

Tooth formation in patients with oligodontia

Yvonne Schalk van der Weide, PhD; Birte Prah Andersen, PhD;
Frederik Bosman, PhD

The literature contains no clear consensus concerning the definition of oligodontia. In general, the term refers to the congenital absence of many teeth.^{1,2} In this study, oligodontia is defined as the congenital absence of six or more teeth, excluding the third molars. This definition holds with the definition of Hobkirk and Brook³ and Whist, Thunold and Böe.⁴ Researchers in both studies defined severe hypodontia as the congenital absence of six or more teeth.

Oligodontia is reported to be associated with dental abnormalities, such as reduced size of the teeth, abnormalities of the enamel and delayed eruption of the teeth.⁵ However, the literature contains no consensus concerning the latter phenomenon.⁶ Bailit, Thomson and Niswander found no significant difference in the eruption of the teeth of patients with oligodontia and a control group.⁷ On the other hand, Rune and Särnas, evaluating tooth formation in patients with oligodontia, found

delayed tooth formation in patients with severe hypodontia.⁸ Garn and Lewis also described a delay in tooth formation in patients with third molar agenesis.⁹ Delayed tooth formation is also found in patients with multiple anomalies such as cleft palate and Down syndrome.¹⁰ Many of these syndromes also include the congenital absence of teeth.

Several methods have been proposed for assessing dental development. Simply counting the number of teeth present in the mouth is one method.¹¹ Rating the calcification stages of the teeth is another approach.¹²

Demirjian¹³ and Haavikko¹⁴ each described methods for assessing dental development. Demirjian described eight stages of mineralization and gave each one a score.¹³ The sum of scores for an individual provides an estimate of dental maturity. The sum is converted to a dental age between 3 and 16 years. Haavikko identified 12 radiographic stages of tooth formation for each tooth, from which a median age

Abstract

Oligodontia is defined as the congenital absence of many teeth. The aim of the present study was to evaluate tooth formation in patients with oligodontia who were missing six or more teeth. The oligodontia sample was compared to a control group matched for race, sex and age. Calcification stages of the teeth of the left side of the mandible were rated.

Patients with oligodontia showed a tendency for delayed tooth formation. Significant differences in tooth formation were found between males with oligodontia and the control group for several stages of tooth formation. In females only stage 5 of the second mandibular molar showed a significant difference. Because of the great individual variation in tooth formation in patients with oligodontia, tooth formation in patients with oligodontia should be individually examined before starting treatment.

