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Concomitant hypodontia and hyperodontia—An analysis of nine patients

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Abstract

Introduction. Congenital absence of tooth germs and presence of supernumerary teeth (CHH) are anomalies which are classified as embryologically determined disorders. Both disorders can occur together (CHH), but relatively rarely. ***Objective.*** The aim of the present study was to present and analyze nine cases collected during the past 24 years, where congenitally missing teeth occurred with supernumerary teeth in the same patient. ***Materials and methods.*** Dental records, radiographs and casts of nine patients, six boys and three girls (aged 9–29 years, mean age: 19 years) were examined. All the family members of the patients were also investigated regarding the presence of tooth disorders. ***Results.*** The boys showed hypo-hyperodontia twice as often as the girls (6:3). Congenital absence of tooth germs was more common than the presence of supernumerary teeth (25:11). CAP were not present in the other family members. ***Conclusions.*** The full diagnosis of hypo-hyperodontia is only possible on the basis of radiographic examination, because some teeth may remain unerupted or missing. The results of this study showed that concomitant hypo-hyperodontia is rare and sex-related, with predominance of hypodontia. The genetic influence seems unclear.

Key Words: *hyperodontia, hypodontia, supernumerary teeth, tooth absence*

Introduction

Concomitant congenital absence of tooth germs and presence of supernumerary teeth are anomalies which are classified as disorders associated with the number of teeth. In 1967, Camilleri [1] introduced the term 'concomitant hypodontia and hyperodontia' to specify the prevalence of congenitally missing teeth and supernumerary teeth in the same individual. Gibson [2] in 1979 introduced its abbreviation as 'hypo-hyperodontia' and Varela et al. [3] used a term 'concomitant hypo-hyperodontia' (CHH). The condition can affect deciduous teeth, permanent teeth or both [3,4].

The prevalence of hypodontia in the permanent dentition is not common and is reported to vary from 2.6–11.3% [3], while the prevalence of hyperodontia occurs even more rarely and is reported to vary from 0.1–3.8% [3].

The etiology of hypodontia is multifactorial. Important etiological factors for hypodontia presence include genetic and environmental influences or a combination of both [3,5]. Genes responsible for congenital absence of teeth germs are diagnosed as: e.g. MSX1, MSX2, PAX9, AXIN2 [6,7]. In the etiology of hyperodontia heredity may also play a role [8]. There are few reports on CHH in the permanent dentition in the literature, mostly describing one or three cases, sometimes with limited documentation (Table I). Varela et al. [3] described six patients with CHH in the permanent dentition. However, most of the reports included only a short description of patients with CHH. Since different etiological factors have been proposed for the concomitant absence of teeth and presence of extra teeth, a detailed description and search for etiological factors are desirable. Therefore, in the present study we had examined nine patients with CHH in the permanent dentition and also their families regarding their morphology and family pattern of tooth number anomalies to better understand the CHH disorder.

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Table I. Distribution of the tooth number anomaly in 18 patients described in studies on CHH in the permanent dentition. Each patient is separately described.

		Absence		Supernumerary				
Patient's number	Study	Maxilla	Mandible	Maxilla	Mandible	Maxilla only	Mandible only	Both jaws
1	Camilleri [1]	12		Mesiodens		x		
2	Camilleri [1]	12,22		Odontoid		x		
3	Mumms [9]	12,22		25		x		
4	Nathanail [10]		35,45	Mesiodens				x
5	Szymańska-Jachimczak [11]		31	22				x
6	Spyropoulos et al. [5]	12,14, 24,25	35,44,45		41			x
7	Spyropoulos et al. [5]	13	45	Mesiodens				x
8	Spyropoulos et al. [5]		31	Mesiodens				x
9	Moore [12]	13,23		12		x		
10	Segura et al. [13]	22		Mesiodens		x		
11	Górniak et al. [8]		35	Mesiodens				x
12	Matsumoto et al. [14]	25	32	22				x
13	Sharma [15]	23		15, 14, 12, 22, 22, 24, 25	35, 34, 32, 42			x
14	Varela et al. [3]		35		42		x	
15	Varela et al. [3]		35, 45	Mesiodens				x
16	Varela et al. [3]		35	12				x
17	Varela et al. [3]		35, 45		32		x	
18	Varela et al. [3]	22		Mesiodens		x		
19	Varela et al. [3]	22		12		x		
Total: 19		17	16	22	7	7	2	9

The aim of the study was to analyze the records of patients with concomitant congenital absence of tooth germs and supernumerary teeth presence (CHH) and their families in order to present the distribution of the missing and supernumerary teeth in the jaws and to attempt a search for possible etiological factors.

