Tooth replacements in young adults with severe hypodontia: Orthodontic space closure, dental implants, and tooth-supported fixed dental prostheses. A follow-up study

Christina L. Hvaring,[a] Bjørn Øgaard,[b] and Kari Birkeland[c]
Oslo, Norway

Introduction: Children with severe hypodontia have a substantial impairment of their dental health starting early in life. The purpose of this study was to describe types and locations of substitutes for missing teeth in patients with severe hypodontia and to compare the crown and soft tissue morphologies of orthodontic space closure, dental implants, and tooth-supported fixed dental prostheses for replacing teeth in the anterior region. **Methods:** Fifty patients missing 6 or more teeth and aged 18 years or older (mean age, 25.6 years) took part in a follow-up study. The patients were examined clinically with panoramic radiographs and clinical photographs. Crown and soft tissue variables (mucosal discoloration, crown morphology, color, and papilla index) were compared for orthodontic space closure, dental implant fixtures, and fixed dental prostheses. **Results:** Dental implants, orthodontic space closure, and retaining deciduous teeth were the most commonly prescribed treatments. Persisting deciduous teeth showed a good survival rate at the follow-up examination. Mucosal discoloration was seen only for implant fixtures and was evident for almost all fixtures in the anterior mandible and two thirds of those in the anterior maxilla. The papilla index scored poorer for both implant fixtures and fixed dental prostheses compared with orthodontic space closure. **Conclusions:** Dental implants in the anterior region proved to be an inadequate treatment modality in patients with severe hypodontia because of pronounced mucosal discoloration. (Am J Orthod Dentofacial Orthop 2016;150:620-6)

Children congenitally missing a substantial number of teeth have a severe impairment of their dental health starting early in life and face a challenging and tedious, even lifelong, treatment regimen. The clinician's mission is to provide realistic expectations of the treatment course and outcome. Patients and parents expect information on what awaits the patient in the near and distant future. The most demanding task for the clinician is to propose a sensible and feasible treatment plan with a lifelong perspective, based on biologic principles. Severe hypodontia or oligodontia, defined as the congenital absence of 6 or more teeth, is often complicated and requires an interdisciplinary approach at the specialist level, both during the initial evaluation phase and when treatment is provided. The prevalence of hypodontia in the Norwegian population has been reported to be 6.5%,[1] with severe hypodontia affecting less than 0.2% of the general Norwegian and Danish populations.[2,3]

Although a few studies have investigated hypodontia patients attending interdisciplinary clinics, the observation periods are generally limited. In addition, studies comparing the outcome of various treatment strategies in patients with severe hypodontia are yet to be performed. Hobkirk et al[4] investigated 451 patients referred to a hypodontia clinic and reported missing teeth, spacing, and poor appearance as the most common complaints. Surprisingly, 40% reported "no complaints" related to their condition. The study did not, however, address treatment planning and outcome. A Danish study by Worsaae et al[5] reported experience with a centralized interdisciplinary clinic, focusing on treatment planning and surgical treatment in 112 patients with oligodontia. At the end of the follow-up period (mean, 28 months; range, 1-68 months), 51 had finished treatment. The

From the Department of Orthodontics, Faculty of Dentistry, University of Oslo, Oslo, Norway.
[a]Research fellow.
[b]Professor.
[c]Associate professor.
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Address correspondence to: Christina L. Hvaring, Department of Orthodontics, Faculty of Dentistry, University of Oslo, PO Box, 1109 Blindern, N-0317, Oslo, Norway; e-mail, christina.hvaring@odont.uio.no.
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most common treatment options were orthodontic therapy (97%) and dental implants (90%). Shafi et al[6] collected data retrospectively from the records of 108 patients attending a hypodontia clinic over a 5-year period. Most patients had a suspected family history of hypodontia. Orthodontic therapy was most frequently proposed in treatment planning (49%), and 21% were considered for dental implants. Almost 30% did not complete treatment. Dueled et al[7] compared patients' and professionals' evaluations of implant-supported or tooth-supported reconstructions replacing congenitally missing teeth in 129 patients. The esthetic variables were considered acceptable for 92% of the implant-supported reconstructions and 83% of the tooth-supported prostheses. However, mucosal discoloration or visible metal on the buccal side was observed in 57% of the patients with dental implants. A positive correlation was observed between the professional and patient-based evaluations.

