Patterns of Missing Teeth in a Population of Oligodontia Patients

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Purpose: The purpose of this study was to characterize a population of oligodontia patients and identify patterns of tooth agenesis. *Materials and Methods*: A total of 116 patients with nonsyndromic oligodontia were studied, and the Tooth Agenesis Code (TAC) per quadrant was calculated. Oligodontia was defined as the congenital absence of 6 or more permanent teeth, excluding the third molars. The TAC is a unique number, consistent with a specific pattern of tooth agenesis. The authors suggest the use of an overall TAC with which the dentition throughout the mouth can be presented by a single number. Frequency analysis was used to study the prevalence of various patterns. *Results:* There was a great diversity of TACs. In the maxilla, agenesis of both premolars and the lateral incisor or the presence of only the central incisor and first molar were the most common patterns. In the mandible, agenesis of the second premolar or both premolars occurred most frequently. Conclusions: No single pattern of agenesis occurred more than twice when the full mouth was viewed. Hence, the presentation of the dentition in oligodontia is very heterogeneous. Evaluation of treatment strategies in oligodontia patients is a methodologic challenge because homogenous, comparable subgroups of patients are not available. Int J Prosthodont 2007;20:409-413.

Oligodontia is generally defined as the congenital absence of 6 or more permanent teeth, excluding the third molars.^{1,2} The prevalence of oligodontia in Caucasian populations in North America, Australia, and Europe is estimated to be 0.14%, with a higher incidence in women than men.³ Oligodontia can occur as an isolated nonsyndromic condition or as part of a syndrome, such as ectodermal dysplasia, incontinentia pigmenti, Down syndrome, and Rieger syndrome.⁴ In

the last decade, more light has been shed on the multifactorial etiology of oligodontia. Endocrine, local, environmental, and hereditary factors of congenitally missing teeth have been suggested and identified, the latter through molecular genetics. Recently, mutations in the genes MSX1 and PAX9 that encode transcription factors were demonstrated to be associated with isolated, nonsyndromic oligodontia.⁵⁻⁹

Oligodontia has a wide diversity of manifestations.¹⁰ Depending on the number and location of the missing teeth, masticatory, speech, and esthetic problems may arise. Van Wijk and Tan¹¹ recently proposed a practical procedure for assigning unique values for all possible combinations of absent teeth: the Tooth Agenesis Code (TAC), which can be used to describe patterns of missing teeth.

The aim of this study was to characterize a population of nonsyndromic oligodontia patients and use the TAC to identify patterns of tooth agenesis.

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Materials and Methods

Patients

The Utrecht Medical Center is an academic teaching hospital with a center for prosthodontics and special dental care. All patients referred to the center by general practitioners in The Netherlands between 1990 and 2006 who were classified at their first visit as having oligodontia were selected from the hospitals' database (n = 224). Oligodontia was defined as the congenital absence of 6 or more permanent teeth, excluding the third molars.1 The patients' charts were reviewed and the diagnosis of oligodontia was verified from a panoramic radiograph. When no panoramic radiograph was available (eg, data on microfilm) or when the quality of the radiograph did not allow adequate interpretation of the presence or absence of permanent teeth, the patient was discarded from the study population (n = 50). Patients were originally misclassified (eq. hypodontia or tooth extraction as opposed to congenital absence) in 36 cases. Patients with oligodontia as part of a syndrome are usually missing more teeth than patients with an isolated type of oligodontia.² Hence, patients with oligodontia as part of a syndrome were excluded from the study (n = 22). Consequently, from the 225 patients on the original list, 116 remained for data analysis (66 women, 50 men).

