

# Can persistence of primary molars be predicted in subjects with multiple tooth agenesis?

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**SUMMARY** The purpose of this research was to create a method for predicting the persistence of primary molars in patients with multiple agenesis. Dental pantomographs (DPTs) from 51 males with agenesis of 5–17 teeth and 54 females with agenesis of 5–21 teeth were investigated. All patients (6 years 9 months to 16 years 7 months) had agenesis of one or both lower second premolars. Patients with ectodermal dysplasia and craniofacial anomalies were not included. The DPTs were classified into two groups according to tooth morphology and agenesis pattern.

Group I—ectodermal symptoms: screwdriver-shaped maxillary central incisors, invaginations in incisors or narrow incisors, taurodontic molar roots, and atypical agenesis. At least two of these ectodermal symptoms had to be present for classification into group I.

Group II: one or none of the criteria for group I.

Each group was subdivided according to the number of missing teeth. The degree of root resorption of the lower second primary molar was analysed and converted to a metric scale for statistical analysis. Ectodermal status (group I versus group II) was analysed as a binary outcome with agenesis and gender as covariates (logistic regression), whereas ordinary multiple regression was performed in order to study the dependency of root resorption score on gender, ectodermal status, and age.

The study showed that subjects with agenesis of more than seven teeth belonged more often to group I than group II, also when correcting for age differences. Root resorption of the primary molars was more severe in group I than in group II.

## Introduction

In a normal dentition, the primary molar roots undergo gradual resorption concurrently with the eruption of the premolars. The normal interrelationship between the eruption of a permanent tooth and the resorption of the primary tooth is well described (Haavikko, 1973). However, it is not known what initiates and controls the two processes, eruption and resorption. It is commonly acknowledged that several factors are decisive for the eruption process as well as for the resorption process of primary tooth roots (Obersztyn, 1963; Cahill, 1969; Parner *et al.*, 2002).

The eruption of a permanent tooth can provoke resorption of the primary tooth roots, but it is also a general view that this resorption process can occur when the underlying permanent tooth is absent (Rune and Sarnäs, 1984). It is not understood why the primary tooth roots are resorbed in these cases.

It is presumed that a protective factor exists in the periodontium of the primary tooth preventing root resorption under normal conditions. This factor can undergo changes when affected by a permanent tooth in eruption. In hyper-IgE syndrome, where the primary teeth are not resorbed normally, a broadened cement-like layer is observed on the roots of the primary teeth (O'Connell *et al.*, 2000; Becktor

*et al.*, 2001). Focus has also been given to dentitions with agenesis of only one second mandibular premolar (Ith-Hansen and Kjær, 2000). In that study, root resorption of the primary molar had not progressed up to 15 years after the age of natural exfoliation.

It has been suggested that agenesis can be caused by factors related either to the mucosal ectoderm, to the ectomesenchyme, or to innervation (Kjær *et al.*, 1994). In subjects with agenesis of a single tooth, it was considered less likely that the occurrence of agenesis was caused by a general ectodermal factor but more likely by a local factor in the region, e.g. local innervation. Nerve growth factor receptor has been located in normal tooth development (Becktor *et al.*, 2002).

In a study on orthodontically provoked root resorption of permanent teeth, it has been documented that resorption occurs particularly in dentitions with morphological deviations in the incisal crowns and molar roots (taurodontia; Kjær, 1995). In that study, early pathological resorptions in the primary dentitions were also registered.

As the morphology of the crowns and roots is dependent on the inner enamel epithelium and the epithelial sheath of Hertwig, it could be presumed that the epithelium derived from the ectoderm plays a role not only in the formation of the tooth but also in root resorption. This hypothesis

substantiates the idea that an ectodermal cell layer is located in the periodontal membrane and protects against resorption. This ectodermal cell layer has been demonstrated in a recent study (Becktor *et al.*, 2007). Based on this, it is hypothesized that cases of multiple agenesis, presumably caused by genetic deviations in the ectoderm, will show other patterns of resorption in the primary dentition than those exhibited by subjects with agenesis of a single permanent tooth.

The purpose of the present study was to clarify the extent of external root resorption of the second primary mandibular molar in subjects with multiple agenesis, including agenesis of the mandibular second premolar, to enable prediction of the persistence of these primary molars.

### Material and methods

The material consisted of dental pantomographs (DPTs) from 105 children (51 males and 54 females). The males had agenesis of 5–17 teeth and the females of 5–21 teeth. The ages at the time when the radiographs were taken ranged from 7 years 10 months to 16 years 7 months for the males and from 6 years 9 months to 14 years 7 months for the females.

The radiographic material was forwarded from dentists in Denmark to the Department of Orthodontics, Copenhagen School of Dentistry, Denmark, in the years 1992–1995. The material has previously been described (Nodal *et al.*, 1994).

No radiographs were included from children with a known diagnoses of ectodermal dysplasia or craniofacial anomalies. Another criterion was that none of the patients with multiple agenesis had undergone orthodontic treatment before the radiographs were taken. A third criterion was that the patients should have agenesis of one or both second mandibular premolars and persistence of primary molars.

