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# An epidemiological study of dental agenesis in a primary health area in Spain: Estimated prevalence and associated factors

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## Abstract

Objectives: To evaluate the prevalence of dental agenesis and its possible association with other developmental dental anomalies and systemic entities.

Setting and Sample Population: Descriptive transversal study, for which 1518 clinical records, of patients visited by the Odontological Service of the Primary Health Centre of Cassà de la Selva (Girona-Spain) between December 2002 and February 2006 were reviewed. The data were recorded in relation to the oral and dental anomalies and the associated systemic entities, between the ones referred as concomitant in literature.

Results: Values of 9.48% (7.25% excluding the third molars) for dental agenesis and 0.39% for oligodontia were obtained. The presence of dental agenesis concomitant with some other forms of oral and dental anomalies was observed. Attention must be drawn to the fact that a greater number of concomitant systemic entities were observed in those patients that presented a severe phenotypical pattern of dental agenesis.

Conclusions: The results of the present study do not differ from the ones reported in studies of similar characteristics among Occidental and Spanish populations. The relationship observed between certain systemic entities and developmental dental anomalies suggest a possible common genetic etiology.

***Key words:*** *Hypodontia, prevalence, tooth abnormalities, craniofacial development.*

## Introduction

Congenital missing teeth is the dental anomaly which is found with a greater prevalence in Occidental populations, with values between 4.4% and 8% (1-3). It is more frequent in permanent dentition and also amongst women (1,2,4).

Many theories exist about the etiology of dental agenesis, differentiating between environmental, genetic and phylogenetic factors; not existing at the present time one theory capable of completely explain the phenomenon of the congenital absence of dental structures. Recent studies have established the relationship between certain developmental dental anomalies, such as family dental agenesis, and genetic mutations, which in turn are associated with systemic entities and multi-organ syndromes, such as colorectal polyposis and cancer, ovarian pathology, or signs and symptoms from ectodermal organs, amongst others. A relationship also observed among different animal models such as Pax9 and Msx1 deficient mice (5,6), or the relationship between Pax genes and cancerous processes (7).

In order to evaluate the prevalence of dental agenesis and the importance of the ethiological theories in our environment, the clinical records of patients treated at the Odontological Service of the Primary Health Center of Cassà de la Selva (Girona-Spain) were analyzed. The data of prevalence were recorded in relation to the possible oral and dental anomalies and the associated systemic entities, and those referred to as concomitant in recent literature (5-9).

## Population and Methods

The Odontological Service of the Primary Health Center of Cassà de la Selva (Girona-Spain), receives patients from the neighbouring towns, totaling an approximate population of 14,000 inhabitants (information supplied by the Institut d´Assistencia Sanitària de Girona and the census of the municipalities).

In order to carry out this study 1660 clinical records belonging to the Odontological Service of the Primary Health Centre were reviewed, following the regulations of the ethics committee of the Institut d´Assistencia Sanitària de Girona. These records correspond to all the patients visited in the health centre for any dental pathology during the period between December 2002 and February 2006. 142 of these clinical records were excluded from the study because the information therein was incomplete (for example radiological evidence was not available). For this reason, our study consists of 1518 clinical records: 683 corresponding to males and 835 to females, in an age group ranging from 6 to 83 years old. Then the clinical records were classified according to the clinical and radiological features observed, taking into account the following parameters:

1. Presence of congenital missing teeth, specifying the affected dental organ.

2. The relationship between dental agenesis and other types of oral and dental anomalies:

-The concomitant presence of supernumerary teeth, teeth that exceed the normal dental formula, independent of their size or shape.

-Shape anomalies like peg-shaped teeth, (that usually correspond to unirradicular teeth and are characterized by their tapered shape); and taurodontic teeth(characterized by an altered crown-to-root ratio,

corresponding to an increase in size of the middle third of the tooth) and the converse situation, cynodontia; being taken into consideration the radiological evaluation of the first and second mandibulary molars.

-Size anomalies: microdontia (characterized by a decrease in the normal size of a tooth); macrodontia (the opposite situation).

