and non-CLP children had mean dental ages ahead of their mean chronological ages with a mean dental age delay of $-0.32 + 0.62$ year and $-0.88 + 0.55$ year, respectively [Table 2].

This UCLP group was delayed in dental maturation compared to the non-CLP group by a mean of $0.55 + 0.75$ year and this delay was found to be statistically significant ($p < 0.001$) [Table 3].

Hypodontia and dental development

There were 38 UCLP children with hypodontia in the permanent dentition. There was no significant difference in the dental age delay between UCLP children with and without hypodontia ($p = 0.602$). Furthermore, Spearman’s correlation coefficient test showed no linear relationship between the number of missing teeth and the dental age difference in UCLP patients with hypodontia.

Asymmetric tooth formation

The UCLP group had a total of 791 tooth pairs, out of which, 187 (23.6%) developed asymmetrically. The non-CLP group had a total of 829 tooth pairs, out of which, 52 (6.27%) developed asymmetrically.

Poisson regression analysis showed that the UCLP group had a significantly higher risk of asymmetric tooth pairs compared with the non-CLP group (Relative Risk [RR]= 3.77, 95% Confidence Interval [CI] 2.77–5.12; $p < 0.001$).

Considering the teeth on the cleft side, it was found that the teeth on the maxilla had a significantly higher risk of delayed development of teeth compared with the teeth on the mandible (RR=2.39, 95% CI 1.11–5.17; $p = 0.027$). However, no statistically significant difference was found in the dental maturation of the upper and lower jaws on the noncleft side (RR=0.78, 95% CI 0.36–1.66; $p = 0.516$).

In both maxilla and mandible, the cleft side has a significantly higher risk of delayed development of teeth than noncleft side ($p < 0.001$; maxilla: RR= 5.28, 95% CI 2.41–11.55; mandible: RR= 1.85, 95% CI 1.40–2.44).

The most commonly delayed tooth in the maxilla is the cleft-sided lateral incisors (73.3%), followed by the cleft-sided central incisors (37.3%). In the mandible, the cleft-sided canines and first premolars were the most frequently affected (21.7%) [Table 4].

Table 2: Differences between chronological and dental ages of UCLP and non-CLP children

	Mean chronological age		Mean dental age		Mean dental age difference	
	Mean (years)	SD	Mean (years)	SD	Mean (years)	SD
UCLP	6.64	0.90	6.97	0.83	-0.32	0.62
Non-CLP	6.64	0.90	7.52	0.92	-0.88	0.55

Table 3: Paired t-test comparing mean dental age difference between UCLP and non-CLP group

	Mean	SD	95% confidence interval of the difference		p value
			Lower	Upper	

DISCUSSION

Most published studies in the English language, investigating dental development of CLP patients were based on Caucasian populations thus far, with the exception of one study by Lai et al., which was conducted on a southern Chinese population in Hong Kong.^[11] This retrospective study investigating the dental development of UCLP children is the first to be carried out in Singapore.

There were several published reports on dental formation and dental age of CLP patients, in which the authors did not group samples by cleft types.^[8,10,11,16,17] This introduces confounding factors as different cleft types of different severities may exert varying influences on the cleft area, affecting the prevalence of dental anomalies, dental development, and overall growth. Cleft palate has also been proposed to be developmentally and genetically different from cleft lip, with or without cleft palate.^[18,19] These differences may likewise affect the dental development in various ways.

Furthermore, it will not be possible to compare asymmetric tooth formation between cleft and noncleft side in patients with bilateral CLP. Hence, this study was limited to UCLP patients to reduce the confounding factors, and to facilitate comparisons with future studies on similar cleft types.

Most of the earlier studies did not have control groups, or compared the cleft groups to unmatched groups of control subjects. This study used age, gender, and race-matched controls from the same country to minimize confounding factors.

