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Birth Weight and Congenital Absence of Teeth in Twins

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SUMMARY

The relationship between hypodontia and birth weight was investigated in 262 twins from the naval recruit population at Great Lakes. The overall frequency of hypodontia including third molars was 34.8%. Excluding third molars, hypodontia frequency was 11.6%. Anatomic distribution of congenitally missing teeth and frequency of third molar hypodontia were similar to other studies, but hypodontia of teeth other than third molars occurred two-three times as frequently in twins as in the general population.

The frequency of low birth weight (2500 g or less) in the twin sample was 47.7%. Hypodontia occurred more frequently in twins with low birth weight (42.6%) than in twins with birth weights over 2500 g (28.6%). Mean birth weight of twins with hypodontia was 2479 g, and without hypodontia, 2675 g. Twins with missing teeth other than third molars had a mean birth weight of 2357 g.

Mean birth weight was lower and hypodontia frequency was higher in MZ than in DZ twins. Discordances in anatomic location and number of missing teeth occurred in several MZ and DZ twin pairs. Discordances within MZ sets tended to be related to anatomic location rather than number of missing teeth. Six twin pairs (5 MZ and 1 DZ) out of 20 (13 MZ and 7 DZ) having one or more missing teeth had perfect concordance in both number and location of missing teeth. In 28 pairs neither member had hypodontia.

Mean intrapair variance in number of congenitally missing teeth was almost ten times greater in DZ than in MZ pairs, indicating the presence of a strong genetic component in connection with tooth number variability in twins.

Numerical variations in the human dentition may involve either an increase or decrease in the normal complement of 32 permanent teeth. Total anodontia or complete absence of teeth is quite rare; however, partial anodontia (hypodontia) is a relatively common anomaly which occurs with variable frequency and intensity in different populations (Brothwell et al, 1963).

The complex and etiologically heterogeneous nature of hypodontia has been mentioned by many authors (Werther and Rothenberg, 1939; Grahnen, 1956; Gorlin and Pindborg, 1964). Genetic factors are apparently of prime importance, and

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dominant, irregular dominant, recessive, sex-linked and non-sex-linked patterns of inheritance have been described (Dolder, 1936; Grahnen, 1956; Gedda et al, 1966).

Teeth may fail to develop in association with chromosome aberrations (Gorlin and Pindborg, 1964), or as part of a generalized syndrome such as chondroectodermal dysplasia (Winter and Geddes, 1967). Viruses, irradiation, and other teratogenic agents such as thalidomide which interfere with normal fetal development may also cause tooth bud agenesis to occur (Evans, 1944; Axrup et al, 1966; Weyman, 1968). In some cases such as congenital syphilis, tooth buds may be destroyed by infection (Putkonen, 1962).

The purpose of the present investigation is threefold. The first objective is to record the frequency of hypodontia and the anatomical distribution of congenitally missing teeth in a sample of twins from the naval recruit population at Great Lakes, Illinois, and to compare the findings with similar data in the literature.

The second objective of the study is to examine the relationship between birth weight and tooth number variability in the twins. In particular, this aspect of the study critically tests a previous finding (Keene, 1965) which indicated that naval recruits with a history of multiple birth had an increased frequency of hypodontia, especially when their birth weight was 2500, or less. Several authors (Allen and Kallmann, 1955; Hendricks, 1966) have noted an overrepresentation of twins in populations of mentally retarded individuals as well as an increased risk of certain congenital malformations in the offspring of multiple pregnancies. Prematurity and/or intrauterine growth retardation apparently contribute considerably to the increased morbidity and mortality associated with multiple birth (Berg and Yerushalmy, 1966) and it would be of considerable interest to determine whether these variables are in any way related to hypodontia.

The third objective of the study is to evaluate, from an analysis of twin pairs, the relative importance of genetic and nongenetic factors in the etiology of hypodontia. Consideration is given to the significance of tooth number discordances in twins and their possible relationship to intrapair birth weight disparity. Interest was aroused in this aspect of the study due to the findings of recent investigations on MZ twins which indicate a less favorable prognosis for normal physical and mental development of the lighter twin (Fogel et al, 1965). Since twins in general tend to be relatively retarded in growth at birth when compared to singletons of similar gestational age (McKeown and Record, 1952), a study of adult twins (especially twin pairs with intrapair birth weight differences) should be of considerable value in determining whether anomalies such as hypodontia are related to low birth weight due to intrauterine growth retardation (Warkany et al, 1961) or to prematurity in a temporal sense. In a previous study (Keene, 1966) it was not possible to determine whether the increased frequency of low birth weight (2500, or less) occurring in singletons with hypodontia was associated with intrauterine growth retardation or a shortened gestational period.

Methods

A total of 262 white male naval recruits with a history of twin birth were selected from the 1964-1965 recruit population at the US Naval Training Center, Great Lakes, Illinois. These men, whose ages ranged from 17 to 23 years, had passed all physical and mental requirements for entrance into the naval service at the time of examination.

The sample consisted of 49 twin pairs and 164 " single " twins. Zygosity of twin pairs was diagnosed as 17 pairs " definitely DZ " and 32 pairs " probably MZ " by multiple factor analysis which included the blood groups (ABO), the Rh series (DCEce), dermatoglyphics, phenylthiocarbamide (PTC) taste sensitivity, and a battery of morphologic observations including iris color and pattern, hair color and distribution, pinna morphology and facial morphology in profile and front view. Seventyfive of the " single " twins stated that their cotwin was female, indicating dizygosity. The zygosity of the remaining 89 " single " twins whose cotwin was male is not known.