Individuals and methods

The study included nine patients, aged from 9–29 years (mean age 19 years), with concomitant hypodontia and hyperodontia of permanent teeth who had been collected at the Department of Orthodontics and two private practices from 1986–2010. The patients' age, gender, number and position of the missing and supernumerary teeth and tooth morphology were recorded during clinical examination. Hypodontia was registered when congenital absence of a tooth and its germ was present while hyperodontia was registered when a supernumerary or odontoma was diagnosed on radiographs (Fig. 1). Study models were also obtained. Panoramic radiographs were taken to detect the tooth germ absence or the presence of unerupted supernumerary teeth.

Parents and siblings (if present) were clinically examined and interviewed to detect the presence of hypodontia or hyperodontia.

Results

The descriptive analysis of the examined individuals is presented in Table II. A predominance of the boys over the girls (6:3) was observed. Twenty-five teeth were congenitally absent in the examined material, that includes 10 upper second premolars, seven lower second premolars, three upper lateral incisors, two lower first molar teeth, two upper canines and one lower central incisor.

The supernumerary teeth (11) were classified as: three mesiodens, four upper lateral incisors, one lower central incisor, two lower second premolars and one odontoma in the maxilla. One patient had oligodontia in permanent dentition (congenital absence of six

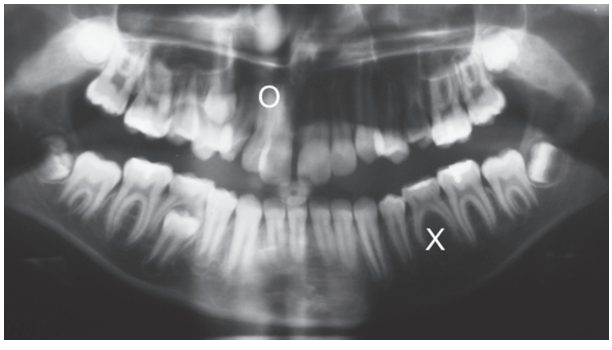


Figure 1. Panoramic radiograph of a 10-year-old female with hypodontia (x) of the lower second premolar and hyperodontia (o) of the upper lateral incisor.

Table II. Distribution of tooth absence and presence of supernumerary teeth in the examined individuals in relation to the position in the maxilla and mandible.

			Absence		Supernumerary				
Patient's number	Sex	Age (years)	Maxilla	Mandible	Maxilla	Mandible	Maxilla only	Mandible only	Both jaws
1	M	8	15, 25		2 Mesiodenses		X		
2	M	9	15, 13, 12	35, 41, 45	Odontoma				X
3	F	10	35		12				X
4	M	10	15		22		X		
5	F	11	15, 25		12		X		
6	M	12		35, 45	Mesiodens				X
7	M	13	12, 22	36, 46		35, 45			X
8	F	17	15, 14, 24, 25	35, 45	22				X
9	M	29	13			41			X
Total: 9	F: 3 M: 6	Mean:19	16	9	8	3	3	None	6

premolars) and supernumerary tooth at the position of the upper left lateral incisor (Table II, case number 8). Dentin dysplasia was recorded in one patient, but not in his family.

Hypodontia was recorded twice as often as hyperodontia (25:11). In the maxilla the hypodontia was registered twice as often as hyperodontia, while in the mandible the hypodontia was registered three-times as often as hyperodontia. CHH was never registered in the mandible only, while its frequency in both jaws was registered twice as often as in the maxilla only. From Table II it is evident that supernumerary teeth were predominantly detected in the anterior maxilla and mostly upper and lower second premolars were congenitally missing in the examined patients with CHH. Hypodontia was detected mostly on the right side of the maxilla and was symmetrically distributed in the mandible.