-Position anomalies, like ectopic eruption or impacted teeth.

-And the relationship with other oral anomalies, such as mandibular prognatism and retrognatism.

3. Being also taken into consideration the anamnestic data and systemic entities such as: different forms of allergies, endocrinal alterations, congenital cardiac anomalies or digestive pathologies (5-9).

All the information obtained was analyzed and processed using the software programme EXCEL- Analyse-it ® (Analyse-it Software, Ltd. Leeds, United Kingdom).

## Results

Among the 1518 clinical records that were reviewed, we obtained a prevalence of dental agenesis of 9.48% (7.25% excluding the third molars) among the population under study, being 43.75% males and a 56.25% females (Fig. 1A).

If the third molars are not taken into account, the dental organs that present a greater incidence of dental agenesis are the second lower premolars, followed by the second upper premolars, the upper lateral incisors, the upper canines and the first upper premolars (Fig. 1C).

Amongst the 1518 clinical records that were reviewed, we observed the presence of two cases of dental agenesis in primary dentition. Both cases were diagnosed in two sisters that presented the congenital absence of the primary second left molars; also, we observed the absence of the second permanent premolars in the panoramic radiography.

0.37% of the population under study presented oligodontia, 80% of the patients affected by this condition being females. It should be mentioned that four of the patients (three females and one male) with oligodontia belong to the same family.

At the same time and in a number of cases, the presence of dental agenesis concomitant with some other forms of oral and dental anomalies was observed (Fig. 1B).