Key Words

Oligodontia • Tooth formation • Calcification stages

Submitted: February 1992 Revised and accepted: June 1992

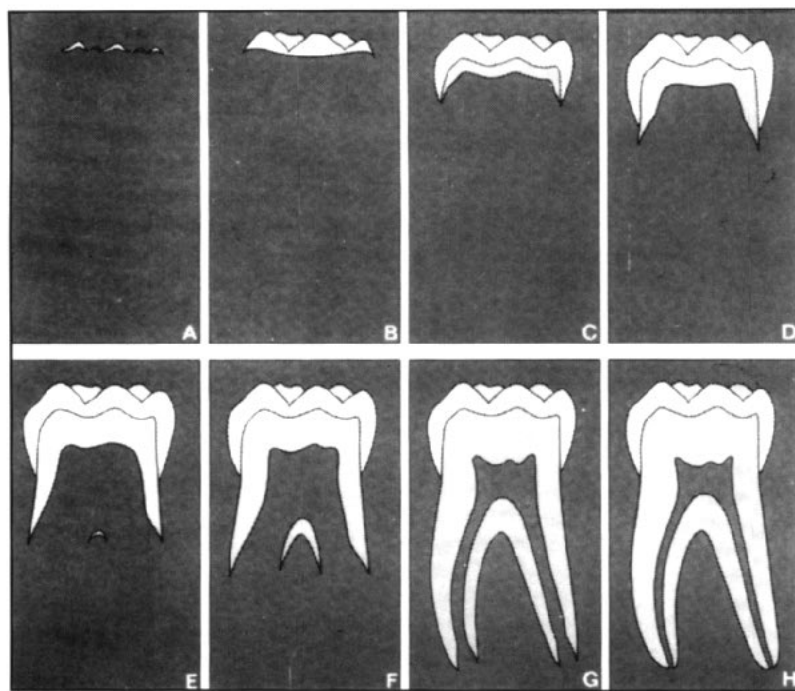


Figure 1

Figure 1
Calcification stages of a molar after the ratings developed by Demirjian.¹³ (Copied with permission from the editor.¹⁷)

for each tooth and its range can be determined. The corresponding ages for all teeth present are summed and divided by the number of missing teeth giving a mean age. Dental age can then be compared to the chronological age of the patient.¹⁴

In the timing of a patient's orthodontic treatment, it is important to pay attention to abnormalities in dental development. Moreover, the knowledge of developmental disturbances is of importance in gaining insight in the etiology of oligodontia.

In the present study the tooth formation of patients with oligodontia is evaluated. The results are compared with a control group matched for race, sex and age. The study is part of a larger study in which the etiology and symptomatology of oligodontia are evaluated.

Materials and methods

Data were collected on 216 patients with oligodontia (121 females and 95 males) from University clinics and private orthodontic practices throughout the Netherlands. The mean age of the patients was 11.3 years (range 5 to 14 years). The group contained 6 patients with ectodermal dysplasia and 210 with isolated oligodontia. The mean number of missing teeth was 10.7 for females and 11.7 for males. A control group was derived from the data of the Nijmegen Growth Study.¹⁵ The Nijmegen Growth Study is a longitudinal study in which the dental age of the children was recorded at more than one age. In the present study the dental age of patients with oligodontia was recorded at one age. Therefore, a sample from the Nijmegen

Growth Study was used in which every child occurred only once. The control group consisted of 443 persons (233 females and 210 males) who were between 4 and 14 years old, and who had full permanent dentition.

The method used in this study was published by Demirjian.¹³ He used the calcification stages of teeth as seen in panoramic radiographs to assess dental development (Figure 2).

Demirjian described eight stages of development, starting from calcification of the top of the cusp to closure of the apex.¹³ The high degree of correlation which exists between the developmental stages of the right and left sides of the maxilla and the mandible allows researchers to study only one side of the arch.¹³ Because of the clear view on a panoramic radiograph of the mandibular teeth, all mandibular teeth on the left side were selected for the study.

Boys and girls were treated separately because of the known sex differences in dental development.^{10,13}

The ratings were assigned by following criteria described by Demirjian¹³ and by comparing the radiograph with illustrations as shown in Figure 1. The ratings were never based on the illustrations alone. Dental development in this study was assessed by only one observer (YS-W).

In the Nijmegen Growth Study the dental maturity scores from children in the age of 4 to 14 years according to the method based on Demirjian¹³ were also obtained by one observer (PB-A).

Inter-observer agreement

In order to calculate the inter-observer agreement dental age was assessed on 20 panoramic radiographs by the two observer's (YS-W; BP-A). Cohen's Kappa was calculated. Cohen's Kappa is a measure of the agreement of two or more observers after the agreement due to chance is excluded.¹⁶

Statistical procedure

The differences in tooth formation between patients with oligodontia and the controls were evaluated by means of a logistic regression technique which was applied to groups separately for each stage.¹⁸

Logistic regression is a regression technique for estimating the probability that a tooth has reached a certain stage of formation, depending on the values of one or more covariates, such as age or sex. Tests are available to determine which covariables contribute significantly to the model. Age, sex and group were significant covariables. Therefore the two groups (oligodontia and Nijmegen Growth Study) were evaluated separately for both males and females.