Diagnosis of congenitally missing teeth (hypodontia) was made by clinical examination supported by radiographic confirmation in all cases. In several instances due to gross destruction and loss of teeth from dental caries, no attempt at hypodontia diagnosis was made. Four of the twins were completely edentulous due to early extraction of all permanent teeth. Since rudimentary or " peg " teeth are frequently considered incomplete expressions of hypodontia, the anatomic location of these structures was also recorded.

Data on birth weight were obtained directly from the twins' parents by questionnaire. No attempt was made to obtain information on length of gestation. In addition to the birth weight, which was initially recorded in pounds and ounces and then converted to grams, the parents were asked to cite the source of the birth weight information given. In most cases, birth certificates and announcements or family health records were consulted; however, " from memory " was also a frequently given source. For a small number of twin pairs in which interesting discordances occurred, confirmation of birth weights was obtained from the hospital in which the birth occurred.

Results

Frequency and Anatomic Distribution of Hypodontia

The distribution of twins by number of congenitally missing teeth is given in Tab. I. Out of 250 twins in which a diagnosis of hypodontia was possible, 163 (65.2%) had no missing teeth (including third molars). In the remaining 87 (34.8%) the number of missing teeth ranged from 1 to 13 (mean = 2.5) and the total number of missing teeth of all morphologic classes was 220. Most of the affected individuals had 1 to 4 teeth missing (91.9%). Four of the twins with no hypodontia had one or more rudimentary (peg) teeth and 3 twins with hypodontia had peg teeth also. The frequency of hypodontia, excluding third molars, was 11.6%.

Tab. II shows the distribution of missing teeth by morphologic class in the maxilla and mandible. Third molars accounted for 71% of the total absent teeth followed by the second premolar, lateral incisor, first premolar, central incisor and canine in order of decreasing frequency. There were no cases of hypodontia of the first and second molar, maxillary central incisor, or mandibular canine and lateral

Tab. I. Twins by number of missing teeth

No. of missing teeth[a]	Frequency N	%	Total teeth
0	163[b]	65.2	0
1	31	12.4	31
2	27	10.8	54
3	11	4.4	33
4	11	4.4	44
5	1	0.4	5
6	2	0.8	12
7	1	0.4	7
9	1	0.4	9
12	1	0.4	12
13	1	0.4	13
Total	250	100.0	220

NOTE: The overall frequency of hypodontia including third molars in 250 twins was 34.8%. The frequency of hypodontia excluding third molars was 11.6%.

[a] Third molars included.

[b] Includes 4 twins with one or more rudimentary (peg) teeth.

Tab. II. Missing teeth in twins by morphologic class

Tooth class	Maxilla R	Maxilla L	Mandible R	Mandible L	Total	%
Third molar	39(5)[a]	36(3)	40	41	156	70.9
Second molar	0	0	0	0	0	0.0
First molar	0	0	0	0	0	0.0
Second premolar	6	7	9	10	32	14.5
First premolar	2	2	0	2	6	2.7
Canine	1	0	0	0	1	0.5
Lateral incisor	12(1)	11(1)	0	0	23	10.5
Central incisor	0	0	1	1	2	0.9
Total	60	56	50	54	220	100.0

[a] Figures in parenthesis indicate rudimentary (peg) teeth.

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incisor. The maxilla was affected by hypodontia more frequently than the mandible, the missing tooth ratio being 116 : 104. A slight excess of hypodontia occurred on the right side of the maxilla and the left side of mandible, but when data for upper and lower jaws were pooled, the right-left asymmetry disappeared (110 : 110). Ten peg-shaped teeth consisting of 8 maxillary third molars and 2 maxillary lateral incisors were observed.

The distribution of 250 twins according to number of congenitally missing third molars is shown in Tab. III. Data from Grahnen (1956) and a previous study (Keene, unpubl.) on 388 caries-free naval recruits are included for comparison. Hypodontia of one or more third molars occurred in 27.6% of the twins. In 46

Tab. III. Third molar agenesis in twins compared with other samples

Third molar agenesis (Maxilla/Mandible)	Twins		Naval recruits[a]	Swedes[b]
	N	%	%	%
0/0	181	72.4	74.2	75.1
1/0	13	5.2	3.1	6.2
0/1	8	3.2	4.9	4.2
1/1	1	0.4	1.5	1.6
2/0	10	4.0	1.5	4.2
0/2	14	5.6	5.9	3.1
2/1	1	0.4	1.8	0.7
1/2	6	2.4	1.8	1.1
2/2	16	6.4	5.2	3.7
Total	250	100.0	99.9	99.9

[a] 388 caries-free white US male naval recruits (Keene, unpubl.).
[b] 547 Swedish male dental students, Group B (Grahnen, 1956).

out of 69 twins with third molar agenesis only one or two third molars were missing. In 6.4% of the entire twin group, all four third molars were missing. The distribution of congenitally missing third molars in the twins is in close agreement with the caries-free group and also with Grahnen's data.