Because severe hypodontia is quite rare, opportunities for longitudinal studies with a sufficient number of patients are limited. Several studies dealing with hypodontia did not delineate patients with severe hypodontia as a separate subgroup, and most publications have been case reports or summaries of clinical experience. Consequently, evidence to support treatment decisions in complicated cases is insufficient.

The aims of this study were to compare the resulting crown and soft tissue morphologies of orthodontic space closure, dental implants, and tooth-supported fixed dental prostheses (FDPs) replacing teeth in the anterior region in patients with severe hypodontia. In addition, the treatment performed and the types and locations of substitutes for missing teeth were assessed.

MATERIAL AND METHODS

A total of 212 patients with nonsyndromic hypodontia were referred between 1998 and 2010 for an evaluation by an interdisciplinary team at the University of Oslo in Norway. The patients were admitted for clinical and radiographic examinations, and a tentative treatment plan was determined by the specialist team. Patients residing within a practical distance to the university clinic were also offered to complete their entire treatment there. The remaining patients received treatment at their local dentist or specialist clinic.

Starting in March 2013, patients missing 6 or more teeth and aged 18 years or more were contacted by mail or telephone and invited to participate in a follow-up study of treatment outcome. The rationale for the chosen age cutoff was that patients 18 years or over could be expected to have completed the majority of the treatment course. A total of 71 patients met the inclusion criteria, of whom 50 (70%) agreed to take part in the study. Of those not attending, 5 could not be reached, 7 did not wish to participate in the study, and 9 declined for practical reasons. The mean age at inclusion was 25.6 years (range, 18-38 years), and the group consisted of 24 women and 26 men. Nine of the 50 patients were unable to travel to the University of Oslo, and the examinations were performed in cooperation with an orthodontist at the patient's place of residence. Written information was given before the study, and informed consent was obtained from each participant. The patients were examined clinically, and panoramic radiographs and clinical photographs were taken.

Patient records from the time of first referral (mean age at referral, 13.9 years; range, 7-25 years) were retrieved, and the following information was noted: diagnosis, number and location of missing teeth, and recommended treatment plan.

The following parameters were recorded for all patients at the follow-up examination: number, location, and type of replacements (artificial or moved natural teeth); number and location of persisting deciduous teeth; and treatment performed. Crown and soft tissue morphology were compared for orthodontic space closure, dental implant fixtures (either single or part of an implant-retained prosthesis), and FDPs (tooth-supported conventional or resin-retained prostheses) replacing at least 1 missing tooth in the anterior region (canine to canine). This concerned 42 patients with a total of 187 teeth replaced by any of these 3 means: 97 in the maxilla and 90 in the mandible. The mean observation time from completion of treatment was 4.6 (0-11) years. The following morphologic measures were assessed objectively from standardized photographs for each replacement: mucosal discoloration, crown morphology, color of the replacement tooth, and papilla level.

Mucosal discoloration in relation to the replacement tooth was scored as follows: 0, no mucosal discoloration; 1, grayish mucosal discoloration on the buccal side; 2, visible metal on the buccal side (Fig 1). The crown morphology of the replacement tooth was evaluated as previously described by Dueled et al[7]: ie, by comparison with the contralateral tooth or, if this tooth was missing, an ideal morphology for the tooth type in question. As a supplement, expected width-to-length ratios based on published data were considered.[8] The following scoring system was used: 0 (match), the replacement tooth was optimal, with a close match to the natural or the ideal tooth; 1 (deviate), the replacement tooth deviated in at least 1 aspect from the natural or ideal tooth, but the overall impression remained acceptable; 2 (mismatch), the replacement tooth clearly deviated from the natural or ideal tooth, giving an unacceptable end result. The

Fig 1. A, Mucosal discoloration score 0, region 23; **B,** mucosal discoloration score 1, region 31; **C,** mucosal discoloration score 2, regions 31 and 42 (Fédération Dentaire Internationale tooth numbers).