We obtained a prevalence of supernumerary teeth of

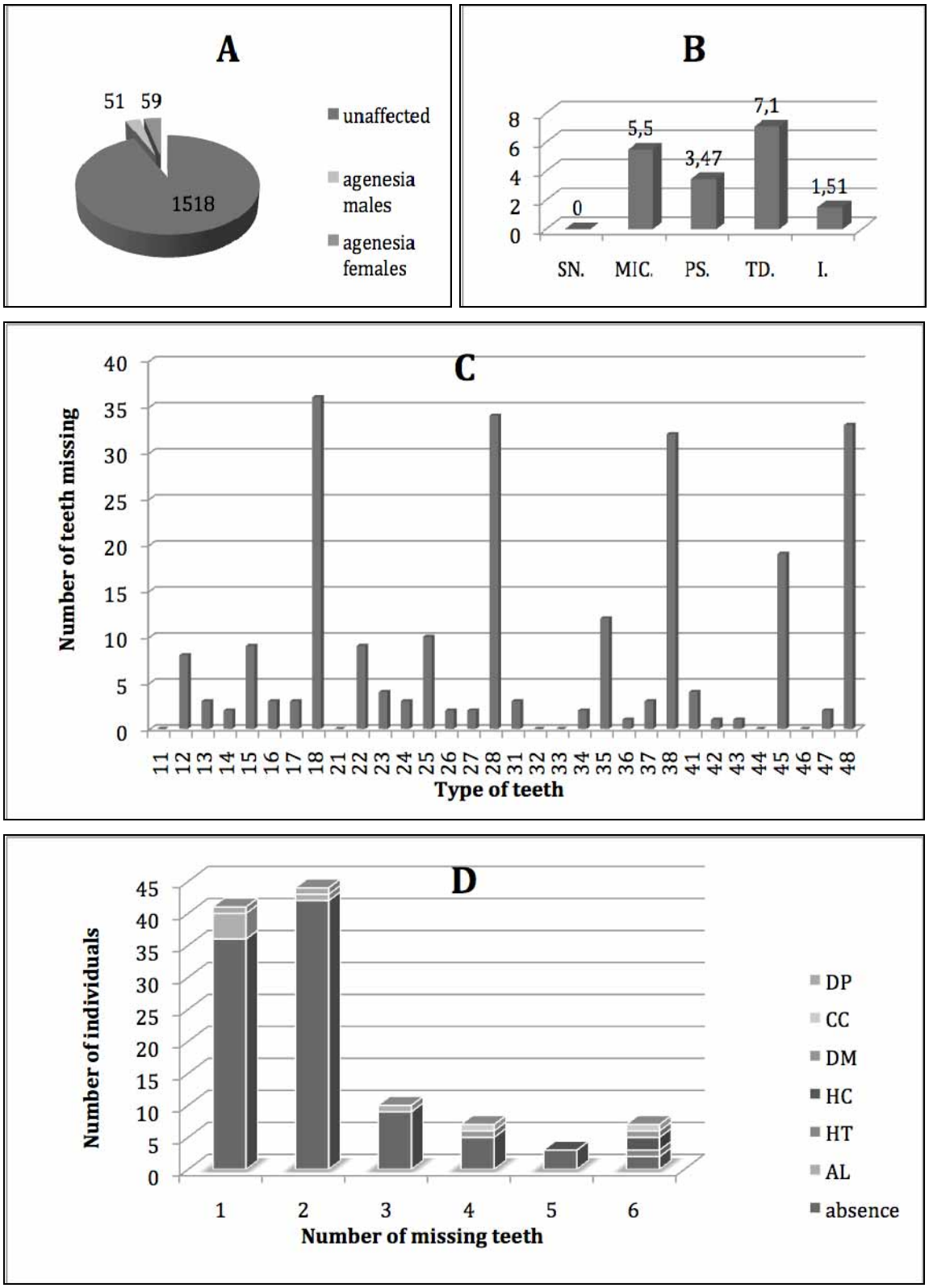


**Fig. 1.** Results: A: Dental agenesis according to gender. B: Concomitant developmental dental anomalies (%) (SN, supernumerary teeth; MIC, microdontia; PS, peg-shaped teeth; TD, taurodontism; I, impacted teeth). C: Dental agenesis according to the type of teeth absent. D: Distribution of dental agenesis according to the number of missing teeth and the presence of systemic entities (DP, digestive pathology; CC, congenital cardiac problems; DM, Diabetes mellitus; HC, hipercolesterolemy; HT, hipertyroidism; AL, allergies; absence of systemic entities).

0.39% among the population under study, 83.3% being male. One of the patients presented multiple supernumerary teeth. 33.3% of the remaining supernumerary teeth corresponded to mesiodens and the others were located in the distal region of the upper third molars. In none of the cases this characteristic trait coincided with dental agenesis.

The presence of microdontic teeth was observed in a 5.5% of the patients affected by dental agenesis, 62.5% being female. The organs most affected by this condition were the upper lateral incisors. In only one male patient, who presented the fusion of the central and lower left lateral temporary incisors, also presented macrodontia of the central lower left incisor with the agenesis of the lateral lower right incisor.

3.47% of the patients affected by dental agenesis presented peg-shaped teeth, all of them being female. The dental organs most frequently affected by this condition are the upper lateral incisors. The presence of concomitant anomalies in the homologous contralateral tooth was observed in 80% of the cases, corresponding to dental agenesis and microdontia. Taurodontism is present in 7.1% of the population under study, a value obtained by the radiological assessment of the first and second lower molars. Higher values, such as 19.3% were observed among the individuals affected by dental agenesis.

The ectopic eruption of a dental organ was observed in 3.7% of the patients presenting the absence of the equivalent tooth on the opposite side. Among the patients affected by dental agenesis the prevalence of impacted teeth (excluding third molars) was 1.51%; and of 1.45% if we only take into consideration the impacted upper canines. In 40% of the patients affected by oligodontia the presence of one or both impacted upper canines was observed.

Only 1.38% of those affected by dental agenesis presented mandibular prognatism. None of the patients affected by congenital missing teeth exhibited mandibular retrognatism.

During the study the eventual presence of some systemic conditions concomitant with dental agenesis was specifically reviewed. Attention must be drawn to the fact that a greater number of concomitant systemic conditions were found among patients that presented a severe phenotypical pattern of dental agenesis (Fig. 1D).