In Tab. IV the results of an analysis of hypodontia frequency for certain individual teeth or tooth groups in the twin sample are presented with comparable data on other North American and European populations. The figures given represent the number of individuals affected with hypodontia expressed as a percentage of the total sample. The overall frequency of hypodontia of the permanent dentition in the twin group, all tooth groups being considered, was 34.8%. In the caries-free group 27.8% of the men had one or more congenitally missing teeth. The increased frequency of overall hypodontia in the twins, when compared with the caries-free group, was statistically not significant. Hypodontia of individual teeth as well as

Tab. IV. Hypodontia in twins compared with other samples

(Percentage of individuals affected)

Type of hypodontia	Twins [N=250]	Caries-free recruits [N=388]	Dolder (1936) ♂ and ♀ [N=10 000]	Grahnen (1956) ♂ [N=1078]	Garn and Rohmann (1966) [gen. pop.]	Brown (1957) ♂ and ♀ [N=5276]	Rose (1966) ♂ and ♀ [N=6000]
One or more miss., incl. M_3	34.8	27.8	—	—	—	—	—
One or more miss. or peg, incl. M_3	36.4	29.9	—	—	—	—	—
One or more M_3 miss.[a]	27.6	25.8	—	24.9	—	—	—
One or more M_3 miss. or peg	29.2	27.3	—	—	—	—	—
Maxillary M_3	18.8	14.9	—	17.6	—	—	—
Mandibular M_3	18.4	21.1	—	14.4	—	—	—
One or more miss., excl. M_3	11.6	3.6	3.4	5.9	—	4.4	4.3
One or more miss. or peg, excl. M_3	12.4	4.4	—	—	—	—	—
Maxillary lateral incisor	5.2	1.03	0.5	1.8	2.6	1.5	1.3
Maxillary lateral incisor miss. or peg	6.0	1.5	—	2.8[b]	—	—	—
Maxillary lateral incisor peg	0.8	0.5	—	—	1.6	—	0.5
One or more pegged, all classes	2.8	3.6	—	—	—	—	—
Maxillary second premolar	3.6	0.5	1.2	1.7	0.5	1.2	1.3
Mandibular second premolar	5.6	1.5	2.3	2.8	2.3	2.5	2.3
Maxillary or mandibular second premolar	7.2	2.1	—	—	—	—	3.5
Maxillary first premolar	0.8	0	0.24	0.6	0.2	—	—
Mandibular first premolar	0.8	0	0.16	0.1	0.2	—	—
Maxillary or mandibular first premolar	1.2	0	—	—	—	—	—
Mandibular central incisor	0.8	0.8	0.1	0.9	0.5	—	—
Mandibular incisor miss. or peg	0.8	1.03	—	—	—	—	—

[a] Rantanen (1967) reviewed 18 studies of third molar (M_3) agenesis in several European and "white American" populations from 1930 to 1966. In 15 out of 18 studies third molar agenesis frequency varied from 19.7 to 32.0%; in 2 studies the frequency was 9.0% and in one study, 14.0%.

[b] Grahnen's Group A, male and female.

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Tab. V. Hypodontia in various populations

Reference	Population	*N*	Frequency[a] %
Dolder, 1936	Swiss school children	10000	3.4
Werther and Rothenberg, 1939	American school children (Pennsylvania)	1000	2.3
Byrd, 1943	American dental patients (New York)	2835	2.8
Öster, 1953	Danish patients (mongoloids)	326	26.0
Clayton, 1956	American dental patients (Missouri)	3557	6.0
Grahnen, 1956	Swedish school children	1006	6.1
Grahnen, 1956	Swedish dental students and nurses	1064	5.0
Brown, 1957	American dental patients (Iowa)	5276	4.4
Valinoti, 1958	American orthodontic patients (New York)	300	6.6
Glenn, 1961	American dental patients (Florida)	777	5.2
McMillan and Kashgarian, 1961	American patients (mongoloids)	174	35.1
Bugg, 1963	American deaf and blind patients (Alabama)	941	9.2
Glenn, 1964	American dental patients (Florida)	925	5.1
Cohen and Winer, 1965	American patients, mongoloids (Massachusetts)	168	34.7
Barkla, 1966	Australian patients (mongoloids)	468	23.3
Horowitz, 1966	American orthodontic patients (New York)	1000	6.5
Rose, 1966	British orthodontic patients	6000	4.3
Keene (unpubl.)	American naval recruits (caries-free)	388	3.6
Keene (present study)	American naval recruits (twins)	250	11.6

[a] Excluding third molars.

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certain tooth groups occurs less frequently in the general population than in the twin group (Tab. IV), with higher frequencies being observed in the twins in 41 out of 44 pairings ($\chi^2 = 22.26$; $df = 1$; $P < 0.001$).

Hypodontia of permanent teeth other than third molars occurred with a frequency of 11.6% in the twins and 3.6% in the caries-free group. The difference was statistically significant ($\chi^2 = 15.45$; $df = 1$; $P < 0.001$). The increased frequency of hypodontia in the twins was primarily due to the large number of individuals with congenitally missing second premolars (7.2%) and maxillary lateral incisors (5.2%). Additional hypodontia data from several European and North American populations are presented in Tab. V for comparison with the twin group. In all cases "frequency" represents the percentage of individuals affected with one or more congenitally missing teeth, excluding third molars.

Hypodontia of the maxillary lateral incisor occurred in 5.2% of the twins and in 1.03% of the caries-free group. The difference was statistically significant ($\chi^2 = 10.19$; $df = 1$; $0.001 < P < 0.01$). Most reports on maxillary lateral incisor hypodontia in the general population indicate a frequency approximating 1-2% (Meskin and Gorlin, 1963).

Birth Weight and Hypodontia

Birth weight data were available on 241 out of 250 twins in which a diagnosis relative to hypodontia was made. The frequency of "prematurity" (birth weight of 2500 g or less) in the twin sample was 47.7%, as compared to 7-8% in single births and 53.0% for plural births in the general population (Silverman, 1964).

