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A controlled study of the association of various dental anomalies with hypodontia of permanent teeth

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Abstract

Although hypodontia, or oligodontia, is one of the most common human dental anomalies observed, there have been few studies on the association of other anomalies occurring with it. The present investigation of 1032 patient records found that 65.7% of patients with hypodontia showed ankylosis of primary molars compared to only 1.5% of control children (P < 0.001). In addition, taurodontism of the mandibular first permanent molar was observed in 34.3% of hypodontia cases compared to 7.1% in the controls (P < 0.001). Other dental anomalies significantly associated with hypodontia include enamel hypoplasia (11.9%, P < 0.01) and conical incisors (8.9%, P < 0.01). In contrast, there were significantly more impacted teeth in control children compared to the hypodontia group. The results indicate that for patients with missing permanent teeth, clinicians should be alert to the possibility of these associated anomalies and their accompanying clinical implications.

Agenesis of a few permanent teeth is one of the most common dental anomalies in man, although it is rare in other mammalian species (Lavelle and Moore 1973). A few terms have been used commonly to describe agenesis of teeth. Hypodontia is used usually to mean the absence of one or a few teeth (Stewart et al. 1982) whereas the term oligodontia is applied often for agenesis of numerous teeth, especially when associated with specific syndromes and/or severe systemic abnormalities (Gorlin et al. 1978; Stewart 1982).

Although it is now well established that agenesis of teeth may result from genetic factors (Grahnen 1956; Woolf 1971; Brook 1984), the modes of inheritance still are unclear. Many authors have suggested Mendelian patterns of inheritance (Thomsen 1952; Grahnen 1956; Alvesalo and Portin 1969), whereas others (Bailit 1975; Brook 1984) proposed that hypodontia may be the result of the interaction of many genetic factors.

Agenesis of teeth is rare in the human primary dentition. When it occurs, it is usually in the incisor region (Stewart et al. 1982). In the permanent dentition, hypodontia is seen most commonly in the third molars with a frequency of around 10%–25% in Caucasian populations (Stewart et al. 1982). However, extensive racial variation is observed. In African Negroes and Australian aborigines, the prevalence of congenitally absent third molars is approximately 1% (Townsend and Brown 1978; Stewart et al. 1982), whereas in a Japanese population the prevalence is approximately 30% (Arita and Iwagaki 1963).

Apart from the third molars, the second premolar appears to be the most commonly missing tooth in the majority of studies (Gimnes 1964; Castaldi et al. 1966; Horowitz 1966; Blayney and Hill 1967; McKibben and Brearley 1971) although some reports have shown the maxillary lateral incisor to be the most frequently missing tooth (Werther and Rothenberg 1939; Muller 1970). However, most studies have indicated that the permanent maxillary lateral incisor is the third most commonly missing tooth after the premolars (Dolder 1936; Byrd 1943; Clayton 1956; Grahnen 1956; Glenn 1961; Rose 1966; McKibben and Brearley 1971).

It is now well known that reduction in tooth size is associated with agenesis of the teeth, with many family and twin studies indicating that this defect is an expression of the same disorder (Garn et al. 1963; Cohen 1971; Gravely and Johnson 1971). In addition, ankylosis of primary molars also has been associated with hypodontia of corresponding premolars (Brown 1981; Brearley and McKibben 1973). However, these two anomalies have been studied in association with hypodontia in isolation only. It is likely that other genetic and environmental influences also may be associated with agenesis of the teeth. The present study examines the association of various dental anomalies in a group of children with hypodontia compared with a control group without the condition.

Patients and Methods

Patient records

Panoramic radiographs and clinical records were the source of data in this study. Current patient records kept at the Pediatric Dentistry Unit of the Dental School, University of Queensland, were screened to obtain 1032 records with panoramic radiographs for analysis. There were a total of 529 males and 503 females. The mean age at the time of radiography was 11 yrs 1 mo \pm 2 yrs 11 mo (range 6-19 years). All the patients were Caucasian and did not suffer dysmorphic medical syndromes expressing accompanying dental abnormalities. Past dental histories were checked to ensure that extractions of permanent teeth were not diagnosed as congenital absence.

