Scientific Article

# Developmental dental changes in isolated cleft lip and palate

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## Abstract

*Isolated cleft lip and/or palate, CL(P), may be associated with multiple changes in the developing dentition. To test the hypothesis that permanent tooth formation is delayed in patients with CL(P), dental maturity was assessed from panoramic radiographs. The dental maturity ratio (DMR, dental age divided by chronological age) of a group of CL(P) patients (23 girls, 30 boys) was compared with matched control subjects (38 girls, 41 boys). The mean DMR in cleft boys (0.97 ±0.01) was significantly lower than in control boys (1.06 ±0.01), P < 0.05. The mean DMR in cleft boys was lower than in cleft girls (1.02 ±0.02), with a tendency toward statistical significance. No significant difference in DMR was found between the cleft versus control girls, or between the control girls and boys. Among cleft boys, the prevalence of dental age delay was 67% (20/30), with a mean delay of 0.6 ± 0.4 years. These results suggest that CL(P) may be associated with delay in permanent tooth formation. (Pediatr Dent 19:109–13, 1997)*

Just as orofacial clefting may affect the development of the nasal airway or the hearing and speech apparati, it can also affect the developing dentition. Initial observations leading researchers to scrutinize this particular aspect of clefting were several reports that teeth immediately adjacent to the cleft (the primary and permanent central and lateral incisors) showed significant abnormalities that occurred at random stages of development.[1,2] Adverse factors during embryonic developmental stages leading to cleft lip and/or palate also may affect development of both primary and permanent dentitions. Teeth adjacent to the cleft are most likely to be affected, but other teeth may also be affected.[2] A better understanding of cleft-associated dental abnormalities might lead to better treatment strategies for cleft-affected children.

Kramer et al.[3] studied the mean emergence time of the primary maxillary incisors, including the cleft-side lateral incisor in the distal cleft segment and in the premaxilla, and that of the mandibular incisors. Compared with normal control children, an emergence delay of 8 months was found for the lateral incisors on the cleft side in the distal segment in children with a cleft lip and alveolus, and 13 months in cleft lip and palate children. The delay was increased by 4 months for lateral incisors located in the premaxillary segment. No delay was observed for the lateral incisors in the noncleft side, the maxillary central incisor, and the mandibular lateral incisors.

Ranta[2] reported delays in the timing of tooth formation in the permanent dentition of children in all cleft groups compared with normal children. The delays were estimated at 0.3 years on average, and found to be similar for all permanent teeth in both the maxilla and mandible. Delays in tooth formation increased to approximately 0.7 years with increasing severity of clefting, and lengthened even more with associated oligodontia and in the older age group. Eruption of primary teeth did not seem to be affected significantly by CL(P). There was also a slight asymmetry in tooth emergence in unilateral cleft lip and palate, as teeth on the cleft side were delayed compared with the noncleft side.[4]

Harris and Hullings[5] found a significant delay in dental development in the permanent dentition situated away from the cleft site in children with isolated cleft lip and palate. They also found that the teeth formed early during the postnatal period (first molars) were most affected, while those formed later (premolars) were less affected, and the teeth that formed latest (second and third molars) were least affected.

Besides clefting, with its multiple associated malformations, other risk factors for delayed development of the dentition have been studied. They included low birth-weight, short gestation, mother's use of drugs during pregnancy, birth order, and relatives having malformations other than oral clefts. However, Poyry and Ranta[4] found only weak association between these other risk factors and tooth development.

This controlled study investigated the dental development of a group of cleft lip and palate children, using a novel method of comparing dental and chronological age — the dental maturity ratio (DMR).

## Subjects and methods

### Subjects with cleft lip and palate

One hundred and eighty-two patients with isolated CL(P) , who had been treated previously or were undergoing comprehensive treatment at Children's Hospital in Boston, were available for prospective consecutive enrollment in this study. Patients with CL(P) as part of syndromes (e.g., Pierre-Robin sequence, hemifacial microsomia, etc.) were excluded. Patients with pre-existing panoramic radiographs obtained during their mixed-dentition stage were grouped for determination of dental maturity. This group included 53 children (23 girls and 30 boys) ranging in age from 4.5 to 11.7 years for girls (average age 7.7 years) and 3.8 to 11.9 years for boys (average age 8.4 years). Except for one radiograph obtained from a private dental office, all panoramic radiographs were exposed using the Planmeca™ unit (Planmeca SF-00810, Helsinki, Finland). The radiographs were processed under standardized conditions for developing and fixing time, temperature, and age of the solutions (5.5 min, 28°C).

