Prevalence of second premolar hypodontia in the Polish cleft lip and palate population

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Background:
Cleft lip and/or palate is the most frequent congenital abnormality occurring in the craniofacial region and is often associated with numerous dental defects such as tooth agenesis, supernumerary teeth, microodontia, taurodontism, crown malformations, or delay in eruption. The prevalence of hypodontia in cleft-affected patients is much higher in comparison with a healthy population. The aim of this study was to evaluate the prevalence of second premolar hypodontia in patients with cleft lip and/or palate.

Material/Methods:
We performed a retrospective evaluation of panoramic radiographs and dental casts in the Department of Dentofacial Orthopedics and Orthodontics, Wroclaw Medical University. Two independent observers evaluated the records of 469 patients with various types of clefts and analyzed dental casts and panoramic radiographs.

Results:
202 individuals met inclusion criteria. The sample comprised 120 UCLP patients, 38 BCLP patients, 28 CP patients, and 17 CLA patients. Hypodontia in the premolar region was observed in 39 individuals (19.3%). A total number of 58 second premolars were missing, of which 35 were maxillary second premolars (U5) and 23 were mandibular second premolars (L5).

Conclusions:
Estimated hypodontia in the Polish CL/P sample was considerably higher than the hypodontia in permanent dentition reported for a European healthy population. The number of congenitally missing second premolars was higher in the maxillary arch than in the mandibular.

MeSH Keywords:
Anodontia • etiology • Cleft Lip • diagnosis • Cleft Palate • diagnosis

Full-text PDF: http://www.medscimonit.com/download/index/idArt/890386
Background

Cleft lip and/or palate is the most frequent congenital abnormality occurring in the craniofacial region. According to the Polish authorities, the prevalence rate for cleft anomalies in Poland is 1.43 cases per 1000 births [1]. Orofacial clefts are often associated with numerous dental defects such as tooth agenesis, supernumerary teeth, microdontia, taurodontism, crown malformations, and delays in eruption [2–11]. Numerous studies have reported hypodontia as the most prevalent dental alteration in cleft patients, with the maxillary lateral incisor being the most common missing tooth, followed by maxillary second premolar and mandibular second premolar [7,8,12–14]. The frequency of tooth agenesis increases with the severity of the cleft and has been found to be the highest in cleft lip and palate patients [15–19]. The prevalence of hypodontia in cleft-affected patients is much higher in comparison with a healthy population, ranging from 27% to 77% [7,17,20,21]. The congenital absence of maxillary lateral incisors, often associated with the proximity between the cleft site and the formation site of the permanent lateral incisor, was found in 39% to 60% of cleft individuals [9,12,22,23]. Studies concerning tooth agenesis indicated that outside the cleft region the prevalence of hypodontia is also increased and the second premolars are affected most often [7,24,25]. Shapira et al. [16] observed missing second premolars in 18% of cleft lip and/or palate patients.

In the general population the prevalence of congenitally missing teeth (except third molars) is much lower and varies from 3.2% to 5.5% for men and from 4.6 to 7.6% for women. The prevalence of dental agenesis in females is 1.37 higher than in males. Agenesis of second premolars alone is even more uncommon and has been estimated at about 1.4% to 1.6% and 2.9% to 3.2% for maxillary and mandibular second premolars, respectively [26,27].

The aim of the present study was to evaluate hypodontia of second premolars in a population of cleft Polish children from south-western Poland.

Material and Methods

The material for the present study was obtained from the records of cleft patients treated in the Department of Dentofacial Orthopedics and Orthodontics, Wroclaw Medical University during the years 1990–2008. The inclusion criteria for the study were: cleft patients, lower Silesia region residency, and Caucasian race. The authors analyzed a total of 469 files of cleft patients. From this material, we excluded 267 individuals who were aged below 6 years; had fuzzy panoramic x-rays; syndromic clefts; unique, atypical types of clefts; or were of non-Polish ethnic origin; plus a small group of 3 CL patients. Thus, 202 subjects with cleft lip and/or palate participated in this study. Detailed information about exclusion process for the study is shown in Table 1. The counts and distribution of missing teeth in the second premolar region in mandibular and maxillary arches were investigated through the use of the patient’s dental casts and at least 1 clear panoramic x-ray. The patient’s records were examined for possible extractions of second premolars. The evaluation was performed separately by 2 operators. In the case of a divergence in outcomes, the operators discussed the case until they reached an agreement. Overall inter-observer agreement was calculated by comparing the findings of the 2 operators. Tooth counts and percentages were used to characterize tooth agenesis. Chi-square and Fisher’s exact test were used to determine significant differences. The statistical significance level was set at 0.05. All analyses were performed using SPSS software 14.0.