Tab. VI shows the relationship between birth weight and hypodontia of the permanent dentition (including third molars) in 241 twins. At the two extremes of the birth weight distribution hypodontia frequency varied from a relatively high 66.7% in twins with birth weights under 1620 g to a low of 20% when birth weight exceeded 3408 g. The frequency of hypodontia in 115 twins with birth weights of 2500 g or less, was 42.6%, while in 126 twins with birth weights over 2500 g, it was 28.6%. A χ^2 analysis of this distribution indicated that the increased frequency of hypodontia in twins whose birth weight did not exceed 2500 g was statistically significant ($\chi^2 = 5.189$; $df = 1$; $0.025 < P < 0.05$).

The relationship between birth weight and number of congenitally missing teeth (including third molars) in 241 twins is shown in Tab. VIII. The mean birth weight of 85 twins with one or more congenitally missing teeth was 2479 g, and for 156 twins with no missing teeth, 2675 g. The difference in mean birth weight between these two groups was statistically significant ($t = 2.58$; $df = 240$; $P = 0.01$). Within the twin group with hypodontia, mean birth weight did not appear to be related to number of congenitally missing teeth. The lowest mean birth weight occurred in twins with only 1 tooth missing (2298 g), and the highest mean birth weight was found in twins with 4 teeth missing (2667 g), which almost equalled the mean birth weight of twins with no congenitally missing teeth.

Tab. VI. Birth weight and hypodontia
[$N = 241$]

Birth weight[a]		No. of twins	Hypodontia[b] N	Hypodontia[b] %	
...-1620	(... to 3-9)	12	8	66.7	
1620-2045	(3-9 to 4-8)	32	12	37.6	42.6*
2046-2500	(4-9 to 5-8)	71	29	40.8	
2501-2954	(5-9 to 6-8)	69	20	29.0	
2955-3408	(6-9 to 7-8)	42	13	31.0	28.6*
3409-...	(7-9 to ...)	15	3	20.0	
	Total	241	85	35.3	

* Difference between groups was statistically significant ($\chi^2 = 5.189$; $df = 1$; $0.025 < P < 0.05$).
[a] Birth weight in grams; corresponding weights in pounds and ounces are indicated in parentheses.
[b] Third molars included.

Tab. VII. Birth weight and missing teeth

No. of congenitally missing teeth[a]		No. of twins	Birth weight Mean	Birth weight Range	Birth weight SD
A.	0	156	2675*	1335-4260	570.3
B.	1 or more[b]	85	2479*	1079-4090	547.3
C.	1	30	2298	1079-3294	
D.	2	26	2604	1477-4090	
E.	3	11	2448	1590-3862	
F.	4	11	2667	1988-3351	
G.	5 or more	7	2530	1590-3351	
	Total	241	2604	1079-4260	562.0

* Difference in mean birth weight between groups A and B statistically significant ($t = 2.581$; $df = 240$; $P = 0.01$).
[a] Including third molars.
[b] Groups C, D, E, F, G are subgroups of B.

An interesting finding shown in Tab. VIII was the relatively low mean birth weight (2357 g) observed in the 19 twins who had agenesis of teeth exclusive of the third molar area, i.e., maxillary lateral incisors and/or maxillary or mandibular premolars. Mean birth weight for these men was significantly lower than in the 156 twins with no hypodontia ($t = 2.28$; $df = 173$; $0.02 < P < 0.05$), but not significantly lower than in the 55 twins with third molar hypodontia. Twins with hypo-

Tab. VIII. Birth weight and location of missing teeth

	Congenitally missing teeth (location)	No. of twins [$N = 241$]	Birth weight (g) Mean	SD	$P \leq 0.05$*
A.	None missing, incl. M_3	156	2675	570.3	A × B
B.	One or more missing, incl. M_3	85	2479	547.3	A × D
C.	Third molars only	55	2530	613.7	A × E
D.	One or more missing, excl. M_3	19	2357	571.7	
E.	One or more missing $\pm$ M_3[a]	30	2386	585.0	
F.	Maxillary lateral incisors	12	2505	557.2	
G.	Max. or mand. premolars	18	2377	696.4	

* Difference in mean birth weight between groups significant at the 5% level or better.

[a] One or more missing teeth excluding third molars, although third molars also missing in some cases.

dontia in the premolar region had a lower mean birth weight (2377 g) than twins with hypodontia of the maxillary lateral incisors (2505 g). The difference was not statistically significant. Twins with third molar hypodontia had a mean birth weight of 2530 g which was not significantly different from that of twins with no hypodontia.

Hypodontia and Birth Weight in Twin Pairs

In the group of 250 twins in which a diagnosis relative to hypodontia was made, 48 pairs were observed, of which 17 were classified as " definitely DZ " and 31 as " probably MZ ". Mean birth weight of the DZ pairs (2852 g) was significantly higher ($t = 4.11$; $df = 92$; $P < 0.001$) than mean birth weight of MZ twin pairs (2401 g). For the 71 DZ twins whose cotwin was female, mean birth weight (2673 g) was also significantly higher ($t = 2.74$; $df = 129$; $0.001 < P < 0.01$) than the MZ twins, but not significantly different from either DZ pairs or twins of unknown zygosity whose male cotwin was not available for examination (Tab. IX). The latter group, probably containing a mixture of MZ and DZ twins, had a mean birth weight of 2613 g, which was significantly lower than DZ pairs ($t = 1.99$; $df = 117$; $P = 0.05$) and significantly larger than MZ pairs ($t = 2.07$; $df = 143$; $0.02 < P < 0.05$). A number of investigations have shown that MZ tend to have lower birth weights than DZ twins (Wilson and Jones, 1931; Naeye and Letts, 1964).