Permanent teeth that were hypoplastic and/or radiographically apparent but not (yet) erupted were considered as "present." Tooth determination was performed by 2 clinicians. Cases were reevaluated when there was initial disagreement as to which tooth was present or absent. Agreement was obtained through discussion. Absent teeth were registered by tooth number. The FDI tooth numbering system was used.¹²

Data Analysis

Patient and clinical information were entered into a database application, which was designed for the study and was used to obtain a uniform data set (Access 2000, Microsoft). The TAC was calculated. The procedure and rationale for the TAC was previously described by van Wijk and Tan,¹¹ so is only summarized here:

- · Each missing tooth type is assigned a specific value.
- For each quadrant, the values are summed. In this manner, a unique value per pattern of tooth agenesis is calculated: the TAC. Reversibly, from each TAC, the unique combination of missing teeth can be deducted (Table 1).

In addition, the authors of the present study constructed a new variable (TACoverall) that was used to identify similar patterns of tooth agenesis throughout the mouth among different patients. This variable is composed of the TAC of each quadrant, as follows:

 $\begin{aligned} \text{TACoverall} &= (\text{TAC first quadrant} \times 10^9) + \\ (\text{TAC second quadrant} \times 10^6) + \\ (\text{TAC third quadrant} \times 10^3) + \\ (\text{TAC fourth quadrant}) \end{aligned}$

The returned value is a unique number in which, when displayed with thousands separators, the 4 underlying TAC scores remain recognizable (ie, 123.100.038.005; TAC first quadrant = 123, TAC second quadrant = 100, TAC third quadrant = 38, and TAC fourth quadrant = 5). (For the convenience of the reader, an Excel spreadsheet [Microsoft] that facilitates swift back and forth calculations of TAC and TACoverall scores can be obtained by contacting the author.)

Statistical analyses mainly consisted of descriptive procedures. Potential differences in the number of absent teeth among male and female patients and between quadrants were tested by means of the independent samples t test and paired samples t test, respectively. A standard statistical program was used (SPSS 11.0, SPSS).

Results

Numeric Approach

A mean number of 12.5 teeth were missing in this population of oligodontia patients (range: 6 to 26). Table 2 shows the tooth agenesis by tooth type.

The lateral incisors and second premolars in both the maxilla and mandible were the most commonly absent teeth. Agenesis of the maxillary central incisors is quite rare, and to a lesser degree, congenital absence of the mandibular canines and maxillary molars was seldom seen.

No statistically significant difference was evident in the number of absent teeth among male and female patients (P=.38) or between the left and right (P=.18) or maxillary and mandibular quadrants (P=1.0).

TAC

All TACs and their prevalence are displayed per quadrant in Fig 1. In the maxilla, TAC 26 and 94 were the most common patterns of agenesis. In addition to functional problems, esthetic aspects play a prominent role in both TACs in the maxilla because of the missing lateral incisor. For the mandible, TACs 16 and 24 showed the highest prevalence.

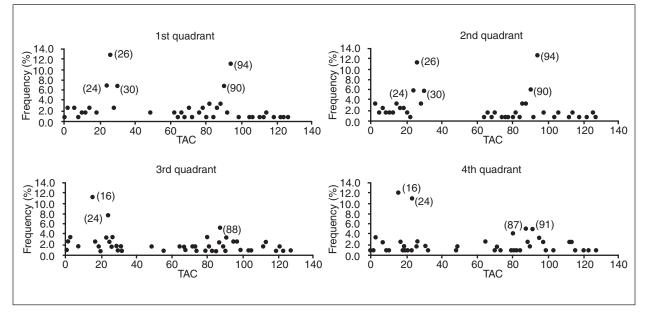


Fig 1 TAC per quadrant.

Table 1 Values for Absent	Teeth*
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Tooth	Value
Central incisor	1
Lateral incisor	2
Canine	4
First premolar	8
Second premolar	16
First molar	32
Second molar	64
Third molar [†]	128

*For example: absence of the lateral incisor, second premolar, and second molar yields the TAC value 82 (2 + 16 + 64) for that particular pattern of agenesis in any quadrant.

[†]Not included in this study.

Full-Mouth Agenesis Pattern

The various combinations of missing teeth were calculated as the TACoverall value. A table containing all observed TACoveral values is available on request. The following TACoverall values occurred twice: 24.024.088.024, 26.026.024.024, and 88.088.088.088. No single pattern occurred more than twice.