Results

The CL/P samples were divided into 5 groups based on the cleft type. Out of 202 subjects, 17 were CLA patients, 120 were UCLP patients, 37 were BCLP patients, and 28 were CP patients. A significant male predominance was observed in the sample (p=0.005). The total number of patients as distributed by cleft type and sex is shown in Table 2.

Table 1. Exclusion criteria and the number of patients included in the study.

| Exclusion criteria                        | Number of patients included |
|-------------------------------------------|-----------------------------|
| Coexisting syndrome                      | 68                          |
| Unclear panoramic x-ray                   | 87                          |
| Age below 6                               | 91                          |
| Unique, atypical type of cleft            | 15                          |
| Ethnic origin                             | 3                           |
| Small sample size (CL group)              | 3                           |
| Total number of patients included         | 202                         |

Hypodontia in the premolar region in the CL/P sample was observed in 39 individuals (19.3%): 20 males (16.5%) and 19 females (23.5%) (Table 3). The difference in frequency of tooth agenesis between sexes was not statistically significant (p=0.275). Similarly, no significant association was observed between cleft type and hypodontia (p=0.110). A total number of 58 second premolars were missing, of which 35 were maxillary second premolars (US) and 23 were mandibular second premolars (LS). The percentage of missing second premolars
out of all possible missing second premolars was found to be 7.2% in the upper and lower arch combined; along with 8.7% and 5.7%, respectively, in the maxillary and mandibular arch. Females (7.7%) showed a higher percentage of missing premolars than males (6.8%), but the difference was not statistically significant (p=.302) (Table 4). The difference in frequency for missing premolars between cleft types was also not significant (p=.143). The overall agreement in diagnosis between investigators was found to be 96%.

Cleft lip and alveolus (CLA)

The number of patients with cleft lip and alveolus was 17 (9 males and 8 females) of whom 7 males and 5 females had left

Table 2. Distribution of cleft types in the analyzed sample.

| Cleft Type | CLA | UCLP | BCLP | CP | Total |
|------------|-----|------|------|----|-------|
| Males      | 9   | 78   | 25   | 9  | 121   |
| Females    | 8   | 42   | 12   | 19 | 81    |
| Total      | 17  | 120  | 37   | 28 | 202   |

Table 3. Distribution of second premolar hypodontia.

| Cleft Type | Number of patients | Number and percentage of patients with missing second premolars |
|------------|--------------------|---------------------------------------------------------------|
|             |                    | Maxilla | Mandible | Males | Females | Total |
| CLA        | 17                 | 0 (0.0%) | 2 (11.8%) | 1 (11.1%) | 1 (12.5%) | 2 (11.8%) |
| UCLP       | 120                | 13 (10.8%) | 10 (8.3%) | 13 (16.7%) | 6 (14.3%) | 19 (15.8%) |
| BCLP       | 37                 | 6 (16.2%) | 2 (5.4%) | 3 (12.0%) | 5 (41.7%) | 8 (21.6%) |
| CP         | 28                 | 7 (25.0%) | 3 (10.7%) | 3 (11.4%) | 7 (26.8%) | 10 (35.7%) |
| Total      | 202                | 26 (13.9%) | 17 (8.4%) | 20 (16.5%) | 19 (23.5%) | 39 (19.3%) |

Table 4. Distribution of number and percentage of congenitally missing second premolars by jaws, gender and cleft type in the sample.