The frequency of hypodontia in general and of certain specific teeth such as the maxillary lateral incisor or maxillary and mandibular premolars appears to be considerably higher in MZ than in DZ twins. It should be noted, however, that in all instances the frequency data (Tab. IX) are based upon less than 100 (and in several cases less than 50) individuals. The 19.4% figure for hypodontia (excluding third molars) and the 11.3% frequency of maxillary lateral incisor agenesis for MZ twins seems rather high, almost approaching and exceeding in some cases the frequencies reported for certain pathologic populations (Tab. V). It is of in-

Tab. IX. Zygosity, birth weight and hypodontia

Zygosity	No. of pairs	Birth weight			Hypodontia frequency (% affected)				
		N	Mean	SD	Incl. M_3	Excl. M_3	Max. lat. incisor	Premolar	M_3
MZ	31	60	2401	548.1	40.3	19.4	11.3	9.7	27.4
DZ	17	34	2852	423.2	32.4	8.8	2.9	5.9	26.5
DZ	75[a]	71	2673	576.5	27.8	7.0	0	7.0	22.2
Unknown	89[b]	85	2613	641.8	37.3	10.8	6.0	6.0	31.3
	Total	250	2605	570.8	34.8	11.6	5.2	7.2	27.6

[a] Female cotwins unexamined.
[b] Male cotwins unexamined.

terest to note in this regard that several authors (McKeown and Record, 1960; Hendricks, 1966) have considered twinning, especially MZ, to be somewhat a pathologic event, marked by high fetal mortality and increased risk of congenital malformations.

Concordance and Discordance in Twin Pairs

Tables X and XI summarize some of the findings pertaining to intrapair differences in birth weight and in hypodontia status for 48 pairs of MZ and DZ twins. In 20 pairs at least one member of the set had hypodontia, and in 28 pairs neither member was affected. If anatomic location and number of congenitally missing teeth are disregarded, 12 out of 13 affected MZ sets and 4 out of 7 affected DZ sets were concordant for hypodontia of one or more teeth. In the discordant MZ set a maxillary third molar was missing in one twin, and not in the other. For the three discordant DZ pairs in which one twin was unaffected, the other twin was missing an upper third molar, both upper third molars, and all four third molars respectively.

Of the 16 twin pairs (12 MZ and 4 DZ) concordant for hypodontia only 6 (5 MZ and 1 DZ) demonstrated perfect concordance in both number and anatomic location of congenitally missing teeth. In several pairs there was concordance for number of missing teeth but discordance in anatomic location. In other cases discordance in both number and location of missing teeth occurred. Discordance in anatomic location of missing teeth within a twin pair may be explained in some cases by the "mirror-image" concept (Price, 1950); however, there was no evidence of such an effect in the present material. Discordance within MZ sets tended to be related to anatomic location rather than total number of missing teeth. Three MZ sets were discordant for number of missing teeth. In set MZ-3, in which a 30% difference in birth weight was observed, the heavier twin (2272 g) had three

Tab. X. Twin concordance in hypodontia

Hypodontia	DZ	MZ	Total
1. No hypodontia (both twins)	10	18	28
2. Hypodontia, one or more teeth[a]			
Concordant	4	12	16
Discordant	3	1	4
3. Hypodontia, one or more teeth[b]			
Concordant	2	10	12
Discordant	5	3	8
4. Hypodontia, one or more teeth[c]			
Concordant	1	5	6
Discordant	6	8	14

NOTE: Groups 2, 3 and 4 each consist of the same 40 individuals.

[a] Disregarding number and location of missing teeth.

[b] Disregarding location, but the number of missing teeth being the same in both twins.

[c] Same location and number of missing teeth in both twins: perfect concordance.

missing teeth and the lighter twin (1590 g) had six missing teeth. In the other two discordant MZ sets (MZ-9 and MZ-10) birth weight disparity was not as great, and there was an intrapair difference of only one missing tooth. It is of interest to note that in the latter cases, the twin with the heavier birth weight had more missing teeth than the lighter twin. In DZ pairs discordant for number of missing teeth, the more severely affected twin had a lower intrapair birth weight in 3 out of 5 cases. When the 8 MZ and DZ twin sets discordant for number of congenitally missing teeth were considered in relation to intrapair birth weight disparity, the more severely affected member of a pair was the lighter twin in 50% of the cases (4 : 4).

An estimation of the relative importance of genetic and environmental factors in tooth number variability in twins may be obtained by calculating mean intrapair variance (S^2) in total tooth number (32 minus number of congenitally missing teeth) for MZ and DZ twin pairs (Osborne, 1962). Disregarding anatomic location of congenitally missing teeth, mean intrapair variance may be obtained by the formula: $S^2 = \Sigma x^2/2N$, where x equals the intrapair difference in tooth number and N equals the number of twin pairs.

As may be seen in Tab. XII, the mean intrapair variance for DZ twins was almost 10 times as great as that for MZ twins, indicating that MZ twin pairs are significantly ($P<0.001$) more alike in total tooth number than DZ twin pairs. Since it is likely that " measurement error " variance is negligible, the data seem to further validate the presence of a strong genetic component in connection with tooth

Tab. XI. Hypodontia and birth weight
[13 MZ and 7 DZ twin pairs]

