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Hypodontia in Children with Various Types of Clefts

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ABSTRACT

The prevalence of hypodontia in children with clefts, both inside and outside the cleft region, and the possible association between the side of the cleft and the side of the missing teeth were studied using radiographs of 278 patients with cleft lip, cleft palate, or both (158 boys and 120 girls), age 5 to 18 years (mean age 10.4 years). A hypodontia prevalence of 77% (excluding third molars) was found for the total cleft sample. This was significantly higher, both statistically and clinically, than the incidence of hypodontia reported for noncleft populations, and considerably higher than the prevalence reported in other studies of children with clefts. The maxillary permanent lateral incisors were the teeth most frequently missing on the cleft side (259 teeth) followed by the maxillary (47 teeth) and mandibular (23 teeth) second premolars, in both boys and girls. The teeth that were most often missing on the noncleft side were the maxillary second premolars (12 teeth), followed by the maxillary lateral incisors (10 teeth) and mandibular second premolars (6 teeth). Hypodontia of both the maxillary lateral incisors and second premolars was found more frequently on the left side, which also has a higher frequency of clefting.

KEY WORDS: Hypodontia, Missing teeth, Cleft lip, Cleft palate, Lateral incisors, Second premolars.

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A variation in the worldwide incidence of hypodontia and a variation in the tooth most frequently involved has been reported in previous studies.^{1,2} Studies of large segments of populations from different locations indicate great variability in the incidence of hypodontia. The incidence of missing permanent teeth, excluding third molars, was 3.4% in Swiss children,³ 4.4% in American children,⁴ 4.6% in Israeli children,⁵ 6.1% in Swedish children,¹ 8% in Finnish children,⁶ and 9.6% in Austrian children.⁷ Furthermore, it is anecdotally reported that, in certain relatively isolated groups of people where familial inbreeding is common, the prevalence of congenitally missing teeth reaches a double-digit figure (personal observation, M. Kuffinec, 1969.).

Some authors reported the permanent maxillary lateral incisor as the tooth most commonly missing in the general population.^{2,8} Others described the mandibular second premolar to be the most frequently missing tooth, with a frequency of 3.4%,^{9,10} followed by the maxillary lateral incisors, at a rate of 2.2%.⁹ Congenital absence of teeth was found more often unilaterally than bilaterally for all the affected teeth¹¹ except for the second premolar, where bilateral absence was found about 1.5 times more often.¹ It is generally accepted that agenesis of teeth is related to an overall reduction in tooth size. Consequently, hypodontia and microdontia tend to occur in the same children.¹²⁻¹⁴ Hypodontia is, to a great degree, genetically determined and transmitted by autosomal dominant inheritance, with incomplete penetrance and variable expression.^{1,15} Environmental factors, however, may also play a role in the etiology of this condition.^{16,17} A mutation in the homeobox gene, MSX1, has been suggested as a factor causing a common developmental dental anomaly, namely familial selective agenesis of the second premolars and third molars.¹⁸

Various dental abnormalities, particularly hypodontia, have a much higher prevalence in certain groups. These dental anomalies have been frequently reported in children who also have cleft lip, cleft palate, or both.¹⁹⁻²¹ Interestingly, these anomalies were found in proportionately higher frequencies as the severity of the cleft increased.^{19,20} The 8% incidence of hypodontia, excluding third molars, reported in normal Finnish children was found to increase up to 31.5% in those affected with an isolated cleft palate.⁶ A prevalence of tooth agenesis of 45.5% in the cleft area was reported by Bohn.¹⁹ Shapira found an incidence of 74% for missing maxillary lateral incisors and 18% for missing second premolars in children with cleft lip, cleft palate, or both.²¹ Hypodontia outside the cleft region was much higher in cleft-affected children than in others.¹⁹⁻²¹ The frequency of missing teeth outside the cleft site for children affected with cleft lip and palate, in descending order of magnitude was 7.5% to 32.3% for the maxillary second premolars, 3.1% to 10.4% for the maxillary lateral incisors, and 0.4 to 10.8% for the mandibular second premolars.²² Hypodontia was found more frequently in the clefted maxilla than in the mandible, and in unilateral clefts this hypodontia was more often on the same side as the cleft, most commonly the left side.¹⁹⁻²¹

In children without clefts, bilaterally missing maxillary lateral incisors and maxillary and mandibular first and second premolars were more common than unilaterally absent teeth.