| Cleft Type | Number of patients | Number and percentage of missing second premolars |
|------------|--------------------|--------------------------------------------------|
|             |                    | Maxilla | Mandible | Males | Females | Total |
| CLA        | 17                 | 0 (0.0%) | 2 (5.9%) | 1 (2.8%) | 1 (3.1%) | 2 (2.9%) |
| UCLP       | 120                | 18 (7.5%) | 15 (6.2%) | 24 (7.7%) | 9 (5.4%) | 33 (6.9%) |
| BCLP       | 37                 | 8 (10.8%) | 2 (2.7%) | 3 (3.0%) | 7 (14.6%) | 10 (6.8%) |
| CP         | 28                 | 9 (16.1%) | 4 (7.1%) | 5 (13.9%) | 8 (10.5%) | 13 (11.6%) |
| Total      | 202                | 35 (8.7%) | 23 (5.7%) | 33 (6.8%) | 25 (7.7%) | 58 (7.2%) |

Table 5. Distribution of hypodontia and missing second premolars in CLA group.

| Sex     | Number of patients | Number of patients with hypodontia | Number of missing premolars |
|---------|--------------------|-----------------------------------|-----------------------------|
|         | Left  | Right | Bilateral | Total | Left  | Right | Bilateral | Left  | Right | Bilateral | Total |
| Male    | 7     | 1     | 1         | 9     | 0     | 0     | 0         | 1     | 0     | 0         | 1     |
| Female  | 5     | 2     | 1         | 8     | 0     | 0     | 0         | 1     | 0     | 0         | 1     |
| Total   | 12    | 3     | 2         | 17    | 0     | 0     | 0         | 2     | 0     | 0         | 2     |
Table 6. Distribution of hypodontia and missing second premolars in UCLP group.

| Sex    | Number of patients | Number of patients with hypodontia | Number of missing premolars |
|--------|--------------------|-----------------------------------|----------------------------|
|        | Left   | Right  | Total  | Left | Right | Total  | Left | Right | Total  |
| Male   | 52     | 26     | 78     | 8    | 5     | 13     | 16   | 8     | 24     |
| Female | 30     | 12     | 42     | 4    | 2     | 6      | 6    | 3     | 9      |
| Total  | 82     | 38     | 120    | 11   | 6     | 19     | 22   | 11    | 33     |

CLA, 1 male and 2 females had right CLA, and 1 male and 1 female had bilateral CLA. Only 2 patients (11.7%) with bilateral CLA (1 boy and 1 girl) had hypodontia of their premolars. The number of missing teeth was 2 (2.9% out of the possible missing second premolars), both in the mandibular arch (5.9%) (Table 5).

**Unilateral cleft lip and palate (UCLP)**

The UCLP sample comprised 120 individuals (78 males and 42 females), of whom 82 had left UCLP and 38 had right UCLP. Nineteen subjects (15.8%) had congenitally absent second premolars: 13 males (16.7%) and 6 females (14.3%). The total number of missing teeth was 33 (6.9%); 24 in the male group (7.7%) and 9 in the female group (5.4%). Eighteen congenitally absent second premolars were found in the upper arch (8.8%) and 15 in the lower arch (6.2%) (Table 6). Thirteen individuals had missing maxillary second premolars: 7 were located ipsilateral to the cleft side, 1 was located contralateral, and 5 had agenesis of both second premolars. Out of 10 subjects with hypodontia in the lower premolar region, 5 had missing ipsilateral premolars and 5 had both teeth missing.

**Bilateral cleft lip and palate (BCLP)**

The number of patients with bilateral cleft lip and palate was 37 (25 males and 12 females). Hypodontia in the second premolar region was found in 8 subjects (21.6%): 3 males (12%) and 5 females (41.7%). Altogether, 10 premolars were missing (6.8%); of which 3 were observed in the male group (3%) and 7 in the female group (14.6%). Eight congenitally absent teeth were found in the maxilla (10.8%) and 2 in the mandible (2.7%).

**Cleft palate (CP)**

The CP group included 28 individuals (9 males and 19 females). At least 1 second premolar was missing in 10 subjects (35.7%): 3 males (33.3%) and 7 females (26.8%). A total number of 9 maxillary (16.1%) and 4 mandibular (7.1%) congenitally absent teeth were found, of which 5 were in the male group (13.9%) and 8 in the female group (10.5%).