Twin pairs	Missing teeth (location)[a]	Birth weight	Difference (%)
DZ-3a	1	3181	
DZ-3b	none	3692	13.8
DZ-8a	none	3067	
DZ-8b	1, 16, 17, 32	3351	8.5
DZ-11a	none	2442	
DZ-11b	1, 16	2897	15.7
DZ-12a	1, 13, 16, 17, 20, 29, 32	2783	
DZ-12b	13	2812	1.0
DZ-15a	17, 32	3294	
DZ-15b	17, 32	3380	2.5
DZ-16a	7	3209	
DZ-16b	(1), (7), 16	3238	0.9
DZ-17a	17, 32	2130	
DZ-17b	32	2300	7.4
MZ-3a	1, 16, 17, 20, 21, 32	1590	
MZ-3b	1, 4, 16	2272	30.0
MZ-4a	32	1590	
MZ-4b	32	1818	12.5
MZ-5a	4, 13, 29	1590	
MZ-5b	4, 13, 16	1931	17.6
MZ-6a	17, 32	2045	
MZ-6b	17, 32	2215	7.7
MZ-9a	1, 4, 5, 6, 7, 10, 12, 13, 16, 17, 20, 32	3096	
MZ-9b	1, 4, 5, 7, 10, 12, 13, 16, 17, 20, 21, 29, 32	3351	7.6
MZ-10a	none	2272	
MZ-10b	1	2670	14.9
MZ-16a	7, 10	2045	
MZ-16b	7, 10	2386	14.3
MZ-21a	1, 16	2414	
MZ-21b	1, 16	2954	18.3
MZ-23a	16	1164	
MZ-23b	17	1363	14.6
MZ-25a	17, 32	2158	
MZ-25b	1, (10), (16), 17	2272	5.0
MZ-28a	24	2102	
MZ-28b	7	2329	9.8
MZ-32a	7, 10	2329	
MZ-32b	7, 10	2414	3.5
MZ-34a	1, 16, 17	2584	
MZ-34b	16, 17, 32	2755	6.2

[a] Universal nomenclature system (Frykholm and Lysell, 1962). Maxillary teeth are numbered from 1 to 16 starting with the right third molar, mandibular teeth are numbered from 17 to 32 starting with the left third molar. For example, 1, 16, 17, 32 are the third molars; 7, 10 are the right and left maxillary lateral incisors. Teeth numbers enclosed by parentheses indicate rudimentary (peg) teeth.

number variability in twins. As Gedda et al (1966) have shown in a pedigree of 32 individuals from 4 generations, concordant hypodontia in an isolated MZ twin set may be simply the result of a straight-forward autosomal dominant pattern of inheritance. If however, as the literature suggests, nongenetic factors are also involved in the etiology of hypodontia, it is likely that phenocopies may occur which are indistinguishable from the " inherited " type. In the absence of additional in-

Tab. XII. Analysis of intrapair variance in tooth number

Zygosity	N^a	S^{2b}	F	P
DZ	17	1.706	9.64	<0.001
MZ	31	0.177		

NOTE: Tooth number = 32 minus number of congenitally missing teeth.

[a] Number of twin pairs.

[b] Mean variance = $\Sigma x^2/2N$ (x = difference in tooth number between pairs).

formation relative to hypodontia in the families of the twin pairs in the present study, it would be presumptuous to conclude from the available data that the anomalies in question are " inherited ", especially since twins seem to experience a greater frequency of hypodontia than is found in the general population.

An interesting aspect of the biology of twinning is the problem of birth weight disparity between members of a twin pair. In some of the present cases (MZ-3) the difference in birth weight was as high as 30%. It is of interest to note (Tab. XIII) that DZ twins tended to be slightly more alike in birth weight than MZ twins. Mean intrapair variance in birth weight for MZ twins was 1.02 times higher than for DZ twins, but the differences were not statistically significant. Other investigators have reported similar findings (Bender, 1959; Naeye and Letts, 1964).

Tab. XIII. Analysis of intrapair variance in birth weight

Zygosity	N^a	S^{2b}	F	P
DZ	17	59295.6	1.02	>0.05
MZ	30	60563.2		

NOTE: Birth weight in grams.

[a] Number of twin pairs.

[b] Mean variance = $\Sigma x^2/2N$ (x = difference in birth weight between pairs).

Discussion

The increased frequency of hypodontia which was noted in an earlier study (Keene, 1965) on a limited number of twins from the naval recruit population at Great Lakes has been confirmed in the present report on 262 twins from the same population. Hypodontia of one or more teeth including third molars occurred with a frequency of 34.8%; excluding third molars, the frequency was 11.6%. In view of the many references to this interesting subject which have appeared in the literature during the past fifty years it is remarkable that data pertaining to frequency of hypodontia in twins are not available for comparison. In many instances, single case reports are given, noting " interesting " discordances or concordances in various types of twin pairs; the method of determining zygosity is rarely described. In studies of larger numbers of twins, techniques used for zygosity diagnosis may be given, but data are often presented in an ambiguous manner which precludes the possibility of calculating hypodontia frequency.

Siemens and Hunold (1924), in a study of 56 MZ and 35 DZ pairs, found 1 MZ pair concordant for bilateral agenesis of the maxillary lateral incisors. Praeger (1924) studied 26 MZ pairs and noted concordance for maxillary premolar hypodontia in 1 set. Korkhaus (1929) reported concordant hypodontia in 1 DZ pair and 2 MZ pairs. In 1 of his MZ pairs the first twin was missing both maxillary lateral incisors and the cotwin lacked second bicuspids. Goldberg (1930), in a series of 50 MZ pairs, noted 2 pairs with concordant unilateral maxillary lateral incisor hypodontia with reversed asymmetry (mirror-image) in both sets. Two cases of perfect hypodontia concordance in MZ twins were reported by Zeiger and Winkler (1931). In the first set each twin had bilateral second premolar agenesis; in the other set maxillary and mandibular second premolars and mandibular second molars were missing bilaterally in each twin. Perfect concordance for number and location of missing teeth in MZ twins has also been reported by Burman (1944), Lasker (1950), Horowitz (1963) and Garn et al (1963).