Four patients had Class I malocclusion with crowding, one patient had Class II div.1, one patient had Class II div.2 and three patients had Class III malocclusions.

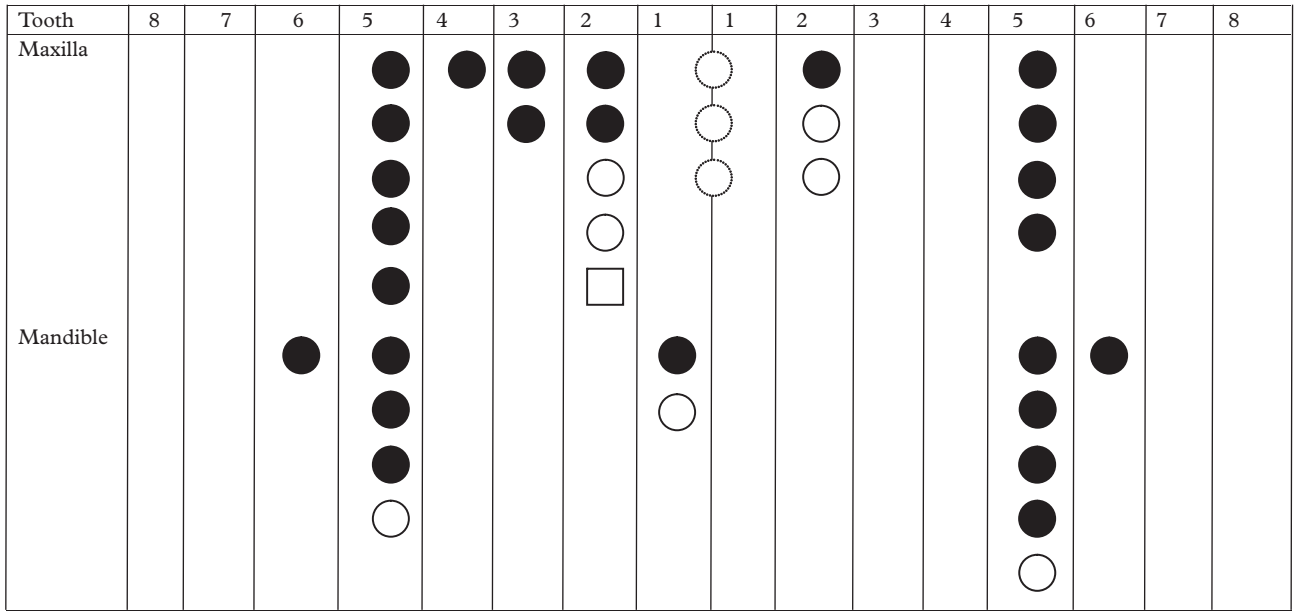
Concomitant hypo-hyperodontia (CHH) was not present in any of the examined family members.

Discussion

This study shows that both a reduction and an increase in the number of teeth can be present in the same individual. Previous papers have introduced the problem mostly in single case reports (Table I) and the present study was aiming also to present additional background factors such as the tooth distribution and family pattern in nine patients. The localization of the missing and supernumerary teeth described in the previous papers on CCH (Table I) is similar to the localization of CHH in our study (Table II), also including the presence of midline maxillary odontoma in one of our patients, similar to the Camilleri [1] report. In 1967, Mumms [9] published a case of hypodontia of two upper incisors and hyperodontia in the upper left second premolar. The CHH anomaly appeared only in the upper arch, similarly to two of our patients. In 1970, Nathanail [10] described the co-existence of missing two lower second bicuspids and the presence of a mesiodens—the case was identical to one of ours.