In children with isolated cleft palate, bilateral absence of mandibular second premolars, maxillary and mandibular canines, and mandibular central incisors was more common than unilateral absence, but with no statistically significant differences between the groups. Rates as high as 59% missing maxillary lateral incisors and 54% missing maxillary second premolars have been reported in these children.⁶ The prevalence of hypodontia in the isolated cleft palate group was reported to be 4 times that of the noncleft group, and higher in the maxilla (sixfold) than in the mandible (threefold).⁶ The increased incidence of hypodontia in children with clefts might be a result not only of the genetic factors directly affecting hypodontia, but also of the factors causing the cleft itself.^{23,24} This suggests that the same etiologic factors may be responsible for both the formation of the clefts and the hypodontia in affected children.²⁵ The congenital absence of the maxillary lateral incisors is associated with other dental changes, both in the size and number of teeth.²⁶ When not absent, the maxillary lateral incisor on the cleft site is nearly always abnormal in size and shape, and the second premolars show delay in development and eruption.^{27,28} Deviation from the normal size and shape of the lateral incisor (small or peg-shaped) on the contralateral side, and the delayed eruption of the second premolars observed in the majority of reports, were suggested as representing a mild expression of hypodontia.^{19,20,29,30,31} In all cleft groups, hypodontia was more frequent in girls than in boys, but the differences were not statistically



The purpose of the present investigation was to determine the prevalence of hypodontia of permanent teeth inside and outside the cleft region of children with various types of clefts, and to compare these findings with those reported for the noncleft population. In addition, the association between the location of the cleft and the side of the missing teeth was examined.

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The material for the present investigation was collected from 4 cleft palate centers in metropolitan New York City (Suffolk Cleft Palate Rehabilitation Center [n = 105]; New York Medical Center, Institute for Reconstructive Plastic Surgery [n = 75]; Montefiore Medical Center, Center for Craniofacial Disorders [n = 56]; and New York University College of Dentistry, Department of Orthodontics [n = 42]), and included records of 278 patients with cleft lip, cleft palate, or both, 158 boys and 120 girls, 5 to 18 years old (mean age 10.4 years). A thorough examination for hypodontia of permanent teeth (excluding third molars) was undertaken using panoramic, periapical, and occlusal radiographs. Children whose cleft was part of a craniofacial syndrome and children whose radiographs were not of diagnostic clarity were excluded from the sample. In addition, children under the age of 5 years and those who had teeth extracted due to dental decay or orthodontic treatment were excluded.

The sample consisted of cleft patients who were enrolled in 1 of 4 centers for treatment or follow-up and consisted of children of varied racial and ethnic origins and of mixed socioeconomic backgrounds. Statistical analysis using the chi-square test was performed to determine the significance of the findings.


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Based on the type of cleft, the sample was divided into 4 groups and hypodontia of permanent teeth was studied separately in each group. The patient's sex and the affected side were also recorded. In the total sample of 278 patients with cleft lip, cleft palate, or both, hypodontia was found in 213 (77%), including 127 (46%) boys and 86 (31%) girls. No hypodontia was found in either jaw in 65 (23%) of children with clefts, (31 [11%] boys and 34 [12%] girls; [Table 1](#) ). In total, 339 teeth were absent in the entire cleft sample, including 312 (92%) teeth missing on the cleft side and 27 (8%) on the noncleft side. Of these, 269 (79%) were maxillary permanent lateral incisors (259 on the cleft side and 10 on the noncleft side). A statistically significant difference ($P = .000001$) was found between missing lateral incisors on the cleft side compared with the noncleft side. Seventy missing teeth (21%) were second premolars, of which 47 (67%) were missing from the maxilla and 23 (33%) from the mandible. Of the 70 missing second premolars, 53 were missing on the cleft side (35 in the maxilla, 18 in the mandible), and 17 on the noncleft side (12 in the maxilla and 5 in the mandible; [Table 2](#) ). The cleft side presents considerably more missing second premolars than the noncleft side ($P = .037$).


