

# A controlled study of associated dental anomalies

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In 1963, Garn and coworkers<sup>1</sup> pointed out that "little attention has been given to the real possibility that tooth number polymorphisms in man are not isolated phenomena but bear a fundamental relationship to the size, development, and calcification timing of the dentition as a whole." The association among (1) aplasia of third molars, (2) marked increase in the number of other missing teeth, (3) delayed calcification of posterior teeth in both affected children and in their siblings, and (4) differences in tooth sequence polymorphism was demonstrated. A significant relationship between variations in cusp and groove patterns of mandibular permanent molars and aplasia of these teeth was found a few years later.<sup>2</sup> Elimination of the hypoconulid

in the first two mandibular molars and absence of the third molars were described as common dental polymorphisms in man. Further contributions<sup>3-5</sup> definitely confirmed the significant direct relationship between aplasia and reduced tooth size, especially when mesiodistal crown diameter is considered.

The spectrum of possible associations among tooth anomalies was enlarged by Hoffmeister in reports between 1975 and 1985.<sup>6-8</sup> The following manifestations were found over three generations in one family: multiple missing teeth, aplasia of upper lateral incisors, peg-shaped incisors, ectopic eruption of maxillary first permanent molars, and intraosseous displacement of maxillary canines. Furthermore, 69 of 80 subjects pre-

## Abstract

The purpose of this study was to reveal patterns of association among seven types of dental anomalies (aplasia of second premolars, small size of maxillary lateral incisors, infraocclusion of primary molars, enamel hypoplasia, ectopic eruption of first molars, supernumerary teeth, and palatal displacement of maxillary canines) in an untreated orthodontic population, ages 7 to 14. The prevalence of associated tooth anomalies in seven groups of 100 subjects selected according to one primarily diagnosed dental anomaly was compared with the prevalence of the examined dental anomalies in a control group of 1,000 subjects. Significant reciprocal associations ( $p < 0.005$ ) were found among five of the anomalies (aplasia of second premolars, small size of maxillary lateral incisors, infraocclusion of primary molars, enamel hypoplasia, and palatal displacement of maxillary canines), suggesting a common genetic origin for these conditions. Supernumerary teeth appeared to be a separate etiological entity with respect to all other examined tooth anomalies. The existence of associations between different tooth anomalies is clinically relevant, as the early diagnosis of one anomaly may indicate an increased risk for others.

## Key Words

Tooth anomalies • Tooth eruption • Dental genetics.

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**Table 1**  
**Age and sex distribution of examined groups**

Groups	Number of subjects	Mean age	Males(n)	Females(n)
Aplasia of second premolars	100	10 y 2 m ( $\pm$ 2 y 4 m)	39	61
Small size of maxillary lateral incisors	100	9 y 11 m ( $\pm$ 2 y 2 m)	42	58
Infraocclusion of primary molars	100	9 y 4 m ( $\pm$ 2 y 6 m)	48	52
Enamel hypoplasia	100	10 y 1 m ( $\pm$ 2 y 8 m)	46	54
Ectopic eruption of first molars	100	8 y 7 m ( $\pm$ 1 y 4 m)	53	47
Supernumerary teeth	100	11 y 2 m ( $\pm$ 2 y 3 m)	55	45
Palatal displacement of maxillary canines	100	11 y 9 m ( $\pm$ 1 y 9 m)	41	59
Control	1,000	10 y 5 m ( $\pm$ 1 y 10 m)	482	518

**Table 2**  
**Prevalence of examined dental anomalies in control group (1000 subjects)**

Dental anomaly	Prevalence(%)
Aplasia of second premolars	5.8
Small size of maxillary lateral incisors	4.7
Infraocclusion of primary molars	5.6
Enamel hypoplasia	4.2
Ectopic eruption of first molars	4.6
Supernumerary teeth	3.9
Palatal displacement of maxillary canines	5.2

sented with various tooth anomalies associated with the infraocclusion of primary molars.

More recently,<sup>9,10</sup> taurodontic mandibular first molars were found in 34.8% of subjects with congenitally missing teeth, and infraocclusion of primary molars was found in 65.7% of the same sample.