Diagnostic criteria

Hypodontia was diagnosed from both clinical and radiographic criteria. The numbers and types of teeth missing were noted. Third molars were excluded in the consideration of hypodontia in this study of young subjects.

Radiographic Methods

Taurodontism was evaluated by measuring the crown-body, as well as the root length of the mandibular first permanent molar from the panoramic radiograph as described in an associated study (Seow and Lai 1989). This study found that measurements of the mandibular first molar did not differ significantly from those taken from a long cone periapical radiograph. Taurodontism was diagnosed if the crown-body to root length ratio was greater than 1:1 (Seow and Lai 1989).

Ankylosis was diagnosed if a tooth showed infraocclusion (Brown 1981), i.e., at least 1 mm below the occlusal plane. In all cases, this was clearly evident from the panoramic radiograph alone.

Other dental anomalies such as fusion, gemination, dilaceration of roots, and impacted teeth also were diagnosed from radiographs.

Clinical Methods

Enamel hypoplasia was diagnosed if at least one tooth showed either a break in the continuity, or surface loss of enamel which was not related to dental caries or trauma (Ainamo and Cutress 1982). This was ascertained mainly from clinical records and confirmed with radiographs whenever possible.

Hypodontia patients

Of the total number of patients selected, 66 (6.4%) had agenesis of at least one tooth. In this group there were 36 males and 30 females, and their mean age at the time of radiography was 11 yrs 1 mo \pm 2 yrs 8 mo (range 6-19 years). All these patients showed hypodontia as an

isolated trait and did not suffer these defects as part of an overall syndrome.

Control patients

A control patient who matched the study case for sex and age at radiography was selected for each case of hypodontia (36 males and 30 females). Their mean age at the time of radiography was 11 yrs 6 mo \pm 2 yrs 11 mo (range 6-19 years). All control patients were Caucasian and were shown to have radiographic evidence of complete permanent dentitions which may or may not have been erupted.

Statistical analysis

Student's *t*-test and the X² test, as appropriate, were used for statistical analysis of the data.

Results

Prevalence of hypodontia

Of the 1032 records screened, 66 patients showed hypodontia of at least one permanent tooth, giving an overall prevalence of 6.4%.

Features of hypodontia

I. Frequency of type of tooth missing

The frequency of each type of tooth missing was first analyzed. The results are shown in Fig. 1. The mandibular second premolar was the most commonly missing tooth, constituting 19.4% of all the missing teeth (61 of 314 missing teeth). Only slightly lower in prevalence was the maxillary lateral incisor, which formed 18.8% or 9 teeth. The third most commonly missing tooth was the second maxillary premolar at 39 teeth (12.4%), followed by 33 mandibular central incisors (10.2%). The other missing teeth in decreasing order of frequency include the mandibular lateral incisor (33 teeth, 7.3%), mandibular second molar (21 teeth, 6.7%), mandibular first premolar and maxillary canine (each 17 teeth, 5.4%), maxillary second molar (17 teeth, 5.1%), maxillary first premolar (13 teeth, 4.1%), mandibular canine (9 teeth, 2.9%), and last, the maxillary central incisor, maxillary first molar, and mandibular first molar (each 3 teeth, 1.0%).

II. Number of missing teeth

The mean number of missing teeth found in the hypodontia group of patients was 4.72 ± 5.04 . As shown in Table 1, more than half of these patients were missing up to 3 teeth, and the rest were missing from 4 to more than 6 teeth per subject.

III. Sex differences

The prevalence of hypodontia also was analyzed according to sex. The results showed that there were no significant differences between the sexes, with 7.0% of

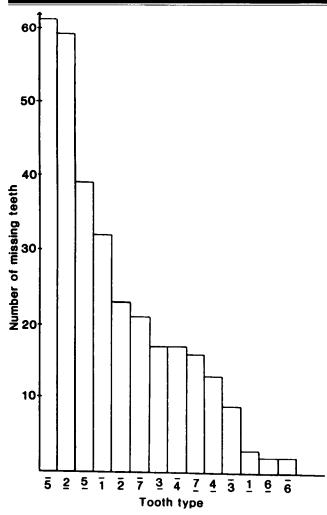


Fig 1. Frequency of each type of missing tooth.

males showing hypodontia compared to 5.8% of females (P > 0.1). In addition, the females did not show a greater tendency for more teeth to be missing than males (Table 1).