Demirjian’s method

The Demirjian’s method^[15] is used in this study as it is an accurate and precise estimation of dental ages, particularly during early childhood.^[20,21] Prior to the start of the study, pre-experimental calibration was carried out to reduce intra-examiner variability. In this study, the intra-examiner calibration performance score of 86.2% was achieved, which compares well with the inter-examiner reliability of 80% reported by Levesque and Demirjian.^[22]

Table 4: Distribution of developmentally delayed teeth

Tooth		Maxilla		Mandible	
		Cleft n (%)	Noncleft n (%)	Cleft n (%)	Noncleft n (%)
		Central incisor	22 (37.29)	1 (1.69)	5 (8.33)
Lateral incisor	22 (73.33)	0 (0.00)	7 (11.67)	4 (6.67)	
Canine	7 (11.86)	2 (3.39)	13 (21.67)	4 (6.67)	
First premolar	11 (18.97)	4 (6.90)	13 (21.67)	4 (6.67)	
Second premolar	9 (19.57)	3 (6.52)	6 (10.17)	5 (8.47)	
First molar	11 (18.33)	4 (6.67)	3 (5.00)	2 (3.33)	
Second molar	13 (22.03)	4 (6.78)	1 (1.67)	1 (1.67)	

The Kappa coefficients for the seven mandibular left teeth showed substantial agreement to perfect agreement between the first and second measurements, indicating that the reproducibility of Demirjian’s method^[15] was high.

However, the 95% CI for Kappa coefficient of the mandibular left second molar showed a wider range (0.40–1.00) from fair agreement to perfect agreement, compared with the other six mandibular left teeth (0.74–1.00), which ranged from substantial agreement to perfect agreement. Levesque and Demirjian also reported a higher discrepancy in the evaluation of second molar developmental stages at 6 years of age. The reason could be because most of the second molars were at developmental stages C and D at the time of evaluation, and it has been shown that delimitation between stages C and D requires more investigation than other stages.^[22]

Dental maturation

The mean delay in dental maturation of the UCLP children compared to non-CLP children was 0.55 + 0.75 year and falls within the range of 0.3–0.9 year previously reported.^[6–11]

Hypodontia and dental maturation

This study found that the presence and extent of hypodontia in UCLP patients did not have an effect on the dental maturation. This was contrary to the results of Ranta^[13] and Lai et al.^[11] who found a longer delay in dental development in the cleft subgroup with hypodontia than in the subgroup without hypodontia, and the more severe the hypodontia, the longer the delay.

Ranta’s study sample only included patients with isolated cleft palate while Lai et al.’s study combined patients of various cleft types together; it was discussed earlier that isolated cleft palate is

etiologically different from other cleft types. Ranta also found that the delay in tooth formation increased with increasing severity of the cleft and the delay was severe in the cleft palate group.^[1,2] Furthermore, increasing cleft severity has also been found to correlate with increased prevalence of hypodontia.^[5,16,23,24] Due to the confounding factors in Ranta and Lai's study, the correlation between hypodontia and dental development could not be substantiated.

Asymmetric tooth-pair formation

The risk of the UCLP group with asymmetric tooth pair formation was 3.8 times higher than the non-CLP group, and this was found to be statistically significant ($p < 0.001$). This finding was similar to other studies which reported that CLP children had asymmetrical tooth formation that was 3 (9) to 4 (11) times more common than those of the control group.

Delayed tooth formation

The teeth exhibiting retarded development in the asymmetrically developing tooth-pairs occurred at a significantly higher risk on the cleft side in both the maxilla and the mandible, a finding also reported by Ranta^[6] and Lai *et al.*^[11] The commonly delayed teeth in this study were the cleft-sided lateral and central incisors in the maxilla and cleft-sided canines and first premolars in the mandible.

Considering the teeth on the cleft side, of the maxilla had a significant higher risk of delayed development compared to the teeth on the mandible. However, there was no statistically significant difference in the maturation of the maxillary and mandibular teeth on the noncleft side.

Since the cleft defect is present only in the maxilla, this finding further reinforces the possible association of common genetic factors between clefting and delayed tooth formation.^[25] It has been postulated that surgical cleft repair results in fibrosis and reduced blood supply, causing damage to the developing tooth buds at the cleft area.^[6] Other etiological factors for delayed tooth formation in the maxillary cleft side include lack of space for tooth formation in the cleft area^[26] and growth attenuation due to improper nutrition.^[6]

CONCLUSION

The UCLP children in Singapore aged 5–9 years demonstrated significant delay in dental maturation by a mean of 0.55 + 0.75 year compared with a matched control group of non-CLP children. The UCLP children also had a significantly higher risk of asymmetrical tooth-pair development compared with the non-CLP children. The most commonly delayed tooth in development was the maxillary cleft-sided lateral incisor.

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