The associations among four anomalies (ectopic eruption of first molars, infraocclusion of primary molars, ectopic eruption of maxillary canines, aplasia of premolars) was investigated by Bjerklin and coworkers in 1992.<sup>11</sup> The findings indicated the presence of a significant reciprocal association between aplasia of premolars and infraocclusion of primary molars, an association also confirmed by a subsequent study using a larger sample.<sup>12</sup> Ectopic eruption of maxillary canines increased significantly whenever any one of the other three conditions was found, while ectopic eruption of first molars increased the prevalence of associated infraocclusion of pri-

mary molars. These results were interpreted to support the hypothesis of a common, presumably hereditary, etiology for the studied tooth disturbances, each disturbance having incomplete penetrance. Finally, a very high prevalence of associated tooth anomalies (76.02%) was calculated for a sample of 169 inherited syndromes presenting with tooth disturbances, strongly suggesting the possibility of genetic relationships between the number, size, shape, and structural characteristics of the teeth.<sup>13</sup>

The aim of the present investigation was to provide further evidence supporting the existence of significant reciprocal (or one-way) associations between different types of dental anomalies in large samples of human subjects during the developmental ages compared with an adequate control group, and to indicate the etiologic and clinical relevance of such associations.

#### Materials and methods

The study was undertaken on an initial sample of 5,450 subjects (2,683 males and 2,767 females), 7 to 14 years old, drawn from the files of the Department of Orthodontics, University of Florence. Dental casts, intraoral photographs, and radiographic material of all subjects were examined. None of the subjects had received any orthodontic treatment. Out of the initial sample of 5,450 subjects, 185 were excluded due to the presence of complex craniofacial malformations, cleft lip and/or palate, sequelae of traumatic injuries to the teeth, or multiple and/or advanced caries. Another 285 subjects were excluded due to incomplete or inadequate records, racial diversity (only Caucasian subjects were included in the study), or familial relationships with other examined subjects (twins and siblings were excluded from the study). The remaining sample

of 4,980 subjects was then randomly divided into two groups. The first group of 1,000 subjects contained 482 males and 518 females and was used as the control group. The reference prevalence rates for the examined dental anomalies were calculated for this group. Seven subgroups of 100 subjects with one primarily diagnosed type of tooth anomaly each were separated from the remaining group of 3,980 subjects (Figure 1 and Table 1). The subjects belonging to one of the seven subgroups were not concomitantly present in any of the other six subgroups. The prevalence rates of the six other types of dental anomalies in association with the primarily diagnosed dental anomaly were calculated for each of the seven subgroups.

The following seven types of dental anomalies were examined:

(1) Aplasia of second premolars was diagnosed using dental casts and panoramic X-rays. In younger subjects, the absence of the germs for the second premolars was confirmed by longitudinal records.

(2) Small size of maxillary lateral incisors, defined as a severe crown-size reduction of the lateral incisors, in some cases associated with a certain degree of narrowing in diameter from the cervix to the incisal edge (peg-shaped lateral incisors).

(3) Infraocclusion of primary molars refers to primary molars that fail to maintain their position relative to adjacent teeth in the dental arch and gradually lose contact with opposing teeth, adopting an inferior position relative to the occlusal plane. Infraocclusion was diagnosed when the distance between the affected teeth and the occlusal plane was more than 1 mm.

(4) Enamel hypoplasia was diagnosed when at least one permanent tooth showed either a break in continuity or surface loss of enamel that was not related to dental caries or trauma.<sup>9,14</sup> Enamel hypoplasia was ascertained mainly from photographs and dental casts, and confirmed with radiographs whenever possible. Uncertain cases were either eliminated or recalled for clinical evaluation.

(5) Ectopic eruption of first permanent molars occurs when the first permanent molar is initially blocked from complete eruption by the adjacent primary molar, which has resorbed prematurely on its distal surface. Two possible evolutions of this conditions may follow: a reversible type of the anomaly, where the permanent molar frees itself and erupts to normal occlusion, and an irreversible type.<sup>15,16</sup> No distinction between the two forms was made in the present study.