IV. Frequency of unilateral and bilateral hypodontia

In the patients with hypodontia, the frequency of unilateral and bilateral hypodontia was determined, and the results are shown in Table 2. In nearly all the tooth types analyzed, the frequency of bilaterally missing teeth exceeded greatly that of unilaterally missing teeth. Overall, 74.2% of all cases showed bilaterally missing teeth ($X^2 = 12.1$, df = 1, *P* < 0.001).

Association of various dental anomalies with hypodontia

Clinical and radiographic records of hypodontia patients and controls were analyzed for the presence of other dental anomalies. The results are shown in Table 3 (see next page). Ankylosis of primary molars was strongly associated with hypodontia, being observed in 65.7% of all hypodontia patients compared to only 1.5% in control patients (P < 0.001).

TABLE 1. Number of Missing Teeth in Patients WithHypodontia

Number of missing Permanent Teeth	Number of patients				
	Male	Female	Both Sexes (%)		
1	5	8	13 (20%)		
2	10	9	19 (29%)		
3	2	3	5 (8%)		
4	6	2	8 (12%)		
5	2	2	4 (6%)		
6	4	2	6 (9%)		
More than 6*	7	4	11 (17%)		
Total	36	30	66 (100%)		

The difference in the mean number of missing teeth in females and males is not significant, P > 0.1.

* Maximum number of missing teeth is 22.

TABLE 2. Patient Distribution of Unilateral Against Bilateral Congenital Absence

	Number of Patients			
	Unilateral	Bilateral	Total	
Maxillary teeth				
Central incisor	1	1	2	
Lateral incisor	11	24	35	
Canine	1	8	9	
First premolar	3	5	8	
Second premolar	5	17	22	
First molar	0	1	1	
Second molar	0	8	8	
Mandibular teeth				
Central incisor	2	15	17	
Lateral incisor	5	9	14	
Canine	1	4	5	
First premolar	5	6	11	
Second premolar	11	25	36	
First molar	0	1	1	
Second molar	1	10	11	
Total	46	134	180	
Percentage	25.6%	74.4%		

The difference in frequency between unilateral and bilateral hypodontia is statistically significant, $\chi^2 = 12.1$, df = 1, *P* < 0.001.

Taurodontism also was observed more commonly in the hypodontia group compared to control (34.3% vs. 7.1%, P < 0.001). In addition, peg-shaped maxillary lateral incisors were observed in 8.9% of the hypodontia group, whereas there was no patient with this trait in the control group. This difference is statistically significant (P < 0.01). Enamel hypoplasia of at least one tooth also appeared significantly more common in the hypodontia group compared to the controls (11.9% vs. 0%, P <0.001).

In contrast, in the control group impacted teeth (mandibular and maxillary canines and premolars) were observed in 28 patients (8.9%), whereas there was no such case in the hypodontia group. This difference is statistically significant (P < 0.02).

Other dental anomalies such as fusion, gemination, and root dilaceration were considered, but no statistical differences between the groups were found.

	No. of Patients Affected (percentage)			
Associated Dental Anomaly	Hypodontia Group (N = 67)	Control Group (N = 67)	P value	
Ankylosis of at least one primary molar	44 (65.7%)	1 (1.5%)	< 0.0011	
Taurodontism of mandi- bular 1st permanent molar	23 (34.3%)	5 (7.1%)	< 0.001 ²	
Peg-shaped incisors	6 (8.9%)	0	< 0.013	
Enamel hypoplasia	8 (11.9%)	0	$< 0.01^{4}$	
Fusion	1 (1.5%)	0	N.S.	
Gemination	1 (1.5%)	0	N.S.	
Root dilaceration	1 (1.5%)	4 (5.7%)	N.S.	
Impacted canines and/or premolars	0	6 (8.9%)	< 0.025	

 $^{^{1}\}chi^{2} = 64.0, df = 1$

Dental anomalies associated with different types of

hypodontia

It is interesting to note the type of hypodontia observed in association with those anomalies previously determined to be significant. The results are shown in Table 4.

Patients with multiple missing teeth also are responsible for 52.3% of all cases of ankylosis, 51.9% of taurodontism, and 75.0% of all cases of enamel hypoplasia.