Specifically it was observed that 4.16% of the individuals affected by dental agenesis also presented certain forms of allergies, mainly to penicillin. And that 50% of the cases presented other associated dental anomalies. Also the relationship with certain digestive pathologies was reviewed; being observed that 3.78% of the patients that presented dental agenesis were affected by hereditary familial polyposis. The presence of two cases of coeliac disease concomitant with congenital missing teeth was also observed, associated in turn with enamel defects and a delay in eruption patterns.

Among the individuals affected by oligodontia it should be mentioned that 60% presented endocrinal pathologies and 40% congenital cardiac valve anomalies.

## Discussion

In our study of the prevalence of dental agenesis among the clinical records reviewed and corresponding to patients visited by the Odontological Service of the Primary Health Centre of Cassà de la Selva (Girona-Spain) during the period from December 2002 to February 2006, a value of 9.48% (7.25% excluding the third molars) was obtained. This figure is within the range of values obtained in several studies carried out in Occidental populations (1-4). These studies, however, are referred to pediatric or orthodontic patients, while our patients present a wider range of age and required different types of dental treatments unrelated to the existence of agenesis, so no selection bias was to be found in our study. While in other prevalence studies higher values in secondary dentition and in females were obtained (2,4,9). The greater incidence detected among females has caused several authors to suggest the possibility that the heredity pattern is related to gender (9).

If we concentrate on the number of dental organs missing, the most common form is the agenesis of two dental organs, being affected the homologous teeth in the same dental arch in 41% of cases, if we exclude the third molars. In literature no significant differences in the prevalence of dental agenesis between left and right sides have been reported. According to Daugaard-Jensen et al. (10) for primary teeth dental agenesis is more common in the upper jaw, not being described as a characteristic pattern for the permanent dentition. With reference to oligodontia, in our study a value of 0.37% was obtained, similar to the ones observed in literature (9,10). Notably, four of the patients affected belong to the same family. This data would reinforce the hypothesis of the genetic etiology of dental agenesis.

Excluding the third molars, the dental organs that showed a greater incidence of agenesis are the second lower premolars, followed by the second upper premolars, the upper lateral incisors, upper canines and the first upper premolars. These values are also obtained in studies of similar characteristics carried out among European populations (2,3,9), although there is a slightly higher value for the lower incisors obtained in our study. This is possibly related to the fact that we found several members of the same family who presented agenesis of this dental structure. The values corresponding to agenesis of the third molars are higher; literature gives us values of between 9% and less than 30% (9). At the same time 71% of the individuals presenting dental agenesis were also devoid of the third molars.

In patients presenting dental agenesis, among the teeth that are apparently unaffected alterations to shape,

size, position in the dental arch and eruption patterns can be observed, characteristics previously described in literature (7,9, 11-13). Peg-shaped lateral incisors are considered as a different phenotypical expression of the same genotype as dental agenesis. In our study, 3.47% of the patients affected by dental agenesis presented peg-shaped lateral incisors. Also, taurodontism has been related to congenital missing teeth by different authors. In the present study the incidence of this characteristic is 7.1% of the population. In literature we find similar results: Darwazeh et al. (14) obtained a result of 8% in a study carried out in a population in Jordania; as did Schalk-van der Weide et al. (15) who obtained a value of 9.9% among a Dutch population. In contrast to the prevalence of 46.4% obtained by MacDonald-Jankowski and Li (16) among a Chinese young adult population. In the present study differences between genders were not observed, a situation also referred to in literature. Higher values were obtained among the individuals presenting dental agenesis, specifically a figure of 19.3% was obtained. Values slightly lower than those found in literature: Schalk-van der Weide et al. (15) obtained a prevalence of 28.9% and Baccetti (12) of 45% among the individuals affected by dental agenesis.

Another of the anomalies taken into account is the alteration of the dental position within the dental arch. A greater frequency of dental agenesis has been reported among those patients that present transposition of canines–first premolars, palatinization of upper canines and transposition of lower lateral incisors–canines (9,11,12). In the present work the ectopic eruption of a dental organ was observed in a 3.7% of the patients presenting the absence of the contralateral equivalent tooth. The prevalence of impacted teeth excluding third molars among the population under study is 1.51% and 1.45% for the upper canines. These values are lower than the ones given in literature, which are around 3% (17).