color of the replacement tooth was scored according to the following system, a modification of that described by Czochrowska et al[9]: 0 (match), the replacement tooth appeared to match the shade and translucency of the contralateral tooth or, if this tooth was missing, the nearest natural neighboring tooth; 1 (deviate), the replacement tooth did not match the shade and translucency of the contralateral tooth or, if this tooth was missing, the nearest natural neighboring tooth, and the deviation was within the range of the patient's tooth shades; 2 (mismatch), the replacement tooth did not match the shade and translucency of the contralateral tooth or, if this tooth was missing, the nearest natural neighboring tooth, and the mismatch was outside the range of the patient's tooth shades and translucency. The papilla index was modified from the papilla index by Jemt[10] and scored as follows: 0, a papilla corresponding to the original papilla, filling up the entire proximal space; 1, at least half of the height of the papilla was present; 2, less than half of the height of the papilla is present. For each replacement tooth, the papillae on both sides were scored, and the average of these scores was used for the replacement tooth in question.

All measurements were recorded by the same examiner (C.L.H.) after calibration. To evaluate measurement error, 50 replacement teeth were chosen at random and remeasured for each of the 4 morphologic measures, 6 weeks after the first measurement. Cohen kappa values were all above 0.7, indicating low measurement error.

Statistical analysis

Replacements in the maxilla and the mandible were analyzed separately. The replacements were classified into 3 categories: orthodontic space closures, dental implant fixtures, and FDPs. Analyzing the data with the individual tooth replacement as the unit of analysis would not be appropriate, since data from teeth belonging to the same patient are not independent. Instead, the patient was chosen as the unit of analysis; ie, measurements were aggregated by patient and type of replacement. For patients with more than 1 replacement of the same type, the mean of the morphologic score values was calculated and rounded to the nearest integer. When comparing 2 treatments in 2 separate patient groups, we used the chi-square test. When 1 patient had replacements from both categories being compared, we chose only the replacement type used more rarely, ensuring that the compared patient groups did not overlap.

A significance level of 5% was used, and the statistical analyses were carried out using SPSS software (version 22.0; IBM, Armonk, NY).

RESULTS

In total, 571 teeth were congenitally missing among the 50 participants. The mean number of missing teeth per subject was 11.4 (range, 6-23).

The total number of sites with substitutes for a missing tooth was 488 distributed as follows: 216 with implant-supported prostheses (fixture or pontic), 45 with FDPs, 114 with a persisting deciduous tooth, 111 closed orthodontically or spontaneously, and 2 with autotransplantations (Fig 2). In addition, 113 gaps remained without a tooth or replacement. In some cases, multiple interventions were necessary to replace 1 missing tooth: eg, orthodontically moving teeth into a favorable position followed by dental implant insertion.

Classifications of original treatment plans obtained from patient records are shown in Table I. At the follow-up examination, an assessment was made of whether the treatment had been carried out as planned. This was the case for most interventions, except for dental implants, which had not yet been performed in 13 patients.

When we compared implant fixtures with orthodontic space closure (Table II), implant fixtures scored poorer for mucosal discoloration and the papilla index, and the differences were statistically significant. All implant fixtures in the anterior mandible and 60% in the anterior maxilla had either mucosal discoloration or visible metal.