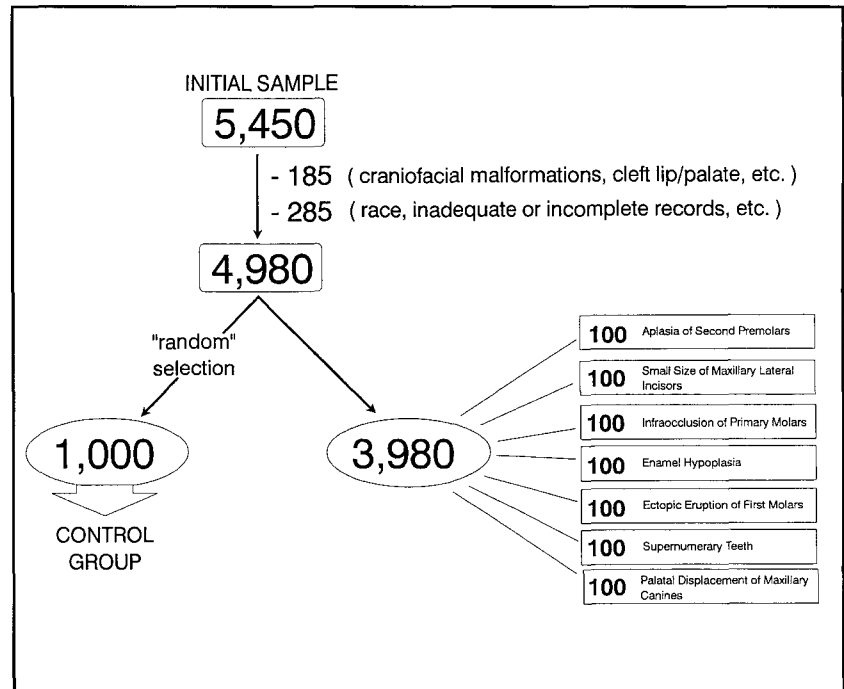


Figure 1

(6) Supernumerary teeth were diagnosed from dental casts and radiographic material.

(7) Palatal displacement of maxillary canines was based on the intraosseous palatal position of the maxillary permanent canines, unilaterally or bilaterally, as assessed on panoramic X-rays and axial and periapical radiographs.

Reproducibility of the diagnosis was assessed by re-examining the records of 200 subjects one year after the first examination. Reproducibility was complete for all dental anomalies except for small size of upper lateral incisors (94.2%) and enamel hypoplasia (94.8%).

Chi square contingency tests were used to statistically compare the prevalence of dental anomalies associated with the primarily diagnosed anomaly in the seven subgroups of 100 subjects each with the prevalence of dental anomalies in the control group of 1,000 subjects.<sup>17</sup> According to Bonferroni's correction, level of significance was increased up to  $p < .005$  in order to avoid statistical type I errors.<sup>18</sup>