The logistic regression models were used to calculate the median points for the various stages for all teeth. The median points represent the point where

50% of the patients had reached the stage in question. The differences between the median of patients with oligodontia and the data from the Nijmegen Growth Study were studied.¹⁹

Results

Inter-observer agreement was calculated by means of a Cohen's Kappa which resulted in a Cohen's Kappa of 0.65. The results of the logistic regression models -- the 50% or median points -- are summarized in Tables I and II. There was a tendency in males for teeth to show delayed dental development in the early stages compared to the control group. In the last three stages the delay was less evident. In males for stage 5 of the mandibular second molar, the first and second premolar and the canine and for the fourth stage of the mandibular second molar significant differences were found between the median points of patients with oligodontia and the control group. Furthermore, significant differences were found for the sixth and seventh stages of the first premolar.

In females only stage 5 of the lower second molar differed significantly between patients with oligodontia and controls (Tables I and II).

Discussion

In patients with oligodontia a tendency for teeth to show delayed tooth formation was observed. The delay was more obvious in males than in females. Unfortunately the present material did not permit an evaluation of the first three stages, because panoramic radiographs were not available. (Patients with oligodontia are often not diagnosed as having congenitally missing teeth until the age of 11 or 12 years. At that age, the patients are often referred to an orthodontic practitioner for the first time.)

The method of counting teeth was not selected as the measure for determining dental age for the following reasons: In humans considerable variation exists in time of eruption of the permanent teeth. The emergence of the teeth is also disturbed by several exogenous factors such as infection, premature extraction of a deciduous tooth and other local factors. The method based on clinical emergence is limited to certain periods, no teeth erupts from 8 to 10 years of age or after the 12th year. The disadvantages related to counting the number of teeth in the mouth to assess dental development do not play a role in using the calcification stages of the teeth as seen on panoramic radiographs for assessing dental age.

By using the method of Demirjian¹³ the results could be compared with the results of the Nijmegen Growth Study.¹⁵ A Cohen's Kappa of 0.65 means that there is substantial agreement between the observers, after the agreement due to chance is



Figure 2

Figure 2
Panoramic radiograph of a patient with oligodontia (age 11 years) as used to score the tooth formation.

excluded. The interobserver agreement was therefore considered satisfactory (Cohen's Kappa = 0.65), making a reliable comparison of the patients with oligodontia and the control group possible.

Rates of calcification of the different teeth are commonly used to calculate dental age, which is then compared to the chronological age. To compute dental age according to the method of Demirjian¹³ it is necessary to rate all four or all seven left or right mandibular teeth. In the present study it was not possible to compute the dental age because of the congenitally missing teeth. Different teeth were missing in each individual, making it impossible to calculate dental age. Moreover a difference in delay for the different types of teeth could be missed using dental age. Therefore in this study logistic regression models were constructed of all stages for each tooth. The points at which 50% of the patients with oligodontia had reached a certain stage of tooth formation were compared with the 50% points calculated from the control group. Standard errors of the 50% points were calculated.

In a study of patients with severe hypodontia (i.e. missing four or more teeth) delayed tooth formation was described by Rune and Särnas.⁸ They calculated the mean age of the individual formation stages of the teeth, according to the method of Haavikko.¹⁴ The mean delay was 2.0 years for males and 1.8 years for females. In general the delay was more severe in the early stages. Those findings of Rune and Särnas⁸ are in concordance with the findings for the males in the present study. However, although in the present study the patients were missing six or more teeth, the delay was less pronounced. Significant differences between the patients with oligodontia and the control group in the 50% points (median) of the logistic regression models were found in only a few stages. In males the fifth stage of second mandibular molar; first and second mandibular premolar and canine differed significantly as well as the fourth stage of the mandibular second molar. Furthermore the sixth and seventh stage of the mandibular first premolar were significantly different compared with the con-

Table I
The 50% points (median) $\pm$ standard error of the curves constructed with logistic regression, for both females with oligodontia (O) and the control group (N).
The 50% point represents the age at which 50% of the persons are in stage 1 through 4, 1 through 5 and so on.