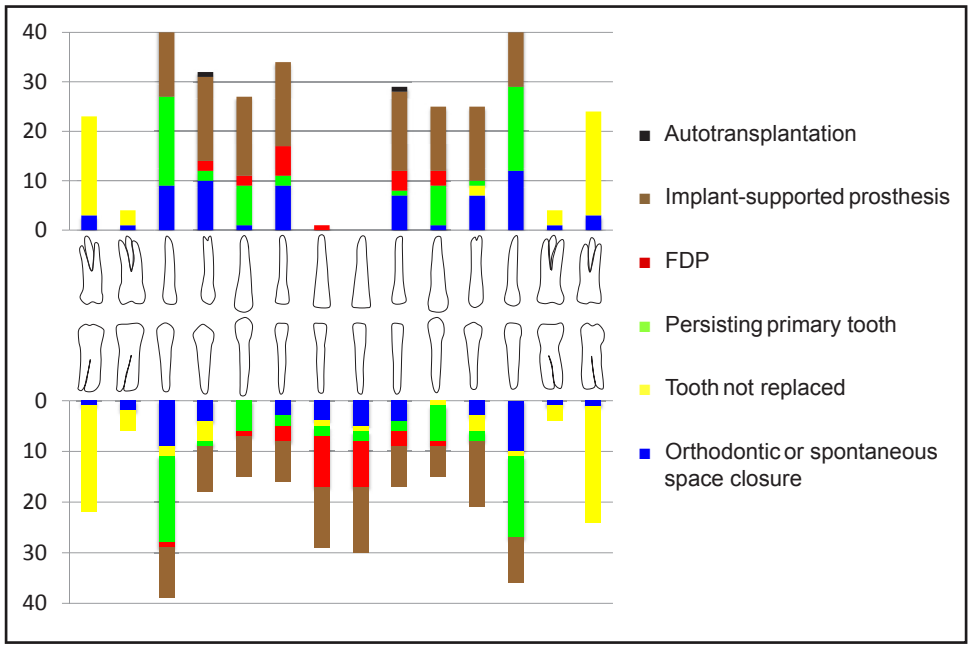

Fig 2. Distribution of substitutes for missing teeth according to location.

Table I. Number of patients in different treatment categories according to the original plan; several interventions could be recommended for each patient

Treatment category	*Treatment planned (n = 50, % of patients)*	*Not completed as planned (n)*
Orthodontic appliance therapy	46 (92%)	3
Implant-supported prosthesis	48 (96%)	13
Retain deciduous tooth	30 (60%)	2
Composite restoration	15 (30%)	-
Tooth-supported FDP	18 (36%)	3
Veneer restoration	10 (20%)	2
Orthognathic surgery	5 (10%)	2
Autotransplantation	1 (2%)	-

Likewise, when implant fixtures were compared with FDPs (Table III), a significantly more pronounced mucosal discoloration was recorded for the fixtures in both jaws. The crown morphology in the anterior mandible was significantly better for the FDPs than the implant fixtures, but the color score was better with an implant fixture than an FDP in the anterior maxilla. For both dental fixtures and FDPs, poor papilla index scores were observed. When we compared space closure with FDPs (Table IV), a significantly better papilla index was observed for space closure in both jaws. The crown morphology in the anterior mandible was considered significantly better for the FDPs.

Table II. Morphology of replacements: dental implant fixtures vs orthodontic space closure and comparisons with the chi-square test

	Anterior maxilla		*Anterior mandible*	
	Implant fixture n (%)	*Orthodontic space closure n (%)*	*Implant fixture* n (%)*	*Orthodontic space closure n (%)*
Mucosa				
0	8 (40%)	11 (100%)	-	7 (100%)
1	7 (35%)	-	3 (27%)	-
2	5 (25%)	-	8 (73%)	-
	P <0.01		*P* <0.01	
Crown				
0	5 (25%)	4 (36%)	-	1 (14.5%)
1	12 (60%)	6 (55%)	11 (92%)	5 (71%)
2	3 (15%)	1 (9%)	1 (8%)	1 (14.5%)
	NS		NS	
Color				
0	15 (75%)	8 (73%)	7 (58.3%)	6 (86%)
1	5 (25%)	3 (27%)	4 (33.3%)	1 (14%)
2	-	-	1 (8.3%)	-
	NS		NS	
Papilla				
0	7 (35%)	10 (91%)	-	6 (86%)
1	8 (40%)	1 (9%)	6 (50%)	1 (14%)
2	5 (25%)	-	6 (50%)	-
	P <0.01		*P* <0.01	

NS, Not significant.
*For 1 implant fixture, the gingival margin was not visible on the photographs; this patient was not scored for mucosal discoloration.

Table III. Morphology of replacements: dental implant fixtures vs FDP and comparisons with the chi-square test

	Anterior maxilla		*Anterior mandible*	
	Implant fixture n (%)	*FDP n (%)*	*Implant fixture* n (%)*	*FDP n (%)*
Mucosa				
0	9 (36%)	7 (100%)	1 (7%)	8 (100%)
1	11 (44%)	-	5 (33%)	-
2	5 (20%)	-	9 (60%)	-
	$P < 0.05$		$P < 0.01$	
Crown				
0	6 (24%)	2 (28.5%)	1 (6%)	6 (75%)
1	15 (60%)	3 (43%)	14 (88%)	1 (12.5%)
2	4 (16%)	2 (28.5%)	1 (6%)	1 (12.5%)
	NS		$P < 0.01$	
Color				
0	20 (80%)	2 (29%)	9 (56%)	4 (50%)
1	5 (20%)	4 (57%)	6 (38%)	3 (38%)
2	-	1 (14%)	1 (6%)	1 (12%)
	$P < 0.05$		NS	
Papilla				
0	8 (32%)	2 (29%)	1 (6%)	-
1	12 (48%)	5 (71%)	9 (56%)	7 (88%)
2	5 (20%)	-	6 (38%)	1 (12%)
	NS		NS	

NS, Not significant.
*For 1 implant fixture, the gingival margin was not visible on the photographs and this patient was not scored for mucosal discoloration.