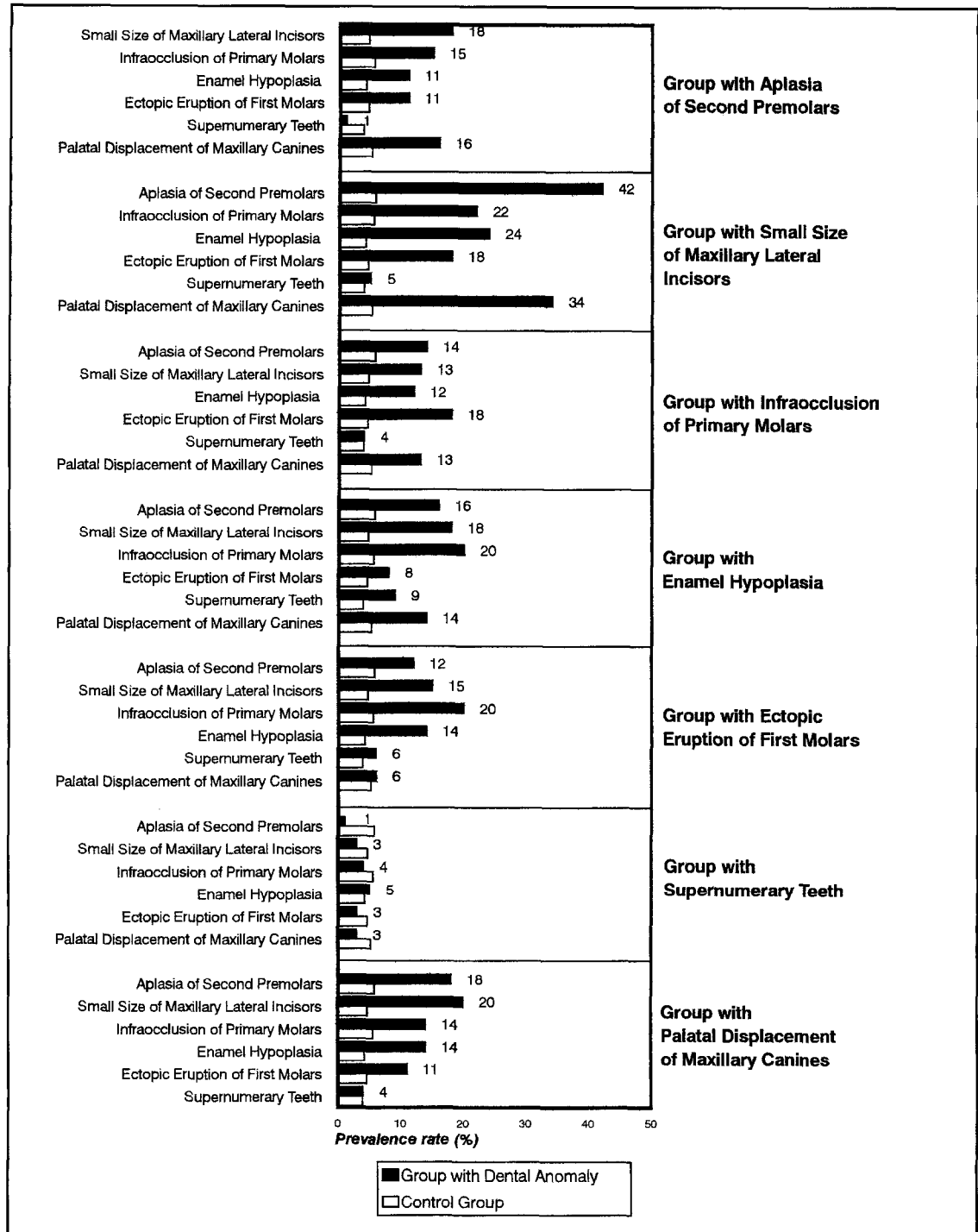
### Results

The prevalence rates of the examined dental anomalies in the control group are shown in Table 2. The prevalence rates of associated dental anomalies in each of the seven subgroups are reported in Figure 2. The results of chi square tests are shown in Table 3.

The group with aplasia of second premolars demonstrated significant associations with small size of maxillary lateral incisors, infraocclusion

Figure 1  
Construction of experimental and control groups (numbers indicate subjects)

**Figure 2**  
**Prevalence of associated dental anomalies in the seven groups of subjects selected according to the presence of one primarily diagnosed dental anomaly. See Table 2 for the prevalence rates of dental anomalies in control group.**



**Figure 2**

of primary molars, enamel hypoplasia, and palatal displacement of maxillary canines; the associations with ectopic eruption of first molars and with supernumerary teeth were not significant.

The group with small size of maxillary lateral incisors and the group with infraocclusion of primary molars showed significant associations with all other examined types of dental anomalies, except for supernumerary teeth.

The group with enamel hypoplasia presented with significant associations with aplasia of second premolars, small size of upper lateral incisors, infraocclusion of primary molars, and palatal displacement of upper canines; associations with ectopic eruption of first molars and with supernumerary teeth were not significant.

The group with ectopic eruption of first molars showed significant associations with all other examined dental anomalies except supernumerary

**Table 3**  
**Results of chi square tests applied to associations between examined dental anomalies**

PRIMARILY DIAGNOSED DENTAL ANOMALY ↓	ASSOCIATED DENTAL ANOMALIES →	Aplasia of Second Premolars	Small Size of Maxillary Lateral Incisors	Infraocclusion of Primary Molars	Enamel Hypoplasia	Ectopic Eruption of First Molars	Supernumerary Teeth	Palatal Displacement of Maxillary Canines
Aplasia of Second Premolars			26,58 *	11,79 *	8,24 *	6,33	1,43	16,46 *
Small Size of Maxillary Lateral Incisors		139,81 *		34,66 *	59,72 *	27,39 *	0,07	100,66 *
Infraocclusion of Primary Molars		8,69 *	10,58 *		10,23 *	27,39 *	0	8,81 *
Enamel Hypoplasia		13,49 *	26,58 *	27,11 *		1,58	4,51	10,96 *
Ectopic Eruption of First Molars		4,87	16,24 *	27,11 *	16,09 *		0,44	0,01
Supernumerary Teeth		3,23	0,27	0,19	0,01	0,23		0,52
Palatal Displacement of Maxillary Canines		19,18 *	34,57 *	9,41 *	16,09 *	6,33	0	

\* significant comparisons with control group  $p < 0.005$

ary teeth and palatal displacement of maxillary canines.

The group with supernumerary teeth did not show any significant associations with the other types of dental anomalies.

The group with palatal displacement of maxillary canines revealed significant associations with all other types of dental anomalies except ectopic eruption of first molars and supernumeraries.