## Control children

Seventy-nine healthy normal children were selected randomly to serve as controls from the general pediatric population in the metropolitan Boston area using existing dental records available at a private pediatric dental office. The control group was matched in race, gender, and age with the cleft group. Subjects with any documented peri- or postnatal medical conditions were excluded. There were 38 girls, ranging in age from 3.7 to 11.9 years (average age 8.2 years) and 41 boys, ranging from 3.5 to 11.8 years (average age 8.3 years).

## Evaluation of dental maturity

In an effort to provide a standardized system to evaluate and score dental maturity, Dermirjian et al.[6] reported a highly reproducible method using panoramic radiographs to score different stages of a permanent tooth calcification. Eight defined stages of development, from the tip of the cusp to closure of the apex, were identified, with specific criteria described. All seven permanent teeth on the mandibular left side of the radiographs (central incisor to second molar) were assigned a weighted maturity score at the different stages of development. The individual maturity scores for all seven teeth were added to yield a total maturity score, which was then converted to a dental age based on percentile curves. The lower left of the mandible was used because it showed the least distorted and clearest image. A demonstrated high degree of correlation of the development stages between the left and right, maxilla and mandible, allowed the representation of the entire dentition by the one quadrant of teeth.[6]

Using the above described method, a dental maturity score was obtained and the dental age was determined for each subject. Dental age delay was defined as dental age minus chronological age to indicate whether a subject was dentally delayed (< 0 ), advanced (> 0 ), or normal (= 0). Additionally, a dental maturity ratio (DMR) was defined as the dental age divided by the chronological age. Thus, the DMR indicated whether a subject was dentally delayed (< 1), advanced (> 1), or normal (= 1).

## Statistical analyses

The normal theory *t*-test and the nonparametric Wilcoxon's rank sum test were used to assess differences in dental age delay between groups (control boys versus cleft boys; control girls versus cleft girls). The two-sample *t*-test was used to assess differences in DMR between groups. Two-way analysis of variance was performed on the DMRs between the groups to determine whether gender and/or cleft was significant. For all tests, a $P$-value of $< 0.05$ was considered statistically significant.

Calibration of the dental age scoring method was accomplished with the joint review of five randomly chosen panoramic radiographs (representing 10% of the entire cleft sample) by two examiners (AP and SS). ANOVA demonstrated no significant difference between the DMRs reported by the two examiners ($P > 0.05$). Similarly, intraexaminer variability of the author (AP) assessing dental maturity was not statistically significant ($P > 0.1$).

# Results

Table 1 summarizes the number of cleft and control subjects who had panoramic radiographs appropriate for determining dental maturity according to age distribution and cleft characteristics. The means and stan-

TABLE 1. DENTAL MATURITY RATIO (DMR) IN CLEFT AND CONTROL CHILDREN

| | *Cleft* | | *Control* | |
|---|---|---|---|---|
| | *Females* | *Males* | *Females* | *Males* |
| *Age Groups* | | | | |
| 3.0–6.0 | 5 | 5 | 6 | 6 |
| 6.1–8.0 | 10 | 5 | 14 | 10 |
| 8.1–12.0 | 8 | 20 | 18 | 25 |
| **Total** | **23** | **30** | **38** | **41** |
| *Type of Cleft* | | | | |
| Incomplete alveolar | 4 | 3 | | |
| Complete alveolar | 1 | 2 | | |
| Complete cleft lip and palate | 17 | 22 | | |
| Combination types of cleft* | 1 | 3 | | |
| *Dental Maturity Ratio* | | | | |
| Mean | 1.02 | 0.97† | 1.04 | 1.06† |
| Standard deviation | 0.09 | 0.07 | 0.09 | 0.09 |
| Range | 0.81–1.18 | 0.81–1.19 | 0.83–1.24 | 0.83–1.26 |

* Combination types of cleft include: complete cleft of the secondary palate with bilateral lip and alveolar notching (female), complete right cleft lip and palate with contralateral incomplete cleft lip (male), and complete bilateral cleft of the primary palate. † Significant statistical difference between mean DMRs of cleft and control boys. $P = 0.001$.

dard deviations for the dental maturity ratio also are reported.