### Classification of material

The DPTs were divided into two groups based on the morphological characteristics of the dentitions.

Group I—morphological symptoms:

1. Screwdriver-shaped maxillary central incisors,
2. Incisor invaginations or slim incisors,
3. Taurodontic molar roots, and
4. Agenesis in regions not normally affected by agenesis, e.g. first permanent molars, permanent canines, and permanent central maxillary incisors.

At least two of these ectodermal symptoms had to be present for classification into group I. An example of a DPT of a subject in group I is illustrated in Figure 1. Seventy-one subjects (32 males and 39 females) belonged to group I.

Group II—one or none of the criteria for group I.

An example of a DPT of a subject in group II is illustrated in Figure 2. Thirty-four subjects (19 males and 15 females) belonged to group II.

### Prevalence of agenesis

Each of the two groups was divided into the following four categories according to registered agenesis:

1. Five teeth,
2. Six to seven teeth,
3. Eight to nine teeth, and
4. More than 10 teeth.

### Scoring of root resorption

On each DPT from groups I and II, a scoring of the degree of external root resorption was performed on the lower second primary molar after a method described by Haselden *et al.* (2001). The degree of resorption was converted into a metric scale (number values) for statistics (Bjerklin and Bennett, 2000). The following values were employed:

1. intact tooth—no root resorption,
2. one-quarter of the root resorbed,



**Figure 1** Dental pantomograph of a female aged 11 years 9 months. Agenesis of 10 permanent teeth (except third molars). Classified into group I due to permanent maxillary incisor morphology (screwdriver) and taurodontic first molars.



**Figure 2** Dental pantomograph of a female aged 14 years 2 months. Agenesis of five permanent teeth (except third molars). Classified into group II due to normal morphology of the incisor crowns and molar roots. Agenesis occurred in regions normally affected by agenesis.

3. root half resorbed,
4. root resorbed three-quarters,
5. root fully resorbed but the tooth still persists, and
6. tooth exfoliated.

### Reliability

Both the grouping of the material and scoring of external root resorption were assessed by two independent observers at different times. The agreement of the classification of the material was 97.4 per cent [confidence interval (CI) 86.2–99.9 per cent] equivalent to 37 agreements out of a possible 38. The percentual agreement for the resorption scoring was 92.1 per cent (CI 78.6–98.3 per cent) equivalent to 35 agreements out of a possible 38.

### Statistical analysis

Fisher's exact test was used to test the dependency between ectodermal status, gender, and the number of missing teeth. Logistic regression, with ectodermal status as the outcome, was carried out in order to include both of these as covariates simultaneously. The number of missing teeth was treated either as a linear predictor (the actual number as a continuous variable) or as a factor with agenesis grouped into four groups. The results are presented as odds ratios (ORs) with corresponding CIs.

Root resorption score was analysed in a multiple regression model, including gender, ectodermal status, and age (either as a linear predictor or as a factor corresponding to three age groups) as explanatory variables. The effects were quantified as differences with corresponding CIs.

### Results

Cross tabulation of agenesis group and ectodermal status showed an increased proportion of ectodermal cases (group I cases) in the higher agenesis groups ( $P=0.015$ , Fisher's exact test). When dividing males and females, each gender showed the same tendency although it did not reach significance.

Marginally, i.e. disregarding the agenesis group, no difference between genders in the proportion of ectodermal status could be detected ( $P=0.40$ , Fisher's exact test). These results were confirmed by a logistic regression with ectodermal status as outcome and both agenesis and gender as covariates. In this model, the OR for ectodermal status was estimated to be 1.55 (CI: 0.65–3.70) for females versus males and 1.28 (CI: 1.08–1.51) for each additional missing tooth.

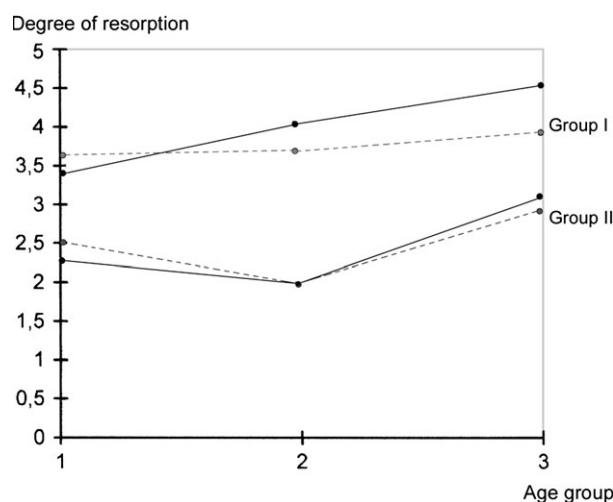
In the multiple regression model relating root resorption score to gender, ectodermal status and age, no effect of gender (estimated score difference between boys and girls was 0.18, CI=−0.37–0.72;  $P=0.52$ ) and of age (estimated increase in score for each year was 1.12, CI=−0.02–0.27;  $P=0.10$ ) was found, whereas the ectodermal group (group I) had a significantly higher score (estimated as 1.46, CI=0.89–2.04;  $P<0.001$ ; Figure 3).