Stage Tooth	1 - 4	1 - 5	1 - 6	1 - 7
37 O	8.3 $\pm$ 1.22 (12)	11.6 $\pm$ 0.51* (33)	13.0 $\pm$ 0.28 (52)	-
37 N	8.8 $\pm$ 0.18 (58)	10.4 $\pm$ 0.15 (100)	12.9 $\pm$ 0.14 (191)	-
36 O	-	-	7.8 $\pm$ 0.69 (6)	10.1 $\pm$ 0.28 (28)
36 N	-	-	8.0 $\pm$ 0.15 (43)	10.4 $\pm$ 0.15 (97)
35 O	-	9.8 $\pm$ 1.12 (8)	13.5 $\pm$ 0.60 (22)	-
35 N	-	10.1 $\pm$ 0.15 (88)	12.4 $\pm$ 0.12 (163)	-
34 O	7.9 $\pm$ 0.75 (6)	9.8 $\pm$ 0.40 (23)	12.0 $\pm$ 0.24 (51)	13.2 $\pm$ 0.26 (68)
34 N	7.7 $\pm$ 0.19 (36)	9.3 $\pm$ 0.17 (68)	11.6 $\pm$ 0.12 (141)	12.9 $\pm$ 0.17 (177)
33 O	6.8 $\pm$ 1.10 (2)	8.6 $\pm$ 0.39 (10)	10.8 $\pm$ 0.24 (42)	13.0 $\pm$ 0.38 (74)
33 N	7.0 $\pm$ 0.18 (17)	8.1 $\pm$ 0.15 (40)	11.0 $\pm$ 0.12 (108)	13.1 $\pm$ 0.21 (167)

() = Number of individuals in each category

* = (P < 0.05)

control group. In females stage 5 of the mandibular second molar showed a significant difference. In some teeth the logistic regression model also showed a significant interaction between age and group, indicating different curves of tooth formation for that tooth in patients with oligodontia and the children of the Nijmegen Growth Study. The higher standard errors of the 50% points of the patients with oligodontia compared to those of the control group (Tables I and II) could be due to the

distributions of individuals in the control group and patients with oligodontia over the stages. The range of age of the patients in one stage is comparable. However, in patients with oligodontia a tendency was found for a leveled distribution, i.e. equal numbers of individuals in the same stage of tooth formation were found in the different age groups. In the control group a tendency for a peak distribution was found, i.e. the highest number of individuals in the same stage was found at a certain

Table II
The 50 % points (median) $\pm$ standard error of the curves constructed with logistic regression, for both males with oligodontia (O) and the control group (N).
The 50 % point represent the age at which 50 % of the persons are in stage 1 through 4, 1 through 5 and so on.

Stage Tooth	1 - 4	1 - 5	1 - 6	1 - 7
37 O	10.5 $\pm$ 0.45* (19)	12.2 $\pm$ 0.26* (33)	13.9 $\pm$ 0.51 (46)	-
37 N	9.0 $\pm$ 0.20 (57)	10.8 $\pm$ 0.18 (96)	13.3 $\pm$ 0.15 (183)	-
36 O	-	-	7.9 $\pm$ 0.64 (12)	11.2 $\pm$ 0.25 (40)
36 N	-	-	8.5 $\pm$ 0.18 (49)	10.9 $\pm$ 0.16 (98)
35 O	9.8 $\pm$ 0.53 (11)	12.1 $\pm$ 0.61* (21)	-	-
35 N	9.2 $\pm$ 0.19 (57)	10.4 $\pm$ 0.18 (80)	-	-
34 O	8.3 $\pm$ 0.58 (7)	10.8 $\pm$ 0.25* (26)	13.2 $\pm$ 0.44* (52)	14.3 $\pm$ 0.44* (61)
34 N	8.5 $\pm$ 0.19 (49)	9.9 $\pm$ 0.16 (69)	12.0 $\pm$ 0.15 (132)	13.3 $\pm$ 0.19 (169)
33 O	7.3 $\pm$ 0.74 (8)	10.4 $\pm$ 0.24* (27)	12.6 $\pm$ 0.43 (52)	14.1 $\pm$ 0.61 (66)
33 N	7.8 $\pm$ 0.19 (35)	8.9 $\pm$ 0.16 (51)	11.7 $\pm$ 0.14 (116)	14.1 $\pm$ 0.25 (178)

* = $P < 0.05$

age group. In the surrounding age groups a lower number of individuals was found.