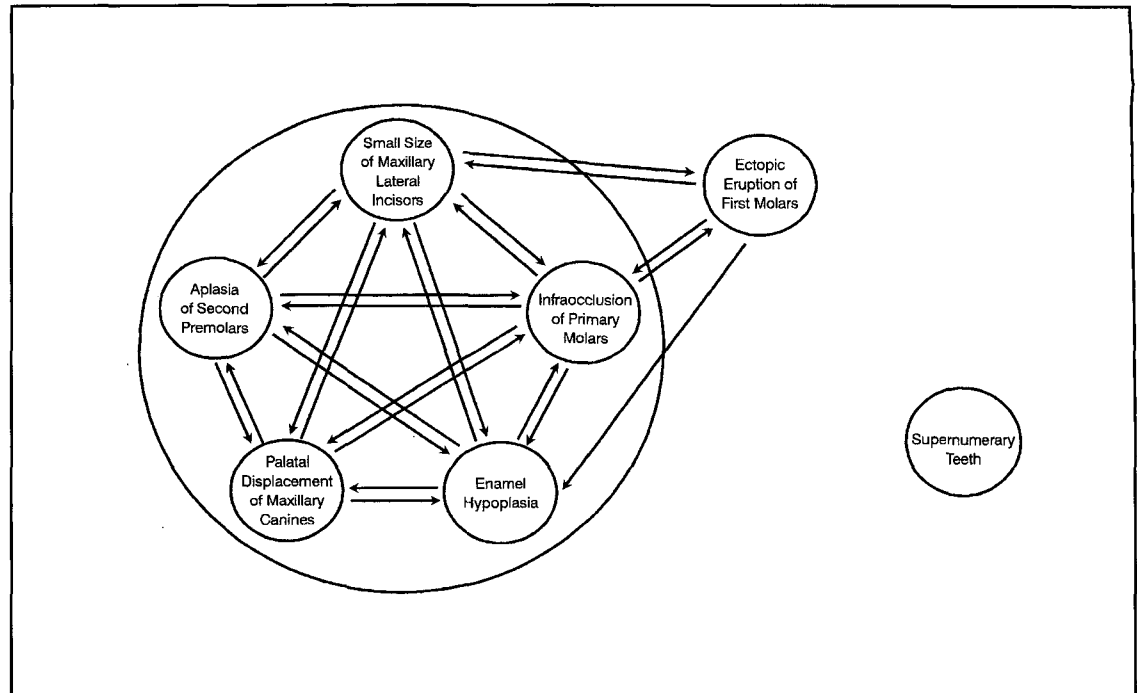
### Discussion

The aim of this study was to analyze the possibility of significant associations among different types of tooth- and eruption-disturbances in a large sample of subjects in the developmental ages. As the examined subjects had been referred to an orthodontic department, the prevalence rates of dental anomalies do not necessarily reflect the prevalence rates of these anomalies in the general population. In fact, the reference prevalence rates for some of the studied dental

anomalies in the control group appear to be quite different from those reported in previous population surveys. The reasons for these discrepancies are connected to three factors: (1) the orthodontic nature of the sample; (2) diagnostic definition of the dental anomalies—the relatively high rate of small lateral incisors is probably due to the fact that both microdontic and properly peg-shaped lateral incisors were included, whereas the relatively low rate of infraoccluded primary molars is explained by the decision to consider only primary molars lying more than 1 mm below the occlusal plane; (3) ethnic provenance of the sample—high rates for supernumeraries and low rates for infraoccluded primary molars have been described in previous studies of Italian samples.<sup>19, 20</sup>

In the present investigation the control group matched the seven experimental groups as to origin (the same untreated orthodontic population), race, age, and sex distribution. The study

**Figure 3**  
Schematic representation of the association patterns between the examined types of dental anomalies. The arrows identify "directions" of significant associations (two arrows with opposite directions connecting two types of dental anomalies represent significant *reciprocal* association)



**Figure 3**

model was constructed this way in order to overcome some methodological drawbacks of previous investigations of associated dental anomalies. Seow and Lai<sup>10</sup> assessed the prevalence of dental anomalies in association with tooth aplasia by comparing a group presenting with aplasia of various teeth with a control group comprising subjects with no tooth aplasia at all. In this way, methodologically, the possibility that one tooth anomaly may be associated with tooth aplasia in controls was excluded *a priori*. Bjerklind and coworkers<sup>11</sup> analyzed the prevalence of associated dental anomalies in individuals selected "mainly for epidemiological reasons" and compared it with data derived from various population surveys carried out by different investigators at different times.