Symmetry of Agenesis

There was symmetry of agenesis between the right and left sides in the maxilla and mandible in 49.1% of the cases. Symmetry between 2 antagonistic quadrants is relatively rare: 9.5% for the right side and 4.3% for the left side. In cases of symmetry in the maxilla, the TACs 26, 94, and 24 were the most common patterns (19.3%, 12.3%, and 8.8%, respectively). TACs 26 and 24 repre-

Table 2	Prevalence of Absent Tooth Types ($n = 116$
Patients)	

Tooth no.	Frequency
11	0.9%
12	71.6%
13	45.7%
14	72.4%
15	72.4%
16	13.8%
17	52.6%
21	0.9%
22	70.7%
23	47.4%
24	67.2%
25	71.6%
26	11.2%
27	49.1%
31	56.9%
32	38.8%
33	20.7%
34	45.7%
35	74.1%
36	17.2%
37	46.6%
41	54.3%
42	43.1%
43	21.6%
44	39.7%
45	76.7%
46	16.4%
47	48.3%

sent absence of the premolars with the lateral incisor either absent or present. In TAC 94 only the first molar and central incisor are present. When there was symmetry in the mandible, TACs 16 and 24 showed the highest prevalence (17.5% and 10.5%, respectively). TAC 16 represents absence of only the second premolar.

Discussion

There are various ways to categorize numeric anomalies of teeth. A trimodal classification would be to group cases into anodontia, hypodontia, and hyperdontia, with syndromic or nonsyndromic as subclasses.¹³ According to the common definition, oligodontia is a subpopulation of both the hypo- and anodontia group in which 6 or more teeth are genetically missing, excluding the third molars.¹ In the literature, patients who suffer from oligodontia are usually characterized in terms of the number of absent teeth, not the patterns of absent teeth. However, this is not always practical, for various reasons:

- Hereditary factors play a role in oligodontia, and the scientific understanding of their significance is increasing. The more specific the cause-effect relationship, the better the evidence. Oligodontia presents in numerous clinical variations (patterns) as a result of different amounts and locations of missing teeth (see Fig 1), with tooth size variations and tooth deformities as coexisting traits.¹ To expand on the knowledge of hereditary factors, one needs to pinpoint different presentations of oligodontia (the clinical phenotype) to specific genetic defects. A useful classification of tooth agenesis takes into account both the phenotypes and genetic background. Thus, simply describing oligodontia in terms of the number of congenitally missing teeth is inadequate.
- The prosthetic rehabilitation of patients with oligodontia is likely to become more comprehensive with a higher number of absent teeth. However, from a restorative point of view, the distribution and types of missing teeth are also relevant, if not even more important. When anterior teeth are missing, esthetic features of treatment become more important. When too many adjacent teeth are missing, fixed partial dentures on natural teeth are not a viable treatment option. Thus, the mere number of absent teeth does not necessarily reflect the restorative complexity, lack of function, or esthetic consequences of each individual case.
- Treatment of oligodontia is typically multidisciplinary. A wide and expanding range of prosthetic, orthodontic, and surgical therapies are currently employed. Consequently, dental treatment for people who suffer from oligodontia can be quite expensive.¹⁴ However, studies addressing the (cost) effectiveness of different treatment strategies are lacking and are often of a retrospective nature or are simple case series or case reports.¹⁵⁻¹⁷ Treatment strategies can only be compared once the therapies under investigation are specific and well defined. In addition, the clinical situations for which they are employed need

to be more or less similar. The number of missing teeth will not suffice for this purpose.