In contrast, half of all cases of peg-shaped incisors are observed in patients with missing premolars, and only 16.7% in patients with multiple missing teeth.

Discussion

The present study of white patients confirms and extends some of the previous observations on hypodon-

TABLE 4. Dental Anomalies Associated With Different Typesof Hypodontia

Associated Dental Anomaly	No. of patients with missing teeth (percentage) ($N = 66$)				
	Central Incisors Only	Lateral Incisors Only	Premolars Only	Multiple Missing Teeth	Total
Ankylosis of at least one primary molar	1 (2.3%)	8 (18.2%)	12 (27.3%)	23 (52.3%)	44
Taurodontism of mandibular 1st permanent molar	0	3 (11.1%)	10 (37.0%)	14 (51.9%)	27
Peg-shaped incisors	0	2 (33.3%)	3 (50.0%)	1 (16.7%)	6
Enamel hypoplasia of at least one tooth	1 (14.3%)	0	1 (14.3%)	6 (75.0%)	8

tia. Excluding third molars, a prevalence of hypodontia in permanent teeth of 6.4% was noted, well within the range of 2.8% (Byrd 1943) and 10.2% (Ferguson et al. 1973) that had been reported previously. This suggests that although the subjects in this study were obtained from a treatment center, there is probably minimal patient selection bias.

It also was shown that the mandibular second premolar was the most commonly missing tooth, confirming the results of previous investigators (Byrd 1943; Clayton 1956; Grahnen 1956; Glenn 1961; Castaldi et al. 1966; Blayney and Hill 1967; McKibben and Brearley 1971; Hundstadbraten 1973; Silverman and Ackerman 1979). In addition, we have shown that the second most frequently missing tooth is the maxillary lateral incisor, supporting the results of Grahnen (1956), Glenn (1964), as well as Silverman and Ackerman (1979).

An important finding of this study is that hypodontia is associated with several other anomalies of the dentition. Ankylosis or submergence of primary teeth was the most frequent dental anomaly associated with missing teeth, being present in more than 65% of the oligodontia patients. Although this finding has been observed by previous investigators studying ankylosis of teeth (Steigman et al. 1973; Brown 1981), the possible reason for the association of the two anomalies has not been established. It may be postulated that both environmental and genetic factors are involved. As all the ankylosed primary teeth were associated with oligodontia of corresponding premolars, it is likely that the absence of a premolar alters the delicate physiological balance of root resorption and repair in the primary molar, resulting in ankylosis of the tooth. Alternatively, it also is possible that both hypodontia and ankylosis may be inherited as associated traits (Roberts 1973; Steigman et al. 1973).

Taurodontism, which describes a tendency for the body of a tooth to enlarge at the expense of the roots (Jaspers 1981), has been noted previously in syndromes with malformation of multiple systems which also demonstrate oligodontia. This includes the tricho-dento-osseous (TDO) syndrome (Lichtenstein et al. 1972), Kleinfelter syndrome (Stewart 1974), otodentodental dysplasia (Levin et al. 1975), ectodermal dysplasia (Stevnick et al. 1972), Down syndrome (Jaspers 1981), and Nance-Horan syndrome (Seow et al. 1985). In contrast, the present study shows that taurodontism also may be seen in patients with hypodontia uncomplicated by systemic involvement.

The finding that there is a significant association between enamel hypoplasia and hypodontia not involving systemic syndromes has not been noted

 $^{^{2}\}chi_{2} = 14.9, df = 1$

 $^{^{3}\}chi^{2} = 6.7, df = 1$

 $^{{}^{4}\}chi^{2} = 8.1, df = 1$ ${}^{5}\chi^{2} = 5.8, df = 1$

previously. It may indicate a common origin for both dental anomalies, most likely an aberration of ectodermal derivative (Barjian 1960). However, it also is possible that a single or concurrent environmental factor may have been responsible for the etiology of both defects. For example, previous authors have noted that local infection, as well as radiation, may cause both hypodontia and enamel hypoplasia (Werther and Rothenberg 1937; McCormack and Filostrat 1967; Weyman 1968).