**Discussion**

In this study the congenital absence of at least 1 second premolar was observed in 19.3% of cleft individuals. This result is very similar to the outcomes, ranging from 17.3% to 19.2%, previously reported by others for CL/P samples [7,20] and considerably higher than the 5.5% calculated for hypodontia in the permanent dentition for the European population [26]. On the other hand, the results of a study performed on dental casts for cleft patients from Indonesia revealed a lack of missing second premolars in the CLA and CLP groups. This discrepancy could be attributed to cleft palate operations, different methodologies in surveys, and/or the ethnic compositions of the samples [28]. Although not statistically significant, in this study, as in the general population and other surveys concerning hypodontia in cleft patients, females (23.5%) were more affected than males (16.5%) [7,15,20,26]. The prevalence of missing second premolars (7.2%) in this sample is comparable to the outcomes, ranging from 6.3% to 9.6%, observed in other surveys [7,17,20]. The percentages of missing upper (8.7%) and lower second premolars (5.7%) in cleft-affected patients are considerably higher than the 1.4% to 1.6% for US and 2.9% to 3.2% for LS reported in the healthy population [26].

In the group of patients with cleft lip and alveolus prevalence of second premolars, hypodontia was calculated at 11.8% and the percentage of missing premolars at 2.9%, which is considerably less than the 18.2% to 24% and 6% to 7%, respectively, reached in different studies [17,20]. These differences may be explained by the small sample size and the different composition of the groups studied.

The frequency of patients with missing second premolars and the percentage of missing premolars in the largest UCLP group were 15.8% and 6.9%, respectively. Very similar outcomes have been published previously [7]. Hypodontia in the maxillary arch was observed in 10.3% of cleft individuals, which is in agreement with other studies [7,9,14]. Also, the results obtained in the mandibular arch (8.3%) are consistent with the findings reported by Wong et al. [14]. However, Menezes and Vieira [7] reported the frequency of hypodontia in the lower second premolar
region to be 1.9%, which is even lower than the prevalence of second premolar agenesis in the healthy population [26].

The highest incidence of hypodontia in the second premolar region (21.6%) was observed in the BCLP group. This outcome is consistent with the data, ranging from 21.3% to 29.6%, reported in other surveys [7,29]. The frequency of premolar hypodontia observed in our sample in the maxillary arch (16.2%) is considerably higher than the values (8.8%) revealed by Camporesi et al. [9], but lower than the values ranging from 21.3% to 27.8% obtained in other studies [7,10,29]. In the mandibular arch, our findings (5.4%) were similar to those obtained by Menezes and Vieira [7] and Tereza et al. (2010) [10], but substantially smaller than others [14,29,30]. The different composition or small size of our sample may be responsible for all of these inconsistencies. The percentage of missing second premolars was found to be 6.8%, which is consistent with the studies of Menezes and Vieira [7] and Halpern and Noble [31]. Also, as in other surveys, the percentage of congenitally missing US (10.8%) was substantially higher than LS (2.7%), and the percentage of lower second premolars was similar to the noncleft population [7,31].

In the CP group, the prevalence of hypodontia of second premolars was 35.7%. This outcome is similar to the values (35.3%) obtained by Menezes and Vieira [7] but much lower than the values (10%) reported by Shapira et al. [15] Surprisingly, the prevalence of hypodontia in the maxillary arch (25%) was notably higher than in mandibular premolars (10.7%). This is an unexpected finding, because other studies reported either opposite results or no difference in the frequency between upper and lower premolar [7,14,16]. The number of missing premolars in the CP group was 13, which was 11.6% out of the total possible second premolars. This finding compares favorably with other previously published studies [16,17,32,33]. Only Menezes and Vieira [7] reported a higher (20.6%) incidence.

Other authors observed a gradual increase in the frequency of hypodontia in cleft-affected groups, along with the severity of the cleft [16–18]. This finding is consistent with our results, with the CLA group (11.7%) being the least affected, and the BCLP group (21.6%) the most (Table 3).

**Conclusions**

Of the basis of the results obtained in this survey, we conclude that:

- The prevalence of second premolar hypodontia in the Polish CL/P sample was considerably higher than the hypodontia in permanent dentition reported for European healthy populations.
- The number of congenitally missing second premolars was higher in the maxillary than in the mandibular arch.

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