Cleft lip


The cleft lip group comprised 6 children, 3 boys and 3 girls. Maxillary permanent lateral incisors were absent on the cleft side in 2 children, 1 boy and 1 girl, both on the left side. In 1 boy, the missing lateral incisor was on the noncleft side; thus, 3 teeth were absent in this group. No other teeth were missing ([Table 3](#) ).

Cleft palate


The cleft palate group comprised 30 children, 7 boys and 23 girls. Hypodontia was detected in 9 children, 2 boys and 7 girls, with a total of 15 teeth absent. Six children, 1 boy and 5 girls, had 11 missing maxillary lateral incisors, and 3, 1 boy and 2 girls, had a total of 4 missing second premolars, all in the mandibular arch ([Table 4](#) ).


Cleft lip and alveolus

A total of 44 children, 20 boys and 24 girls, had cleft lip and alveolus. Hypodontia was found in 38 children, 21 (55%) boys and 17 (45%) girls, with 46 missing teeth, of which 35 (76%) were maxillary lateral incisors and 11 (24%) were second premolars. Twenty-four teeth, of which 15 (63%) were maxillary lateral incisors and 9 (37%) were second premolars, were missing in boys. Twenty-two teeth, of which 20 (91%) were lateral incisors and 2 (9%) were second premolars, were absent in the girls ([Table 5](#) ).

Hypodontia on the left side included 22 (48%) teeth (18 lateral incisors and 4 second premolars). On the right side, 8 (17%) teeth (7 lateral incisors and 1 second premolar) were missing, while 16 (35%) teeth were absent bilaterally (10 lateral incisors and 6 second premolars). Hypodontia was found on the left side approximately 3 times more frequently than on the right side (22 and 8, respectively). Lateral incisors were missing 3 times more often than second premolars (35 and 11, respectively; [Table 6](#) ).

Cleft lip and palate





The cleft lip and palate group was the largest group in our sample and included 198 children, 128 boys and 70 girls. Hypodontia of lateral incisors was observed in the maxilla, and hypodontia of second premolars was observed in both the maxilla and the mandible. Altogether, 275 teeth were absent, of which 220 (80%) were lateral incisors and 55 (20%) were second premolars. Boys were missing 174 teeth, including 139 (80%) lateral incisors and 35 (20%) second premolars. Girls were missing 101 teeth, including 81 (80%) lateral incisors and 20 (20%) second premolars ([Table 7](#) ).


The left side was affected with 96 (35%) missing teeth (78 lateral incisors and 18 second premolars), and the right side with 61 (22%) missing teeth (54 lateral incisors and 7 second premolars). One hundred eighteen (43%) teeth were absent bilaterally (88 lateral incisors and 30 second premolars; [Table 8](#) ).



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

The present study revealed a prevalence of 77% hypodontia in the children with cleft lip, cleft palate, or both examined, excluding third molars. This frequency was statistically higher than the 1.6% to 9.6% reported for a noncleft population,⁷ and considerably higher than the 45.5% previously reported for children with cleft lip and palate.¹⁹ Hypodontia of the maxillary permanent lateral incisors, observed in 74% in our total cleft sample, was significantly higher than the 2.2% reported for the normal population,⁹ or the 56.9% previously reported for children with cleft lip and palate.³²

Similarly, a finding of 18% missing second premolars found in children with cleft lip and palate²¹ was considerably higher than the 3.4%⁹ to 6.6%³³ found in children without clefts. This was only slightly lower than the 24% missing second premolars reported for children with cleft lip and palate.³⁰ The sample population in the reported studies were more homogenous than ours, which was of varied racial and ethnic origin. This could possibly explain these differences.