Table IV. Morphology of replacements: orthodontic space closure vs FDP and comparisons with the chi-square test

	Anterior maxilla		*Anterior mandible*	
	Orthodontic space closure n (%)	*FDP n (%)*	*Orthodontic space closure n (%)*	*FDP n (%)*
Mucosa				
0	11 (100%)	7 (100%)	6 (100%)	8 (100%)
1	-	-	-	-
2	-	-	-	-
	NS		NS	
Crown				
0	4 (36%)	2 (28.5%)	1 (17%)	6 (75%)
1	6 (55%)	3 (43%)	5 (83%)	1 (12.5%)
2	1 (9%)	2 (28.5%)	-	1 (12.5%)
	NS		$P < 0.05$	
Color				
0	8 (73%)	2 (29%)	5 (83%)	4 (50%)
1	3 (27%)	4 (57%)	1 (17%)	3 (37.5%)
2	-	1 (14%)	-	1 (12.5%)
	NS		NS	
Papilla				
0	10 (91%)	2 (29%)	5 (83%)	-
1	1 (9%)	5 (71%)	1 (17%)	7 (87.5%)
2	-	-	-	1 (12.5%)
	$P < 0.01$		$P < 0.01$	

NS, Not significant.

DISCUSSION

In this clinical study, we found dental implants in the anterior region to be an inadequate treatment modality in patients with severe hypodontia because visible metal or grayish discoloration was evident in almost all patients. This is consistent with findings by Dueled et al,[7] who reported such discoloration in almost 60% of the studied hypodontia patients. Consequently, other therapeutic approaches such as orthodontic space closure, FDPs, or, when possible, preservation of deciduous teeth, should be considered for this patient group. Figures 3 and 4 show examples of patients treated with dental implants and orthodontic space closure, respectively. The studied patients with dental implants replacing missing teeth in the anterior mandible all had large reconstructions, with at least 2 and up to 4 fixtures. This lack of several adjacent teeth results in a low amount of alveolar bone, leading to a biologic limitation of the success of dental implant insertion in this region. A minimum bone support of 2 mm is required surrounding the implant, and preferably 3 to 4 mm mesiodistally is needed to secure an adequate blood supply, reducing the risk of bone resorption.[11-14] Because the scope of this study was limited to morphologic variables, no biologic measures of the connective tissue or bone level surrounding the artificial or moved teeth were carried out either clinically or radiographically. However, intraoral radiographs, as used in similar studies, will show only mesial or distal bone loss, whereas buccal bone destruction is not evident.[7] Future studies with 3-dimensional technology such as cone-beam computed tomography would provide interesting follow-up data about bone level surrounding dental implants in this population group of young, healthy adults with a low prevalence of periodontitis.

A direct relationship between the buccal bone level and marginal soft tissue recession has previously been demonstrated.[15] The visible metal or grayish discoloration observed in this study therefore indicates a considerable loss of buccal alveolar bone. In addition, these patients are prone to further breakdown of connective tissue because the unattached gingiva does not provide a protective barrier around the fixture. The buccal alveolar plate is more susceptible to resorption because its endosteal origin leads to a more porous structure.[16,17] Animal studies have also shown that the bone formed in the buccal areas of the mandible is particularly prone to resorption with aging.[18,19] With this basic knowledge, the buccal bone resorption already present in these patients with a mean age of 25.6 years can be expected to progress, foretelling an uncertain long-term prognosis.