Ritter (1933) observed discordance for hypodontia in 6 MZ pairs, which was explained by variation in penetrance of a very unstable genetic factor. He also noted discordances in 9 DZ pairs with " incomplete discordance " in 2 pairs. In a later publication Ritter (1937) described 5 pairs of MZ twins discordant for hypodontia. MZ twins discordant for number and/or location of congenitally missing teeth have been reported by Euler and Ritter (1939), Kennedy (1950), Tanner (1955) and Greenberg (1961). In one report (Horowitz, 1963) the discordance was due to reversed asymmetry, suggesting a " mirror-image " effect.

Discordances may be associated with agenesis in one twin and not the other; agenesis in both twins, but differences in number (location also) of missing teeth; concordance in number of missing teeth but discordance in anatomic location (not including cases with evidence of " mirror-image " effect). In the present study, all types of discordance (excluding " mirror-image " type) were observed in MZ

twins. Out of 13 MZ twin sets in which at least one member of the set was affected, only 5 demonstrated perfect concordance for number and location of missing teeth.

In some cases hypodontia in twins may be associated with ectodermal dysplasia. Stern and Cranin (1955) reported an interesting pedigree with a DZ male twin and a male sib affected. The female cotwin and a female sib were unaffected. The mother, father, two aunts and two uncles (mother's side) were unaffected. Three of the mother's brothers had ectodermal dysplasia and two of these were feeble-minded. Roberts and Gardiner (1959), noted partial concordance for hypodontia in a pair of unlike-sex DZ twins with ectodermal dysplasia (hydrotic type). The female twin (birth weight 2272 g) had 25 missing permanent teeth and the male twin (birth weight 2726 g) had 18 missing teeth. The male DZ twins reported by Hinrichsen (1963) were concordant for ectodermal dysplasia and partially concordant for hypodontia, with one of the twins missing 29 permanent teeth and the other, 26 teeth.

Although the present data indicate a birth weight effect on hypodontia frequency with higher frequencies occurring in the lower birth weight groups, the results permit neither acceptance nor rejection of the hypothesis pertaining to the influence of birth weight disparity (intrauterine growth retardation) on tooth number variability within MZ twin pairs. Discordance in tooth number occurred in only 3 MZ pairs. In 1 set, in which a 30% difference in birth weight was observed, the heavier twin had three congenitally missing teeth and the lighter twin had six missing teeth. In the other 2 sets, in which an intrapair difference of only one missing tooth was noted, birth weight differences of 7.4% and 7.6% were observed, with the lighter twin being the less severely affected in each pair. A larger number of twin pairs with at least one member affected with hypodontia and a more complete range of birth weight disparities would be necessary to more adequately test the " retardation-hypodontia " hypothesis in MZ twins. Preliminary studies of the craniofacial complex have indicated that the magnitude of intrapair differences in third molar development (Keene, 1968a) and in maxillary dental arch width (Keene, 1968b) within MZ twin pairs may be related to the degree of birth weight disparity (growth retardation of the lighter twin) exhibited by the pairs. The less favorable prognosis for survival and for normal physical and mental development of the lighter member of a MZ twin pairs has been noted by Babson et al (1964), Fogel et al (1965), and Churchill (1965).

An interesting finding in the present study was the relatively low mean birth weight (2357 g) of twins with congenitally missing teeth other than third molars (Tab. VIII). Twins with no hypodontia had a mean birth weight of 2675 g and twins with only third molars missing had a mean birth weight of 2530 g. In a previous study (Keene, 1966), an increased frequency of low birth weight (2500 g or less) was observed in naval recruits with bilateral agenesis of mandibular third molars. Some of the men in this group had teeth other than third molars missing as well. During the selection of this hypodontia group several of the men with hypodontia (excluding third molars) were eliminated because they did not have the requisite agenesis of both mandibular third molars. The data on all men were reexam-

Tab. XIV. Hypodontia and birth weight in naval recruits

Hypodontia	N	Birth weight Mean	Birth weight SD	$P<0.01$
A. None	136	3404	550.1	A×E***
B. Third molars only	126	3402	617.7	B×E**
C. Other than third molars	49	3260	652.9	
D. Maxillary lateral incisor	26	3300	639.6	
E. Premolars	25	3073	449.0	

NOTE: Data recalculated from previous study (Keene, 1966).

** $t = 2.52$; $df = 149$; $0.01<P<0.02$
*** $t = 2.83$; $df = 159$; $0.001<P<0.01$

ined for possible lack of homogeneity in mean birth weight associated with different types of hypodontia. The results shown in Tab. XIV agree somewhat with the findings in twins and demonstrate further that individuals with congenitally missing teeth outside the third molar region (especially premolars) tend to have a lower mean birth weight than either men with no agenesis or men with agenesis of third molars alone (cf. Tab. VIII).

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Riassunto

È stato studiato, su di un campione di 262 gemelli, estratto dalla popolazione dei coscritti della marina militare a Great Lakes, il rapporto fra ipodonzia e peso alla nascita. La frequenza generale dell'ipodonzia è risultata del 34.8% includendo i terzi molari e dell'11.6% escludendoli. La distribuzione anatomica dei denti congenitamente assenti e la frequenza dell'ipodonzia dei terzi molari sono apparse in accordo con altri studi, ma l'ipodonzia di altri denti è risultata due-tre volte superiore nei gemelli che nella popolazione generale.