The association of reduced incisors with agenesis of other teeth has been well documented already (Alvesalo and Portin 1969; Baum and Cohen 1971; Keene 1971; Sofaer et al. 1971). However, genetic implications of this in the inheritance of hypodontia are under debate currently. Some researchers have suggested that hypodontia is caused by a single gene with pleiotropic manifestations, which controls the size of individual teeth. Hence, peg-shaped incisors are believed to be reduced forms of the hypodontic trait. In contrast, other workers (Rose 1966; Baum and Cohen 1971) have indicated that hypodontia is likely to be controlled by a polygenic system. Various statistical analyses using single locus and polygenic models have demonstrated this possibility (Grahnen 1956; Suarez and Spence 1974).

In conclusion, the present study has found significant association of hypodontia with ankylosis of primary molars, taurodontism, enamel hypoplasia, and peg-shaped incisors. While numerous family studies have established without doubt that hypodontia is an inherited trait, the etiology of the dental anomalies associated with it is more difficult to determine. Nevertheless, the clinician should be alert to the possibility of associated anomalies in all patients with hypodontia and the accompanying clinical problems.

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- Ainamo J, Cutress TW: An epidemiological index of developmental defects of dental enamel (DDE Index). Int Dent J 32:159-67, 1982.
- Alvesalo L, Portin P: The inheritance pattern of missing, peg-shaped and strongly mesiodistally reduced upper lateral incisors. Acta Odontol Scand 27:563-75, 1969.
- Arita M, Iwagaki H: Studies on the serial observations of dentofacial regions in Japanese children. Monograph, Tokyo: Nikon University School of Dentistry, 1963.
- Bailit HL: Dental variation among populations. In Poole AE ed. Symposium on Genetics. Philadelphia: WB Saunders, 1975. p 125.
- Barjian H: The effect of early dental treatment of anhidrotic ectodermal dysplasia. J Am Dent Assoc 61:555-69, 1960.

- Baum BJ, Cohen MM: Studies on agenesis in the permanent dentition. Am J Phys Anthropol 35:125-28, 1971.
- Blayney JD, Hill IN: Congenitally missing teeth. J Am Dent Assoc 74:298-99, 1967.
- Brearley LJ, McKibben DH: Ankylosis of primary molars. ASDC J Dent Child 40:54-63, 1973.
- Brook AH: A unifying aetiological explanation for anomalies of human tooth number and size. Arch Oral Biol 29:373-78, 1984.
- Brown ID: Some further observations on submerging deciduous molars. Br J Orthod 8:99-107, 1981.
- Byrd ED: Incidence of supernumerary and congenitally missing teeth. ASDC J Dent Child 10:84-86, 1943.
- Castaldi CR, Bodnarchuk A, McRae PD: Incidence of congenital anomalies in permanent teeth of a group of Canadian children aged 6 to 9. J Can Dent Assoc 32:154-59, 1966.
- Chosack A, Eidelman E, Cohen T: Hypodontia: a polygenic trait in a family study among Israeli Jews. J Dent Res 54:16-19, 1975.
- Clayton JM: Congenital dental anomalies occurring in 3537 children. ASDC J Dent Child 23:206-8, 1956.
- Cohen MM: Agenesis and tooth size in the permanent dentition. J Int Assoc Dent Child 2:88-89, 1971.
- Dolder E: Deficient dentition. Dent Record 57:142-43, 1937.
- Ferguson NC, Worth HM, Dillabaugh GH: An investigation of the occurrence of diastemata and supernumerary teeth. J Am Dent Assoc 87:1409-10, 1973.
- Garn SM, Lewis AB, Kerewsky RS: Third molar agenesis and size reduction of the remaining teeth. Nature 200:488-89, 1963.
- Gimnes H: Congenital absence of teeth in Oslo school children. Dent Abstr 9:236-37, 1964.
- Glenn FB: Incidence of congenitally missing permanent teeth in a private pedodontic practice. ASDC J Dent Child 28:317-20, 1961.
- Glenn FB: A consecutive six year study of the prevalence of congenitally missing teeth in private practice of two geographically separated areas. ASDC J Dent Child 31:264-70, 1964.
- Gorlin RJ, Pindborg JJ, Cohen MM. Syndromes of the head and neck. New York: McGraw-Hill, 1976. pp 298-303.
- Grahnen H: Hypodontia in the permanent dentition. Odontol Revy 7(3):28-41, 1956.
- Gravely JF, Johnson DB: Variation in the expression of hypodontia in monozygotic twins. Dent Pract Dent Rec 21:212-20, 1971.
- Horowitz JM: Aplasia and malocclusion: a survey and appraisal. Am J Orthod 52:440-53, 1966.
- Hundstadbraten K: Hypodontia in the permanent dentition. ASDC J Dent Child 40:115-17, 1973.
- Jaspers MT: Taurodontism in the Down syndrome. Oral Surg 51:632-36, 1981.