Garn and Lewis described a posterior-anterior gradient of delay in tooth-formation timing.⁹ In our study no such gradient was found. Rune and Särnas⁸ also failed to ascertain such a gradient. Several other studies report a delay of tooth formation in cleft lip/palate patients with and without hypodontia.^{20,21} In those studies the dental age of patients was compared to the chronological age.

The mean delay in tooth formation in cleft affected children varies from 0.3 to 0.7 years.²¹ Prah Andersen reported a tendency for children to show delayed tooth formation until the age of 9 years.²² However, in a later study no difference was found in females with or without clefts, although formation was delayed in boys with clefts.²³ These findings are in agreement with the findings in the present study, where tooth formation was more severely delayed in the early stages. The delay was

more obvious in males than in females.

Ranta described, in contrast with the previous findings, a significantly longer delay in the older age group.²⁰

Harris and Hullings reported the delay in tooth formation in patients with isolated cleft lip/palate to be more severe in the early formed teeth (first molar, canine) than in the later formed teeth (second and third molar).²⁶ Those results were not confirmed in our study. Harris and Hullings described an asymmetry in the development of teeth in isolated cleft lip/palate patients.²⁴ In the present study no asymmetry in tooth development was found in either patients or controls. Even the teeth which were most likely to be congenitally missing (mandibular and maxillary second premolars and maxillary lateral incisors) showed no asymmetry in tooth development. The differences in asymmetry found in development of the teeth can probably be explained by the differences in distribution of missing teeth. In the study by Harris and Hullings 35 patients had unilateral clefts.²⁴ In patients with unilateral cleft lip/palate the distribution of the maxillary missing teeth is often asymmetric.²¹ Rune and Särnas⁸ reported a marked delay of the contralaterals to missing teeth. Apparently the distribution of missing teeth was asymmetric. In the present study there was a high degree of symmetry in the distribution of missing teeth in both the mandible and maxilla. This could explain the symmetry found in development in our study in contrast to the findings of Harris and Hullings²⁴ and Rune and Särnas.⁸

Conclusions

The results of this study showed a great individual variation in tooth formation in patients with oligodontia. Some patients with oligodontia showed a severely delayed tooth formation whereas others showed normal tooth formation. Tooth formation

for individuals in the control group stayed within the normal biological variation. Therefore the tooth formation of each patient with oligodontia should be individually examined before starting treatment.

There is a delay in tooth formation in patients with oligodontia compared to the control group. This delay is more obvious in males than in females.

Acknowledgments

The authors would like to express their gratitude to Dr. Ir. J.A.J. Faber (Center of Biostatistics, Utrecht, The Netherlands) for the great help in the statistical analysis. We thank the Departments of Oral-Maxillofacial Surgery, Prosthodontics and Special Dental Care of Nijmegen and Amsterdam and the private orthodontic practitioners for submitting the panoramic radiographs of their patients with oligodontia. We thank the committee Nijmegen Growth Study for the opportunity to use the data of the Nijmegen Growth Study as a control group.

Author Address

Yvonne Schalk van der Weide
Department of Oral-Maxillofacial Surgery,
Prosthodontics and Special Dental Care
University of Utrecht
Padualaan 14, 3584 CH Utrecht
The Netherlands

Y. van der Schalk-Weide is an Assistant Professor, Department of Oral-Maxillofacial Surgery, Prosthodontics and Special Dental Care, University of Utrecht, The Netherlands.

B. Prahl-Anderson is a Professor, Academic Center of Dentistry Amsterdam, University of Amsterdam, Department of Orthodontics.