### Discussion

Resorption of primary second molars in subjects with agenesis has been the subject of two classic scientific studies (Rune and Sarnäs, 1984; Sad, 1997). These concentrated on local changes in the primary teeth and did not show how these local changes were connected with the morphology of the permanent dentition. The present investigation focused on the necessity of assessing permanent tooth morphology and mapping the agenesis pattern in patients with multiple agenesis in order to predict root resorption tendency of the primary teeth.

It has previously been shown that evaluation of morphological tooth characteristics is important for predicting root resorption tendencies of permanent teeth prior to orthodontic treatment (Kjær, 1995). That study also focused on the association between predisposition and resorption in the primary and permanent dentitions. Compared with the investigation by Ith-Hansen and Kjær (2000) showing resistance to root resorption of primary molars in subjects with agenesis of only one premolar, it is obvious that the number of missing teeth is a factor that influences the resorption pattern in the primary dentition.

How reliable are the findings of the present research with regard to subjectivity, and where are the factors of uncertainty? The material in this study was collected from different orthodontic treatment units and is therefore not homogenous regarding photographic technique, distortion, enlargement, etc. The quality of the DPTs varies and is limited, which means that it was not possible to make



**Figure 3** Resorption of the primary molars related to age in groups I and II (males and females). The degree of resorption is converted to a metric scale (y-axis; Bjerklin and Bennett, 2000). Higher numbers indicate a more severe degree of resorption. In the x-axis, the ages are divided into three categories: (1) younger than 10 years, (2) between 10 and 12 years, and (3) older than 12 years. The upper two curves indicate group I and the two lower curves group II. Solid lines males and dashed lines females. Resorption of the primary second molars was more severe in group I than in group II, also when age was considered.

a confident quantitative measurement of root length. Radiographic material, particularly DPTs, are always distorted and flawed as the object is three-dimensional and projected photographically down to two planes. The morphology of the dentition is not optimal because the radiographic tube revolves around the head and serially exposes the patient who is able to move during the exposure resulting in 'shaking' and lack of clarity. This distorts the impression of the morphology of the dentition and may lead to uncertainty in the scoring of the degree of root resorption. In addition, the head posture according to beam direction is important. Most likely, the patients were positioned differently, and their head postures were varied resulting in specific rendition and enlargement.

It can be assumed that the factor, which is decisive for the ability of the periodontal membrane to resist root resorption, also affects morphological development of the teeth. In the present study, it was hypothesized that this factor, which is probably genetically determined, is connected to the ectoderm. The factor or the gene in question is not known.

It is interesting to note that the material, which came from patients who, according to the anamnestic information, had no typical phenotypic signs of ectodermal dysplasia, still showed morphological signs of ectodermal deviations. Therefore, it can be presumed that some of these patients had undiagnosed ectodermal dysplasia. This apparent contradiction can only be accepted if ectodermal dysplasia is given a wider definition than at present. The current definition of ectodermal dysplasia is not only characterized by morphological deviations in the dentition and multiple agenesis, but also by deviations in other ectodermally derived structures such as hair, skin, nails, and glands.

It is well known that ectodermal dysplasia is caused by different genes with different phenotypical manifestations. For example, acro-dermato-ungual-lacrima-tooth syndrome, affecting extremities, skin, nails, tears, and teeth caused by deviations in the P63 gene, causes malformations in teeth and external structures. The question is how the gene acts in different tissue types. In a recent study, P63 gene product was identified in the oral ectoderm (Kock *et al.*, 2005).

In the present study, the patients in group I can probably be further subdivided according to phenotype into several groups of ectodermal dysplasias that have not yet been described phenotypically or genotypically.

In order to understand how an ectodermal factor can influence both the shape of the tooth and the protection of the root in the periodontal membrane against root resorption, it is important not only to locate the ectodermal tissue in the periodontium (Becktor *et al.*, 2007) but also to clarify the actual function and genes of this ectodermal cell layer.

Classification of multiple agenesis subjects into groups according to characteristics of tooth morphology and the pattern of agenesis appears to be useful for predicting the degree of resorption of primary mandibular molars, although it must be remembered that the present study was cross-

sectional. The main point is that it is necessary to evaluate the morphology of the permanent dentition and the number of missing teeth for a prediction of the persistence of the primary molars—a factor that could be important for treatment planning during puberty when growth of the jaws prohibits insertion of implants.

## Conclusion

A classification of multiple agenesis subjects into groups according to tooth morphological characteristics and pattern and number of agenesis appears to be useful for predicting the degree of resorption of primary mandibular molars. Thus, for patients whose permanent dentitions have ectodermal phenotypes such as screwdriver-shaped incisors and/or invaginations, slim incisors and taurodontic root morphology as well as an unusual agenesis pattern (group I), the primary molars are not expected to have a long-term persistence.

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