# THE PREVALENCE OF HYPODONTIA IN CHILDREN WITH CLEFT AND NONRELATED CONTROLS

**Claudia COREGA, Alin SERBANESCU, Mihai COREGA, Mihaela BACIUT**
University of Medicine and Pharmacy ”Iuliu Hatieganu” Cluj-Napoca, Romania

THE PREVALENCE OF HYPODONTIA IN CHILDREN WITH CLEFT AND NONRELATED CONTROLS (**Abstract**): The aim of this study was to compare the occurrence of hypodontia, dental age, and asymmetric dental development in children with cleft with a non-sibling control group. The study sample consisted of 30 children with cleft (aged 7.2 to 17.1 years) and 60 controls without cleft (aged between 7 and 18.8 years). Hypodontia, dental age, and asymmetric dental development were assessed on panoramic radiographs of the children with cleft and the control children without cleft. The cleft (p.001) group showed a significantly higher frequency of hypodontia and a significantly higher occurrence (cleft p.01) of asymmetric dental development, compared with the control group. Only a small, but insignificant delay in dental development could be found in the cleft group. The cleft subjects showed a significantly higher occurrence of hypodontia and asymmetric dental development than the non-cleft control group. This may suggest a genetic component for the occurrence of hypodontia and asymmetric dental development.

**Keywords**: cleft, hypodontia, tooth formation

## INTRODUCTION

Some dental traits such as hypodontia, supernumerary teeth, peg-shaped teeth, dental delay and dental asymmetry occur with higher frequency in individuals affected with cleft lip, cleft palate, or both (Ranta, 1986). The literature includes a large number of studies dealing with tooth formation in patients with cleft with a range of findings. Several studies report a delayed formation of the permanent teeth (Bailit et al., 1968; Ranta, 1972, 1982; Harris and Hullings, 1990; Brouwers and Kuijpers-Jagtman, 1991). Other studies report only a delayed dental development in boys until the age of 9 years (Prahl- Andersen, 1978; Prahl-Andersen et al., 1979). In the study of Loevy and Aduss (1988), early development in boys with clefts was observed. Left-to-right differences in tooth formation are also greater in children with cleft (Ranta 1973; Harris and Hullings, 1990). The incidence of hypodontia away from the cleft area in individuals is also markedly increased as compared with the population without cleft (Haataja et al., 1971; Ranta, 1986; Jiroutova and Mu¨ llerova, 1994). In particular, hypodontia most frequently involves the second premolars in the upper and lower jaw and the upper lateral incisor on the noncleft side (Ranta, 1986).

Some studies (Jordan et al., 1966; Schroeder and Green, 1975) report an increase in dental aberrations such as abnormal shape of teeth and supernumerary or missing teeth in siblings of children with cleft, compared with the general population.

However, these studies were only descriptive with little statistical analysis and in the meantime the dental age was not investigated. Investigations of Adams and Niswander (1967) and Bhatia (1972) support the idea that the same etiological factors that cause the formation of the cleft can affect the development of the dentition. Significant associations of

some patients with cleft lip and palate with tranforming growth factor alpha and retinoic acid receptor loci (Chevenix-Trench et al., 1992) were found.

Since there are few studies on children with a cleft, the aim of the present study was to compare hypodontia, dental delay, and asymmetric dental development in children affected with cleft lip or palate with a group of control children.

## MATERIALS AND METHODS

### Sample Selection:

The cleft group consisted of 30 children (20 girls and 10 boys), aged 7 years 2 months to 17 years 1 month (mean age 10 years 2 months). All were of Caucasian origin with nonsyndromic clefting. Twenty of these children had a complete cleft lip and palate, 6 children showed an isolated cleftpalate, and only 4 children had a cleft lip with cleft alveolar process. They were all enrolled for treatment at the Department of Orthodontics at the University of Medicine and Pharmacy ”Iuliu Hatieganu” Cluj-Napoca, Romania. The nonsibling control group consisted of 60 children (40 girls and 20 boys) whose age ranged from 7 years to 18 years 9 months (mean age 11 years 3 months). At the time of the orthopantomogram, none had been treated orthodontically. The children of the noncleft sibling and control groups were of Caucasian origin and were nonsyndromic.

### Method:

An orthopantomogram was taken of each child to assess the frequency of hypodontia and the dental maturation (dental age). The sample for evaluating the frequency of hypodontia consisted of 30 children with cleft.

Dental age was calculated using the method of Demirjian and Goldstein (1976). A computer system and individual data sheets were used to train the evaluators in scoring the stages of development correctly and consistently. Individual radiologic appearances of the seven permanent teeth on the left side of the mandible were evaluated according to developmental criteria. Development of each tooth was categorized into one of eight stages. These individual scores were entered into a clinical evaluation program, which converted them, depending on the sex of the child, into a maturation and dental age score. Panoramic X-rays, which showed a full maturation score, or bilateral agenesis or extraction of at least one tooth in the lower jaw were excluded. Thus the final sample for evaluating the dental development consisted of 30 children affected with namely 20 with cleft lip and palate, 6 with a cleft palate, and 4 with a cleft lip and alveolus. In order to assess the reliability of this method, the scores of 30 children were measured twice with an interval of 1 month by two examiners as a pilot study.

To investigate the symmetry of permanent tooth formation, individual tooth developmental stages of seven left and right mandibular teeth were compared. A pair of teeth was regarded as having undergone asymmetrical development when the tooth development stage of the left tooth deviated from that of the antimeric tooth by at least one developmental stage.