The considerably higher prevalence of hypodontia of both maxillary lateral incisors and maxillary and mandibular second premolars in the entire cleft sample; a total of 339 teeth missing from the maxillary and mandibular arches, is reported ([Table 2](#) ). The great majority of missing teeth was from the maxilla. There were 269 missing lateral incisors, of which 259 were on the cleft side and 10 on the noncleft side. The difference was statistically significant ($P = .000001$). In addition, 47 second premolars were missing in the maxillary arch, of which 35 were on the cleft side and 12 on the noncleft side. In addition, 23 second premolars were absent in the mandible, of which 18 were on the cleft side and 5 on the noncleft side ([Table 2](#) ). It is evident that the cleft side, which is found in a substantially higher incidence on the left side, presents considerably more missing second premolars than the noncleft side ($P = .037$). This is true both in the maxilla and the mandible (53 and 17, respectively; [Table 2](#) ). In fact, hypodontia was found an order of magnitude more frequently on the cleft side than on the noncleft side (312 and 27, respectively). Similarly, substantially more missing teeth were detected in the maxillary cleft side (259 lateral incisors and 35 second premolars), compared with its antimere noncleft side (10 lateral incisors and 12 second premolars, [Table 2](#) ).

Missing second premolars were found 3 times more often on the left side than the noncleft side, both in the maxilla (35 to 12) and in mandible (18 to 5). They were absent approximately twice as often on the maxillary cleft side than the mandibular cleft side (35 and 18, respectively). Similarly, second premolars were missing twice as often on the maxillary noncleft side than the mandibular noncleft side (12 and 5, respectively, [Table 2](#) )

The distribution of hypodontia by cleft type, tooth, and jaw side is presented in [Table 9](#) . Left side predominance for hypodontia is evident for all cleft types. Maxillary lateral incisors were missing over 50% more often on the left side than on the right (100 and 61, respectively). Maxillary second premolars were absent more than twice as often on the left side than on the right (14 and 6, respectively). Similarly, mandibular second premolars were absent approximately 3 times more often on the left side than on the right (8 and 3, respectively; [Table 9](#) )

Hypodontia distribution by cleft type indicates a significantly high incidence of missing lateral incisors ([Figure 1](#) ). They were absent on the cleft side of our isolated cleft lip group ([Table 3](#) ) and in the group of isolated cleft of the secondary palate ([Table 4](#) ). Neither of these involves the alveolar ridge of the cleft. This confirms early reports stating that even a small degree of clefting may be associated with a seemingly unrelated dental anomaly.^{34–36}

Many theories have been advanced attempting to explain why so many teeth are missing in children with clefts. These theories include multiple genetic and environmental factors, mesenchyme deficiency, and direct effect of the cleft on the primordial tissues related to the development of the lateral incisor.³⁷ As of this time, there is not enough knowledge to explain the left-side predominance for clefts and hypodontia, which was confirmed in this study.

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The prevalence of 77% hypodontia found in our combined sample of children with cleft lip, cleft palate, or both was statistically significantly higher than the rate for children without clefts, and considerably higher than the rate reported earlier for children with clefts. Hypodontia was found considerably more frequently on the left side in both the maxilla and the mandible, and clefts occurred more often on the left side, clearly indicating left-side predominance for this anomaly.

Congenital absence of permanent teeth has direct clinical implications. Early evaluation of the number of missing teeth and consideration of the size and number of the remaining teeth in both jaws should aid the clinician in planning and managing treatment. The high incidence of missing teeth in children with cleft lip and palate presents additional complications for treatment planning. Therefore, it is important to determine, at an early age, the presence or absence of specific permanent teeth and then to plan treatment accordingly. The type of malocclusion, degree of crowding and facial profile is of prime concern in determining the final treatment plan.