Of the 20 boys with delayed dental age, 10 had unilateral complete cleft lip and palate (mean dental age delay = -0.7 ± 0.5 years), six had bilateral cleft lip and palate (mean delay = -0.5 ± 0.2 years), three had unilateral cleft lip (mean delay = -0.2 ± 0.2 years), and one had bilateral cleft lip, alveolus, and palate (delay = -1.4 years). The prevalence of dental age delay among cleft boys was 67% (20/30), with a mean delay of -0.6 ± 0.4 years.

Both the normal theory *t*-test and the nonparametric Wilcoxon's test showed that the dental age delay for cleft boys was different from that for control boys (0.25 ± 0.11 year versus -0.57 ± 0.12 year, $P < 0.05$) and that there were no statistical differences in dental age delay between cleft girls and control girls. Similar results were obtained from the analysis of DMR, as summarized in Table 1. Two-sample *t*-tests showed that cleft boys were delayed compared with control boys, with a highly significant difference in the DMR (0.97 ± 0.01 versus 1.06 ± 0.01, $P < 0.001$). No significant differences were found in the DMR between the female cleft and control groups (1.02 ± 0.02 versus 1.04 ± 0.01, $P > 0.05$) or between the female and male control groups (1.04 ± 0.01 versus 1.06 ± 0.01, $P > 0.05$). However, there was a tendency toward significant difference between the female and male cleft groups ($P = 0.059$).

The frequency distributions in Figs 1 and 2 showed a normal distribution of DMR for the control populations (boys and girls), centering approximately at DMR = 1.05. Cleft boys, however, exhibited skewing of the curve toward DMR < 1, while cleft girls showed a near normal distribution.

The dental age delay in cleft subjects relative to control subjects may be observed qualitatively from scatterplots of paired dental and chronological age data (Figs 3 and 4). In Fig 3, dental age delay in cleft boys is more apparent in the older age group (8–12 years) than in the younger age group (< 8 years). Dental age delay in cleft girls is less evident (Fig 4), although the linear regressions indicate a possible tendency for cleft girls to be slower in dental age (slope 0.92 ± 0.09 versus 1.05 ± 0.05).

Two-way ANOVA between groups revealed that the gender factor did not have a significant effect on the DMR ($P > 0.05$), but that the cleft factor did ($P = 0.001$). A borderline *P*-value ($P = 0.051$) indicated that there was some interaction between the gender and cleft factors to affect the DMR.

TABLE 2. PARAMETERS FOR THE LINEAR REGRESSIONS OF DENTAL AGE TO CHRONOLOGICAL AGE

| *Parameters* | *Control Girls* | *Cleft Girls* | *Control Boys* | *Cleft Boys* |
|---|---|---|---|---|
| Slope | 1.05 ± 0.05 | 0.92 ± 0.09 | 1.12 ± 0.06* | 0.90 ± 0.05* |
| Constant | -0.05 ± 0.45 | 0.71 ± 0.71 | -0.42 ± 0.56 | 0.55 ± 0.43 |

* Combination types of cleft include: complete cleft of the secondary palate with bilateral lip and alveolar notching (female), complete right cleft lip and palate with contralateral incomplete cleft lip (male), and complete bilateral cleft of the primary palate.

## Discussion

This study employed a novel method of analyzing dental development using the dental maturity ratio, which is defined as the dental age divided by the chronological age. Previous studies[5] have shown that rate of dental development in normal children may vary among the age groups. Thus, the use of the DMR in this sample with a wide range of chronological age groups eliminates any possible skew, which can be shown by arithmetic differences between chronological and dental ages.

This study demonstrated that boys with clefts were slower in dental age than control boys. In addition, cleft boys tended to be more dentally delayed than cleft girls, although the difference was not statistically significant. These findings support other observations made regarding dental delays in boys with clefts. Prahl-Andersen[7] reported a tendency for cleft children to show delayed tooth formation until age 9 years, with a greater delay in boys than in girls. Brouwers and Kuijpers-Jagtman also found a significant delay in the formation of permanent teeth in both boys and girls with unilateral cleft lip and palate, but boys were significantly delayed across all ages, while girls were delayed at only a few ages.[8] It is unclear why boys with clefts are more dentally delayed than girls with clefts. Dermirjian has observed that the mechanisms controlling dental development are independent of somatic and/or gender maturity, and that these unknown controlling factors seem to be highly influenced by the same etiologic factors of clefting.[9] The fact that boys mature slightly later than girls and are also slower to mature dentally than girls, was not seen among the control groups in this study because the Dermirjian's method has already corrected for this difference.[6]

Dental age delay among the older age group (9 years and older) has been observed,[4] although there has been no explanation for the observation. As discussed earlier, underestimation of dental age may occur in the older age group due to teeth that have completed growth. However, as shown in Figs 3 and 4, scattergraphs of dental age versus chronological age do not show leveling or decrease of dental age in the older age groups.