In our study we obtained a prevalence of supernumerary teeth of 0.39%. This value is lower than the ones that we find in literature, which range from between 0.5 and 3.8% (18). Probably this difference can be explained by the fact that these studies, as also in the case of the results related to impacted teeth, have been based on patients visited at maxillary-facial surgery services.

Dental development is regulated by a high number of genes, that determine the number, size, shape and position of the different dental organs; and which in turn are also expressed in other organs and tissues. Specifically mutations in the genes MSX1, PAX9 and AXIN2, which are responsible for the regulation of the initial stages of odontogenesis, have been related to family forms of dental agenesis in humans (8,19,20). At the same time PAX9 gene has been related to neoplastic pathology and coeliac illnesses and alterations of the vertebral column development; and the MSX1 with pathologies as varied as cleft syndromes, cherubism, ovary neoplasia and, more recently, digestive tract neoplasia, as well as causing complex deformity-syndromes in experimental animals. The importance of the alterations in gene AXIN2 must be emphasized, because these alterations participate both in dental development and in the homeostatic maintenance of the colonic epithelium. Mutations in this gene have been described as the aetiology of dental agenesis and a high predisposition to colorectal cancer. The fact that a genetic anomaly causes alterations restricted to teeth, can be due to the fact that this gene is critical for the correct development of a dental organ and not for the other organs and tissues. Also this factor can be due to the non-dental effects only appearing under specific conditions that in many cases remain unidentified.

In our study, 3.47% of the patients affected by dental agenesis also presented different forms of allergy. In literature a relationship has been described between congenital missing teeth and signs and alterations of the ectodermal organs like hair and nail anomalies and low salivary flow rates (7).

Three patients suffering endocrinal pathologies presented a severe form of dental agenesis, as well as dental position, size and shape anomalies. Two of these patients, a woman and a man belonging to the same family, had familial hypercholesterolemia as well as Diabetes mellitus (I), in the male, and hyperthyroidism, in the female. According to Castell et al. (21) the Diabetes mellitus (I) prevalence in the Catalan population is 6.1% (30-64 year old age group). The factors significantly associated with this endocrine alteration are age, obesity, hypertension and a family history of diabetes. The prevalence of hyperthyroidism in the Spanish population is 2.7% for females and 0.2% for males. These values are very similar to the ones observed in the population with hypodontia, but if we only take into account the patients that have multiple congenital missing teeth these values are slightly higher.

It should be mentioned that 40% of the patients affected by oligodontia presented congenital cardiac valve alterations. Cardiac valve development is under genetic control, being related to numerous gene disruptions with alterations of valve phenotypes. Some of these have been also related with dental development, like the transmembrane protein Notch and the Wnt/β-catenin complex.

3.78% of the patients that presented dental agenesis were affected by hereditary familial polyposis, a previously described relationship (8). The presence of two individuals affected of coeliac disease concomitant with dental agenesis, enamel defects and a delay in eruption patterns was observed. No reference to the relationship between dental agenesis and this entity

has been found in literature. However Balli et al. (22) reported the existence of a positive correlation between dental and bone development delay and chronic diarrhea and coeliac disease in childhood .

The present study corresponds to a descriptive transversal study in which the prevalence of dental agenesis was analyzed in patients of the Odontological Service of the Primary Health Centre of Cassà de la Selva (Girona-Spain). Values of 9.48% (7.25% excluding third molars) were obtained; these values do not differ from the ones obtained in studies of similar characteristics among Occidental and Spanish populations.

Recently, numerous studies have established a possible etiological relationship between oral and dental anomalies and certain systemic entities. The fact that individual members of the same family were found to be affected by congenital missing teeth and other dental anomalies, as well as systemic entities, reinforces the genetic theory of the aetiology of dental agenesis. Further studies are needed to ascertain the multiple functions that each gene can have in different organs and systems.

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