In 2001, Górniak et al. [8] presented the case of a 7-year-old girl who was diagnosed with agenesis of one lower second bicuspid and concomitant presence of a mesiodens. In both papers, hypodontia was diagnosed in the lower teeth and hyperodontia in the upper teeth. The same distribution of CHH was present in one of our patients (Table III). Therefore, the observed distribution of CHH in our study was similar as in the previous papers and probably represents 'normal' or most common localization of CHH in the mouth. In the present study the CHH was not detected only in the mandible in any of the presented patients. Previous reports described the presence of CHH mostly in both jaws or in maxilla

Table III. The schematic distribution of missing and supernumerary teeth in the examined individuals.



●, missing teeth; ○, supernumerary tooth; ◌, mesiodens; □, odontoma.

Figure 2. (A) Panoramic radiograph of a 13-year-old non-syndromic patient, showing congenital absence (x) upper lateral incisors and lower first molars, bilaterally supernumerary (o) lower premolars. Upper first molars were endodontically treated due to the previous periodontal problems including gingival recessions and marked hypersensitivity. Signs of possible moderate degree of Taurodontism and dentine dysplasia are present. (B) Intra-oral views of the upper and lower dental arches.

only, which was an interesting finding, except two cases described by Varela et al. [3], in whom the concomitant absence of second premolars and presence of supernumerary laterals was registered in the mandible only. Hypodontia of maxillary second premolars is the most common agenesis, but lack of association with the presence of supernumerary teeth in the mandible, except two cases described by Varela et al. [3] may be differently determined for the lower and the upper jaw, since the CHH was observed in the maxilla only (Table I). Therefore, the CHH localization may be different than hypodontia alone and may be determined differently. In our study it was also noted that hypo-hyperodontia is gender-related. The dominance of boys over girls is typical for hyperodontia, but not for hypodontia patients. Excluding mesiodenses, hypo-hyperodontia occurred mainly in the region of incisors and premolars as also previously reported [16].

Hypodontia is a multi-causal disturbance and both genetic and environmental factors have been believed to be involved in the etiology. In the etiology of hyperodontia the following factors are mentioned: anomalies in the organ structure of the first visceral arch, environmental factors and family influence [8]. The exogenous factors may change expression patterns and the extent of gene activity. So far, 300 genes have been identified to take part in the process of odontogenesis [7,17]. Many genes have been recognized to be responsible for congenital absence of teeth [3,6,17]. From the results of our study we can speculate that concomitant hypo-hyperodontia may not have family predisposition, since no CHH was present in the families of patients with CHH. In one of our patients (Figure 2A) possible signs of moderate Taurodontism were present. This condition is often associated with ectodermal conditions or X-linked disorders [18,19]. Our patient had no signs of any ectodermal disorders and this was also not detected in his family. Therefore, the etiology of the CHH seems unclear and definite conclusions regarding the genetic pattern cannot be drawn.

Early detection of concomitant hypo-hyperodontia is very important to prevent development of malocclusion in growing patients due to the lack of space (hyperodontia) or the reduction in arch length (hypodontia) (Fig. 3). In selected cases, autotransplantation of developing teeth [20] may be applied to balance the tooth number.

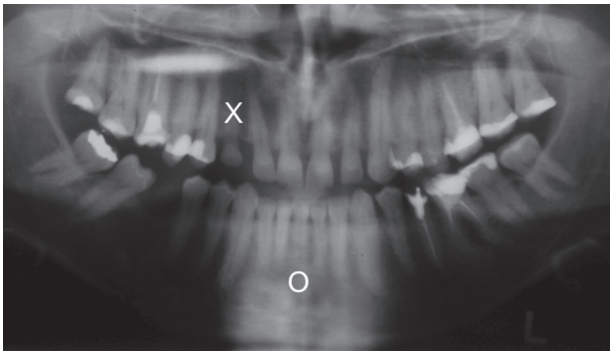


Figure 3. Panoramic radiograph of a 29-year-old patient showing the congenital absence (x) of the right side upper canine and supernumerary (o) lower incisor.

Conclusions

Co-existence of hypodontia and hyperodontia (CHH) is an uncommon disturbance and the predominance of the boys over the girls was observed. In the present study, the CHH was present in both jaws or in the maxilla, but never only in the mandible. The genetic pattern seems unclear and CHH was not registered in families of affected patients.

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Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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