Of the 20 boys with delayed dental age, delay ranged from 0.2 to 1.0 years with a mean of 0.6 years. This was within the range of delay in tooth formation found by Ranta,[2] who reported an average varying from 0.3 years to 0.7 years, and slightly lower than that found by Harris and Hullings[5], who noted that the mean delay for individual teeth (including the canines to the third molars) was 0.9 years. It is important to note that as-

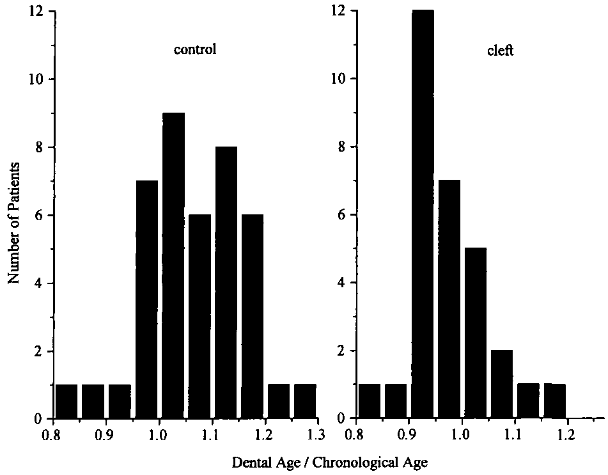


**Fig 1. Frequency distribution of the DMRs for boys. The distribution for control boys is centered at a DMR of 1.05 and the distribution for cleft boys is skewed to a DMR of 0.97.**

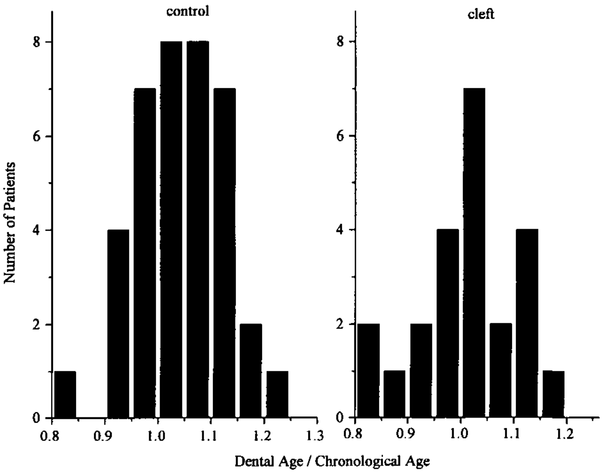


**Fig 2. Frequency distribution of the DMRs for girls. The distributions for control and cleft girls are both centered at a DMR of 1.05.**

sessment of dental age using the lower left quadrant of the mandible may not reflect the full effect of clefting in the maxilla; the dentition in the maxilla may conceivably demonstrate a larger dental age delay.

Using a different method of assessing the dental age (measuring the root length of developing teeth via panoramic radiographs), Brouwers and Kuijpers-Jagtman[7] reported that both maxillary and mandibular teeth were delayed in their formation and eruption in children with cleft lip and palate. Effective use of the Dermirjian method on maxillary teeth would require improved radiographic quality.

The prevalence of delayed dental age in our sample of cleft boys was 67% — lower than that reported by Harris and Hullings (89%, 48/54 subjects).[5] Detailed examination of children with delayed dental ages showed that of 20 affected boys, 10 had unilateral complete cleft lip and palate, six had bilateral cleft lip and palate, three had unilateral cleft lip, and one had bilateral cleft lip, alveolus, and palate. However, our study was not designed to examine the true prevalences of dental developmental changes in subgroups of cleft children.