Cluster analysis and principal component analysis to identify clusters of absent teeth in hypodontia patients were used in a previous study.¹⁸ This is a better approach than numeric classification but cannot be used to classify individual cases. Therefore, in addition to a numeric description, the authors chose the method described by van Wijk and Tan to characterize this population of oligodontia patients,¹¹ which makes use of the TAC and allows individual patterning of absent teeth per quadrant. With a minor modification, it is possible to uniquely characterize tooth agenesis throughout the mouth with a single number. These numbers, in contrast to strings, superiorly facilitate data analysis. The values in Table 2 and Fig 1 identify unique combinations of absent teeth, which are now readily available to other groups, such as for meta-analysis or genetic research. The authors encourage other groups to publish their data on oligodontia patients in a similar manner.

The data show that hypodontia is more common in women than in men, and this is in agreement with the findings of other studies.^{3,19} However, in contrast to Kirkham et al,¹⁸ no difference was found regarding the number of missing teeth among male and female oligodontia patients, perhaps because Kirkham et al included hypodontia patients as well.

Congenital absence of the central incisor, mandibular canine, and maxillary first molar is rare. A relationship has been proposed between tooth formation and innervation of the jaw. The pattern of tooth agenesis seems to follow different neural fields.^{20,21} The maxillary lateral incisor and first or second premolar were missing in 67.2% to 71.6% of all cases, without much difference in prevalence among the 3 tooth types.

TAC 26 in the maxilla, which corresponds with the agenesis of all 3 of these teeth, was the most common pattern of agenesis in the first and second quadrant (12.9% and 11.2%, respectively). In TAC 94, the other common pattern in the maxilla (right: 11.2%, left: 12.9%), only the central incisor and first molar are present. Both patterns of agenesis present functional and esthetic problems. Symmetry in the maxilla occurred in approximately half of the cases, in which left/right symmetry of TACs 26 and 94 occurred the most often (19.3% and 12.3%, respectively). Since all other TACs in the maxilla are relatively rare, therapy evaluation in oligodontia patients should be focused on treatment of TAC 26 and 94.

In the mandible, the second premolar was the most frequently absent tooth type in this group of oligodontia patients (right: 76.7%, left: 74.1%). The second premolar is the most common absent tooth in hypodontia in men.³ TAC 16, which is the agenesis of the second premolar only, was the most frequently seen pattern of agenesis in the mandible (right: 12.1%, left: 11.2%). In contrast to patterns in the maxilla, the esthetic component of this pattern plays a less prominent role. No frequently occurring patterns of missing teeth in oligodontia patients were identified.

Conclusions

The presentation of the dentition in oligodontia is very heterogeneous. Thus, the evaluation of treatment strategies in oligodontia patients will be a methodologic challenge because homogenous, comparable subgroups of patients are not available.

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Literature Abstract

Bone formation after sinus augmentation with engineered bone

The aim of the investigation was to quantify the resorption rate of tissue engineered bone grafts in the maxillary sinus using volume measurements. Sinus floor augmentation using autologous bone grafts from the iliac crest (n = 17, group 1) was compared with commercially produced transplants of human cells seeded on polyglycolid-polylactid (PLGA) scaffolds (Oral Bones) (n = 14, group 2). For bone engineering, the periosteum from the lateral aspect of the mandible was harvested to cultivate autologous human osteoblast-like cells. These cells were seeded on laboratory-developed bone chips. The total resorption rate for autologous bone had a bone density of up to 266 to 551 Hounsfield units (HU), while sufficient mineralization of tissue-engineered bone was found in only 1 case (152 HU). This clinical study concluded that the use of autologous cancellous bone grafts in sinus augmentation was more reliable than scaffolds containing cultured osteoblasts. Further tissue-engineered bone transplants should be examined to draw general conclusions about the use of tissue-engineered grafts compared with autologous bone grafts for maxillary sinus augmentation.

Zizelmann C, Schoen R, Metzger MC, et al. *Clin Oral Implants Res* 2007;18:69–73. References: 17. Reprints: Dr Christoph Zizelmann, Clinic and Policlinic for Oral and Maxillofacial Surgery, University Hospital Freiburg, Hugstetter Strasse 55, D-79106 Freiburg, Germany. Fax: +49 761 2704800. E-mail: c.zizelmann@gmx.de—Tee-Khin Neo, Singapore

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