The panoramic X-rays were also studied for congenitally missing teeth outside the cleft region (excluding the lateral incisor in the upper jaw on the cleft side). A tooth germ was considered to be congenitally missing if it was absent on the X-ray, although the child’s age would have supported its being radiographically detectable (Haavikko, 1970). The presence of the preceding deciduous tooth was in most cases a supporting criterion for the diagnosis of

hypodontia. When the deciduous tooth was missing, the patient's file was reviewed and the patient was interviewed in order to exclude the possibility of an extraction.

All data were transferred to Microsoft Excel 97 (Microsoft Corporation, Redmond, Washington) for statistical analysis.

For each patient, missing teeth, the difference between dental and chronological age, the dental delay compared with the controls as well as the asymmetry of dental development were assessed.

For each group (cleft group and control group), the means and the standard deviations of dental age, chronological age, differences between dental and chronological age and dental delay of the cleft compared with the controls were calculated. Differences between the groups were analyzed using the unpaired *t* test and the *F* test for equality of variances. The chi-square test was used in order to test differences (frequency) in hypodontia and dental asymmetry among the two groups.

Probabilities less than .05 were considered to be statistically significant.

## RESULTS

### Error of Method:

No statistically significant differences were found between the means of the intra- and interobserver set of measurements. The intraobserver measurements yielded a correlation of 0.988, which was almost equal to the correlation of the interobserver measurements: 0.994. The measurement error for the dental age was at most one developmental stage.

### Hypodontia:

In the group of 30 children with cleft, 15 children (50%) showed hypodontia of one or more teeth outside the cleft region.A total of 17 teeth were absent (upper/lower jaw 10/7). In the control group of 60 children, 6 children (10%) showed hypodontia of one or more teeth. A total of 9 teeth were absent (upper/lower jaw 6/3).Compared with the nonsibling controls, the cleft group showed a highly significant increase in frequency of hypodontia (*p* .001).

Hypodontia involved mostly the second premolars of the upper and lower jaw and the upper lateral incisor on the contralateral side to the cleft. The most frequently missing teeth in all the groups were the second premolars. No significant difference in hypodontia between the upper and lower jaw or any significant sex differences were found.

### Comparison of the Dental and Chronological Age:

The cleft group had a mean dental age of 10.2 years, which was 0.25 years (3 months) greater than the mean chronological age of 9.11 years of this group. The control group showed a mean dental age of 11.3 years, which was 0.3 years (4 months) older than the mean chronological age of 10.11 years.

### Asymmetric Tooth Formation:

In the group of 30 cleft children, 25 (50%) were found to have one or more asymmetrically developing pair of teeth and in the control group, 17 of 60 children (28.33%) showed asymmetric tooth development. The cleft group showed significantly more asymmetrical dental formation, compared with the control group (chi-square: cleft-control *p* .01). In each group, the premolars most frequently exhibited asymmetric development.

## DISCUSSION

The aim of this study was to compare dental development among a cleft and a control

group. Sample size precluded comparison of scores for different cleft types, which would also influence results.

In the cleft group, some of the children had been treated orthodontically. According to Fanning (1962), orthodontic treatment can influence the eruption but not the root formation of the teeth. Teeth close to the cleft are likely to have various malformations because of some additional environmental factors (Ranta, 1986). Since this study was interested in the genetic issues in hypodontia of children with cleft, we excluded hypodontia in the cleft area.

The most frequently missing teeth on the noncleft side were the premolars and the maxillary lateral incisor. This is in agreement with Ranta (1986). Our findings show a certain gradation in frequency of hypodontia among the two groups: the cleft group shows the highest frequency of hypodontia outside the cleft region (34.5%), followed by the control group (22.6%). This frequency of hypodontia outside the cleft region is in accordance with previous studies (Weise and Erdmann: 1967; 28% in unilateral cleft lip and palate, 17.9% in bilateral cleft lip and palate; Ranta 1983: 31.5% in isolated cleft palate). Concerning the dental development, we preferred to use the method of Demirjian and Goldstein (1976), which uses the teeth of the lower jaw so that local (environmental) factors such as surgical trauma are excluded. We found no significant differences in mean (dental-chronological) age among the two groups. Compared with the controls, the cleft groups show an insignificant mean relative dental delay. Ranta (1986) estimated the delay in tooth formation to var y from 0.3 years to 0.7 years according to the severity of the cleft and the hypodontia. Tooth formation was delayed longer in the more severe cleft cases and in the subgroups with severe hypodontia. This is in agreement with the mean dental delay of 0.2 years reported in this study. With the method of Demirjian and Goldstein, however, we were not able to assess dental age in cases of multiple missing teeth, which were often severe cleft cases.

Concerning the dental age assessment, a consistent overestimation of 3.5 months was found in all groups using the method of Demirjian and Goldstein. This confirms the results of other studies, which found an overestimation from 6 to 10 months with Demirjian and Goldstein's method (Ha¨ gg and Matson, 1985; Staaf et al., 1991). Given this consistent overestimation in all groups (greater overestimation in the control group than in the cleft group), one could wonder whether the cleft group are really as different as results indicate.

No gender difference could be discovered with Demirjian and Goldstein's method because the conversion of the maturity score into a dental age is dependent on the sex.

Significant differences were found in the frequency of asymmetric dental development between the cleft group and the control group. This agrees with the results of several other studies that found a significantly higher frequency of asymmetric dental development in children with cleft (Ranta, 1973, 1986). We should be careful with these results, given the reliability of the method (useable within one development stage).

## CONCLUSIONS

The cleft group showed findings which were significantly different from the control individuals. The children with cleft demonstrated a significantly higher frequency of hypodontia and a significantly higher frequency of dental asymmetries together with a small but nonsignificant mean dental delay relative to controls without cleft. The results of this study suggest that some genetic factors for clefting and tooth development have some relationship.

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