La frequenza di basso peso alla nascita (2500 g o meno) nel campione gemellare era del 47.7%; l'ipodonzia si verificava più frequentemente nei gemelli con basso peso alla nascita (42.6%) che in quelli con peso superiore a 2500 g (28.6%). Il peso medio alla nascita dei gemelli con ipodonzia era di 2479 g; quello dei gemelli senza ipodonzia, di 2675 g. I gemelli con denti mancanti, esclusi i terzi molari, avevano un peso medio alla nascita di 2357 g.

Il peso medio alla nascita era inferiore e la frequenza dell'ipodonzia maggiore nei gemelli MZ che in quelli DZ. Discordanze rispetto alla localizzazione anatomica ed al numero di denti mancanti furono riscontrate in diverse coppie sia DZ che MZ; nelle MZ le discordanze tendevano ad essere limitate alla localizzazione anatomica, più che al numero di denti mancanti. Sei coppie gemellari (5 MZ e 1 DZ) su 20 (13 MZ e 7 DZ) con uno o più denti mancanti presentavano concordanza perfetta sia per numero che per localizzazione. In 28 coppie nessuno dei due gemelli presentava ipodonzia.

La varianza media intracoppia del numero di denti congenitamente assenti è risultata circa dieci volte maggiore nelle coppie DZ che in quelle MZ, il che depone per una elevata componente genetica alla base della variabilità del numero dei denti.

Résumé

Le rapport entre hypodontie et poids à la naissance a été étudié sur un échantillon de 262 jumeaux, tiré de la population des conscrits de la marine militaire à Great Lakes. La fréquence générale de l'hypodontie était de 34.8% comprenant les troisièmes molaires, et de 11.6% en les excluant. La distribution anatomique des dents congénitalement absentes et la fréquence de l'hypodontie des troisièmes molaires étaient en accord avec les données d'autres études, mais l'hypodontie d'autres dents était deux-trois fois plus élevée chez les jumeaux par rapport à la population générale.

La fréquence d'un poids bas à la naissance (2500 g ou moins) dans l'échantillon géméllaire était de 47.7%; l'hypodontie était plus fréquente chez les jumeaux avec poids bas à la naissance (42.6%), par rapport à ceux avec poids supérieur à 2500 g (28.6%). Le poids moyen à la naissance des jumeaux avec hypodontie était de 2479 g; celui des jumeaux sans hypodontie, de 2675 g. Les jumeaux qui manquaient de quelques dents, excluant les troisièmes molaires présentaient un poids moyen à la naissance de 2357 g.

Le poids moyen à la naissance était inférieur, et la fréquence de l'hypodontie plus élevée, chez les jumeaux MZ par rapport aux DZ. Chez différents couples DZ et MZ des discordances ont été remarquées vis-à-vis de la position anatomique et du nombre de dents absentes; chez les couples MZ les discordances tendaient à être limitées à la position anatomique plus qu'au nombre de dents absentes. Six couples (5 MZ et 1 DZ) sur 20 (13 MZ et 7 DZ), avec une ou plusieurs dents absentes, présentaient une parfaite concordance de nombre et de position. Chez 28 couples, aucun des deux jumeaux ne présentait d'hypodontie.

La variance moyenne intra-couple du nombre de dents congénitalement absentes était dix fois plus élevée chez les couples DZ par rapport aux MZ, ce qui indique une influence génétique remarquable à la base de la variabilité du nombre des dents.

Zusammenfassung

An einem Muster von 262 aus der Bevölkerung der Marinerekruten in Great Lakes ausgewählten Zwillingen wurde das Verhältnis zwischen Hypodontie und Geburtsgewicht untersucht. Bei Einbeziehung der Weisheitszähne (dritte Molaren) betrug die Frequenz der Hypodontie allgemein 34.8%, ohne diese 11.6%. Bezüglich der anatomischen Verteilung der bei Geburt fehlenden Zähne und der Häufigkeit des Fehlens der Weisheitszähne stimmten die Ergebnisse mit anderen Forschungen überein, allerdings war die Hypodontie der anderen Zähne bei Zwillingen zwei-drei mal so häufig wie bei der allgemeinen Bevölkerung.

Das Vorkommen eines niedrigen Geburtsgewichts (2500 g oder noch weniger) betrug bei dem Zwillingsmuster 47.7%; Hypodontie wurde häufiger bei Zwillingen mit niedrigem Geburtsgewicht angetroffen (42.6%) als bei solchen mit Geburtsgewicht über 2500 g (28.6%). Das durchschnittliche Geburtsgewicht der Zwillinge mit Hypodontie betrug 2479 g, bei den anderen war es 2675 g. Die Zwillinge, denen Nicht-Weisheitszähne fehlten, hatten ein durchschnittliches Geburtsgewicht von 2357 g.

Bei EZ war das durchschnittliche Geburtsgewicht niedriger und die Hypodontie häufiger als bei ZZ. Bei mehreren sowohl EZ- als ZZ-Paaren bestand eine Diskordanz hinsichtlich der anatomischen Lokalisierung und der Zahl der fehlenden Zähne. Bei 6 (5 EZ und 1 ZZ) der insgesamt 20 (13 EZ und 7 ZZ) Zwillingspaare bestand perfekte Konkordanz sei es in Bezug auf die Zahl als auf die Lokalisierung der fehlenden Zähne. Keinerlei Zähne fehlten 28 Paaren.

Der Unterschied zwischen den jeweiligen Paarlingen war für die angeborene Hypodontie im Durchschnitt ca. zehnmal grösser bei ZZ als bei EZ, was für einen starken Erbeinfluss bei der Variabilität der Zähnezahl spricht.

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