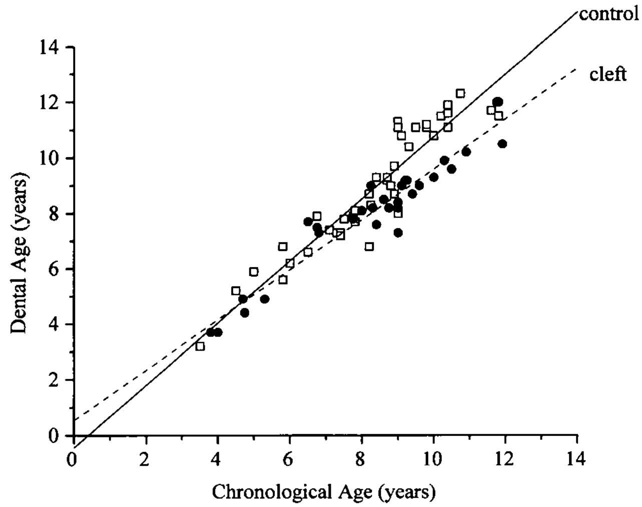


**Fig 3. Scattergraph of dental age and chronological age for boys. The open squares represent control subjects, dark circles represent cleft subjects. The continuous line is the linear regression for the control group (R value = 0.94), and the dashed line is linear regression for the cleft group (R value = 0.96).**

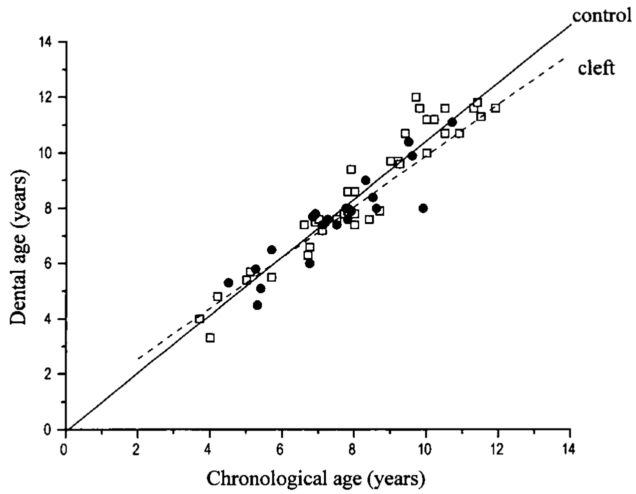


**Fig 4. Scattergraph of dental age and chronological age for girls. The open squares represent control subjects, dark circles represent cleft subjects. The continuous line is the linear regression for the control group (R value = 0.96), and the dashed line is linear regression for the cleft group (R value = 0.91).**

Further studies of dental development among cleft children are needed to delineate the possible relationship between severity of clefting and dental delay.

## Summary and conclusion

In this preliminary report on the developmental dental changes in isolated cleft lip and/or palate, the dental maturity of a group of cleft children was determined and compared with age- and sex-matched control children.

1. Boys with isolated CL(P) were dentally delayed compared with control boys.
2. The average dental age delay in boys with isolated CL(P) was 0.6 years.
3. There was no statistical difference in dental age between control and cleft girls.

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1. Bailit HL, Doykos JD III, Swanson LT: Dental development in children with cleft palates. J Dent Res 47:664, 1968.
2. Ranta R: A review of tooth formation in children with cleft lip/palate. Am J Orthod Dentofacial Orthop 90:11–18, 1986.
3. Kramer GJC, Hoeksma JB, Prahl-Andersen B: Emergence of the primary incisors in CLP children. Eur J Orthod 11:265–70, 1989.
4. Pöyry M, Ranta R: Formation of anterior maxillary teeth in 0- to 3-year-old children with cleft lip and palate and prenatal risk factors for delayed development. J Craniofac Genet Dev Biol 6:15–26, 1986.
5. Harris EF, Hullings JG: Delayed dental development in children with isolated cleft lip and palate. Arch Oral Biol 35:469–73, 1990.
6. Dermirjian A, Goldstein H, Tanner JM: A new system of dental age assessment. Hum Biol 45:211–27, 1973.
7. Prahl-Andersen B: The dental development in patients with cleft lip and palate. European Orthodontic Society Transcription 52nd Congress: 155–61, 1976.
8. Brouwers HJM, Kuijpers-Jagtman AM: Development of permanent tooth length in patients with unilateral cleft lip and palate. Am J Orthod Dentofacial Orthop 99:543–49, 1991.
9. Dermirjian A, Buschang PH, Tanguay R, Kingnorth Patterson D: Interrelationships among measures of somatic, skeletal, dental, and sexual maturity. Am J Orthod 88:433–38, 1985.

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