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TABLE 1. Distribution of Patients with Hypodontia by Sex

Sex	Patients With Hypodontia		Patients With No Hypodontia		Total	
	No.	%	No.	%	No.	%
Male	127	46	31	11	158	57
Female	86	31	34	12	120	43
Total	213	77	65	23	278	100

TABLE 2. Distribution of Hypodontia by Jaw and Location

	Missing Lateral Incisors	Missing Second Premolars			Total Missing Teeth	
		Maxilla	Mandible	Total	No.	%
	Cleft side					
Noncleft side	10	12	5	17	27	8
Total	269	47	23	70	339	100

TABLE 3. Hypodontia in Cleft Lip

Sex	Cleft Patients Total	Patients with Missing Laterals				Normal Side	Total
		Cleft Side*					
		L	R	B	T		
Male	3	1	0	0	1	1	2
Female	3	1	0	0	1	0	1
Total	6	2	0	0	2	1	3

* L indicates left; R, right; B, bilateral; and T, total.

TABLE 4. Hypodontia in Cleft Palate

Sex	Cleft Patients	Patients With Missing Laterals*				Missing Laterals	Patients With Missing Second Premolars*				Missing Premolars	Total Hypodontia
		L	R	B	T		LL	LR	LB	T		
Male	7	0	0	1	1	2	0	0	1	1	2	4
Female	23	1	0	4	5	9	1	1	0	2	2	11
Total	30	1	0	5	6	11	1	1	1	3	4	15

* L indicates left; R, right; B, bilateral; T, total; LL, lower left; LR, lower right; and LB, lower bilateral.

TABLE 5. Hypodontia in Cleft Lip and Alveolus by Tooth and Sex

Sex	Cleft Patients	No. of Patients with Missing Teeth			No. of Missing Teeth		
		Lateral Incisors	Second Premolars	Total	Lateral Incisors	Second Premolars	Total
Male	20	14	7	21	15	9	24
Female	24	16	1	17	20	2	22
Total	44	30	8	38	35	11	46

TABLE 6. Hypodontia in Cleft Lip and Alveolus by Tooth and Side

Side	Missing Lateral Incisors	Missing Second Premolars	Total
Left	18	4	22
Right	7	1	8
Bilateral	10	6	16
Total	35	11	46

TABLE 7. Hypodontia in Cleft Lip and Palate by Tooth and Sex

Sex	Cleft Patients	Missing Lateral Incisors	Missing Second Premolars	Total Missing Teeth
Male	128	139	35	174
Female	70	81	20	101
Total	198	220	55	275

TABLE 8. Hypodontia in Cleft Lip and Palate by Tooth and Side

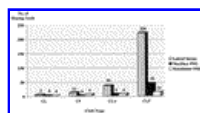
Side	Missing Lateral Incisors	Missing Second Premolars	Total
Left	78	18	96
Right	54	7	61
Bilateral	88	30	118
Total	220	55	275

TABLE 9. Distribution of Hypodontia by Cleft Type, Tooth, and Jaw Side*

Cleft Type	Maxillary Lateral Incisors				Maxillary Second Premolars					Mandibular Second Premolars				Total Missing Premolars	Total Missing Teeth	
	L	R	B	T	L	R	B	UL	UB	T	L	R	B			T
CL	3	0	0	3	0	0	0	0	0	0	0	0	0	0	0	3
CP	1	0	10	11	0	0	0	0	0	0	1	1	2	4	4	15
CLA	18	7	10	35	1	0	4	0	0	5	3	1	2	6	11	46
CLP	78	54	88	220	13	6	20	1	2	42	4	1	8	13	55	275
Total	100	61	108	269	14	6	24	1	2	47	8	3	12	23	70	339

* L indicates left; R, right; B, bilateral; T, total; LL, lower left; UL, upper left; and LB, lower bilateral; UB, upper bilateral; CL, cleft lip; CP, cleft palate; CLA, cleft lip and alveolus; and CLP, cleft lip and palate.

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FIGURE 1. Distribution of hypodontia by cleft type and jaw

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