Keene HJ: Epidemiologic study of tooth size variability in caries-free naval recruits. J Dent Res 50:1331-36, 1971.

- Lavelle CL, Moore WJ: The incidence of agenesis and polygenesis in the primate dentition. Am J Phys Anthropol 38:671-80, 1973.
- Levin LS, Jorgenson RJ, Cook RA: Otodental dysplasia: A "new" ectodermal dysplasia. Clin Genet 8:136-44, 1975.
- Lichtenstein J, Warson R, Jorgenson R, Dorst JP, McKusick VA: The tricho-dento-osseous (TDO) syndrome. Am J Hum Genet 24:569-82, 1972.
- McCormack J, Filostrat DJ: Injury to the teeth of succession by abscess of the temporary teeth. ASDC J Dent Child 34:501-4, 1967.
- McKibben DH, Brearley LJ: Radiographic determination of the prevalence of selected dental anomalies in children. ASDC J Dent Child 38:390-98, 1971.
- Muller TP: A survey of congenitally missing permanent teeth. J Am Dent Assoc 81:101-7, 1970.
- Roberts MW: Multiple familial dental anomalies: report of a case. ASDC J Dent Child 40:482, 1973.
- Rose JS: A survey of congenitally missing teeth, excluding third molars in good orthodontic patients. Dent Pract Dent Rec 17:107-20, 1966.

- Seow WK, Brown JP, Romaniuk K: The Nance-Horan syndrome of dental anomalies, congenital cataracts, microphthalmia and anteverted pinna: case report. Pediatr Dent 7:307-11, 1985.
- Seow WK, Lai PY: Association of taurodontism with hypodontia: A controlled study. Pediatr Dent 11:214-19, 1989.

Clin Genet 6:341-44, 1974.

- Stewart RE, Barber TK, Troutman KC, Wei SHY: Pediatric Dentistry, Scientific Foundations and Clinical Practice. St Louis, CV Mosby Co., 1982. pp 87-109.
- Suarez BK, Spence MA: The genetics of hypodontia. J Dent Res 53:781-87, 1974.
- Thomsen SO: Missing teeth with special reference to the population of Tristam de Cunha. Am J Phys Anthropol 10:155-67, 1952.
- Townsend GC, Brown T: Heritability of permanent tooth size. Am J Phys Anthropol 49:497-504, 1978.
- Werther R, Rothenberg F: Anodontia: a review of its etiology with presentation of a case. Am J Orthod 25:61-81, 1939.
- Weyman J: The effect of irradiation on developing teeth. Oral Surg 25:623-29, 1968.
- Woolf CM: Missing lateral incisors: a genetic study. Am J Hum Genet 23:289-96, 1971.

Patients accept infection control

Most Americans are not intimidated by dentists who wear gloves, masks, and protective eyewear to prevent the possible transmission of infection during dental treatment, according to a survey.

A nationwide telephone poll, conducted by researchers at the University of California-San Francisco, was the first to study how patients perceive infection control procedures in the dentist's office. The findings contradict the contention of some dentists that patients would be "scared away" if infection control procedures were used in their practices.

When asked if their dentists used protective wear, 69% of those surveyed said their dentist wears gloves; 47% said their dentist uses a mask; and 25% said eyewear was worn. The survey further revealed that infection control procedures are used somewhat more often in areas with the nation's highest prevalence of AIDS. These findings parallel reports within the profession about the use of infection control procedures.

The research team also found that one-third of the respondents had considered the risk of contracting AIDS in the dental office. In addition, 25% said it was likely that they could get AIDS from receiving dental treatment, while only 4% said it was impossible to get AIDS from a visit to a dentist. A third of the respondents said they would find another dentist if they knew their dentist was treating a patient who suffered from AIDS.