The results of the present study revealed significant associations among different types of dental anomalies. Particular emphasis should be put on significant *reciprocal* associations between dental anomalies (e.g., the group with aplasia of second premolars showed a higher prevalence rate for small maxillary lateral incisors than the control group and, conversely, the group with

small maxillary laterals showed a higher prevalence rate of aplasia of second premolars when compared with the control group). The significant reciprocal association between defects in tooth number and defects in tooth size confirms previous findings.<sup>1,4,5,13,21</sup> It is worth pointing out that an interpretation for aplasia of succedaneous teeth as an autosomal recessive trait due to the homozygous state of the gene for small-pegged-missing maxillary lateral incisors has been proposed.<sup>22</sup>

Aplasia of second premolars showed reciprocal associations with infraocclusion of primary molars, in agreement with previous studies.<sup>11,12</sup> In 18% of the present sample and in 22% of the sample examined by Baccetti and Tollaro,<sup>12</sup> aplasia of second premolars was associated with infraocclusion of first primary molars, in contrast with the hypothesis that assigns to local factors (absence of the germ of second premolars) the primary role in the etiology of infraocclusion of corresponding deciduous molars<sup>23,24</sup> and supporting a genetic origin for infraocclusion.<sup>25</sup> In addition, infraocclusion of primary molars showed significant reciprocal associations with

ectopic eruption of first molars, small size of upper lateral incisors, enamel hypoplasia, and palatal displacement of maxillary canines.

The suggested prevalence of enamel defects in a more generalized group of tooth and eruption disturbances<sup>13</sup> was demonstrated by the significant reciprocal associations among enamel hypoplasia, small size of maxillary lateral incisors, palatal displacement of maxillary canines, and infraocclusion of primary molars.

An important aspect of dental anomalies, both clinically and speculatively, is the palatally displaced maxillary canine. At least two major theories exist in this regard. One theory is labeled "the guidance theory of palatal canine displacement" and maintains that the displacement of the maxillary canine is caused by local, predisposing factors, such as absent or anomalous lateral incisors. Missing maxillary lateral incisors, small size, or short root length of these teeth are considered "mechanical" determinants for the displacement of the maxillary canine during eruption.<sup>26-29</sup> The other theory points to genetic factors as the primary origin of the palatal displacement of the maxillary canine and of other possibly associated dental anomalies, including missing or small maxillary lateral incisors.<sup>30-32</sup>

Results of the current study corroborate the genetic hypothesis of the etiology of the anomalous position of maxillary canines. Palatal canine displacement showed significant reciprocal associations not only with small size of maxillary laterals, but also with aplasia of second premolars, infraocclusion of primary molars, and enamel hypoplasia. The clinical relevance of these findings is that early diagnosis of one or more of the tooth anomalies significantly associated with palatal canine displacement may reveal a potential risk of subsequent position and eruption anomalies of the upper permanent canines.

Ectopic eruption of first permanent molars with premature and anomalous resorption of adjacent primary molars revealed significant reciprocal associations with small size of lateral incisors and infraocclusion of primary molars, and one-way association with enamel hypoplasia. The lack of any other significant association with

other examined dental anomalies at present suggests an important role of local factors, such as tooth size-arch length discrepancy in the etiology of the dental anomaly.

Although supernumerary teeth may occur with other tooth abnormalities, they did not show any significant association with the other examined types of dental anomalies. Supernumerary teeth appear to be a separate pathologic entity. Etiologic mechanisms leading to the formation of supernumeraries are probably different from those responsible for the onset of an intensely intercorrelated group of dental anomalies. The proposed model that puts supernumerary teeth at one end of a quasi-continuous distribution of tooth size and number and missing and small-sized teeth at the other end<sup>5</sup> is not necessarily discarded.

A schematic representation of the association patterns among the seven examined types of dental anomalies is shown in Figure 3. Five tooth abnormalities (aplasia of second premolars, small size of maxillary lateral incisors, infraocclusion of primary molars, enamel hypoplasia, palatal displacement of maxillary canines) appear to be different manifestations of one syndrome with incomplete penetrance and variable expressivity; one dental anomaly (ectopic eruption of first molars) is significantly associated with only two of the dental anomalies, and supernumerary teeth represent an independent tooth anomaly with respect to the other examined tooth disturbances. The future analysis of a broader spectrum of tooth and eruption anomalies in man may reveal further or different patterns of associations.

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