F. Bosman is Professor, Department of Oral-Maxillofacial Surgery, Prosthodontics and Special Dental Care, University of Utrecht, The Netherlands.

References

1. Stewart RE, Poole AE. The orofacial structures and their associations with congenital abnormalities. *Ped Clin North Am* 1982;29:547-585.
2. Hall RK. Congenitally missing teeth--a diagnostic feature in many syndromes of the head and neck. *J Int Ass Dent Child* 1983;14:69-75.
3. Hobkirk JA, Brook AH. The management of patients with severe hypodontia. *J Oral Rehab* 1980;7:289-298.
4. Whist PJ, Thunold K, Bøe OE. Frequency of hypodontia in relation to tooth size and dental arch width. *Acta Odont Scand* 1974;32:201-206.
5. Weide van der Y, Steen WHA, Beemer FA, Bosman F. Oligodontie Ned Tijdsch Tandheelk 1990;91:55-57.
6. Wabeke KB, Prahl-Andersen B. Oligodontie: etiologie, frequentie, verschijningsvormen, preprothetische orthodontie. *Ned Tijdsch Tandheelk* 1986;93:3-13.
7. Bailit HL, Thomson LA, Niswander JD. Dental eruption and hypodontia. *J Dent Res* 1968;47:669.
8. Rune B, Särnas KV. Tooth size and tooth formation in children with advanced hypodontia. *Angle Orthod* 1974;44:316-321.
9. Garn SM, Lewis AB. The gradient and the pattern of crown-size reduction in simple hypodontia. *Angle Orthod* 1971;40:51-58.
10. Prahl-Andersen B. Gebitsontwikkeling bij agenesieën. *Ned Tijdsch Tandheelk* 1984;91:515-518.
11. Filipson R, Hall K. Correlation between dental maturity, height development and sexual maturation in normal girls. *Ann Hum Biol* 1976;3:205-210.
12. Lewis AB, Garn SM. Relationship between tooth formation and other maturational factors. *Angle Orthod* 1960;30:70-77.
13. Demirjian JA. The Dentition. In Demirjian JA. *Human Growth*. London, Bailliere Tindall, pgs. 413-444, 1978.
14. Haavikko K. The formation and the alveolar and clinical eruption of the permanent teeth. An orthopantomographic study. *Suom Hammaslaeak Toim* 1970;66:103-170.
15. Prahl-Andersen B, Roede MJ. The measurement of skeletal and dental maturity. In: Prahl-Andersen B, Kowalski CJ, Heyendael P. *A mixed-longitudinal interdisciplinary study of growth and development*. New York, Academic Press, pgs. 497-536, 1979.
16. Landis JR, Koch GG. The measurement of observer agreement of categorical data. *Biomet* 1977;33:159-174.
17. Prahl-Andersen B, Linden van der FPGM. The estimation of dental age. *Trans Eur Orthod Soc* 1972;1:535-541.
18. Hosmer DW, Lemeshow S. *Applied logistic regression*. New York, John Wiley and Sons, 1989.
19. Finney DJ. *Statistical method in biological assay*. London, Charles Grifin and Company LTD, 3ed, pg. 82, 1978.
20. Ranta R. Associations of some variables to tooth formation in children with isolated cleft palate. *Scand J Dent Res* 1984;92:496-502.
21. Ranta R. A review of tooth formation in children with cleft lip/palate. *Am J Orthod Dentofac Orthop* 1986;90(1):11-18.
22. Prahl-Andersen B. The dental development in patients with cleft lip and palate. *Europ. Orthod. Soc. Trans. 52nd congress*, pgs. 155-161, 1976.
23. Prahl-Andersen B. Biologisches alter bei kindern mit spaltbildungen. *Stomatol DDR* 1979;29:816-822.
24. Harris EF, Hullings JG. Delayed dental development in children with isolated cleft lip and palate. *Archs Oral Biol* 1990;35(6):469-473.