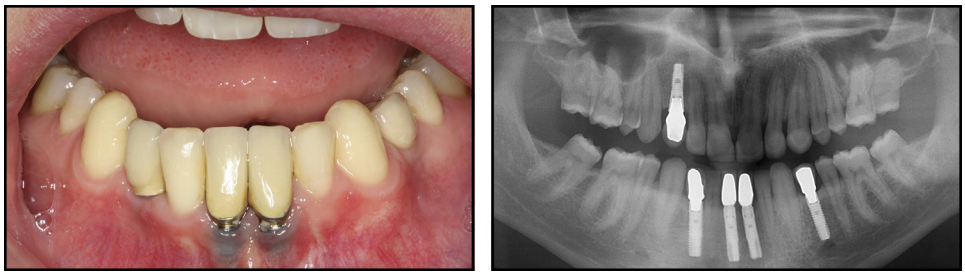

Fig 3. Man aged 24 years with agenesis of teeth 17, 14, 27, 37, 34, 31, 41, 44, and 47 (Fédération Dentaire Internationale tooth numbers). Dental implants were placed in the anterior mandible. Note the extensive mucosal discoloration and visible metal.

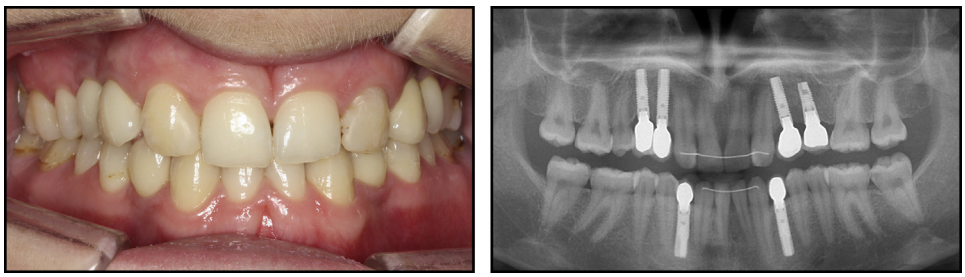

Fig 4. Woman aged 23 years with agenesis of teeth 14, 12, 22, 24, 25, 31, and 41 (Fédération Dentaire Internationale tooth numbers). She was treated with orthodontic space closure in the anterior maxilla and mandible, with dental implants placed posteriorly.

Both FDPs and implant fixtures scored significantly poorer for the papilla index compared with orthodontic space closure. Edentulous spans will lead to lack of alveolar bone and corresponding connective tissue, with a risk of future biologic complications with the final restoration.[20] Accordingly, when possible, even distribution of the existing teeth should be pursued to better maintain the alveolar process and the esthetics of the soft tissues.

The crown morphology of the FDPs scored significantly better than did orthodontic space closure in the anterior mandible. In most instances, orthodontic treatment involved moving the canines mesially to replace missing incisors. In no patient had these moved canines been subject to crown reshaping, which was probably avoided to prevent a further reduction of much-needed tooth substance. When FDPs are chosen, the technician has greater flexibility to design a more anatomically correct replacement.

A customized treatment plan consisting of at least 1 intervention was provided for each patient, and most of them were recommended to receive orthodontic therapy or dental implants (Table I), consistent with the treatment frequencies reported by Worsaae et al[5] and Shafi et al.[6] At the follow-up examination, 13 of 48 patients scheduled for dental implants had not yet had the procedure. The reason for this might be that deciduous teeth were still functioning or that implant insertion was contraindicated because of biologic limitations. Almost half of the missing teeth were substituted by a persisting deciduous tooth or by orthodontic space closure (Fig 2). Missing teeth that were not replaced were mainly located far distally, causing the least esthetic and functional impact.

Among our 50 patients, 30 were recommended to preserve at least 1 healthy deciduous tooth at the planning stage (Table I). Our findings, with a 12-year mean follow-up time from the initial examination at a mean age of 13.9 years, show that 28 patients still had at least 1 primary tooth functioning. This must be regarded as a good survival rate. These observations support previously published data stating that retaining healthy deciduous mandibular molars is a viable treatment alternative.[21-26] The patients in this study, lacking a substantial number of teeth, were recommended to preserve not only their deciduous mandibular molars, but also, when appropriate, essentially all healthy deciduous teeth. These longitudinal data are valuable for future treatment decisions in patients with severe hypodontia.

This study is the first to compare the outcome of different treatment strategies in a group of patients with severe hypodontia, and the findings have the potential to influence clinical practice. Considering the relatively unsatisfactory outcome of dental implants, the question arises whether these patients were suited for implant therapy from the outset. Ideally, a bone volume measurement

should have been performed before implant therapy was considered. This would also have permitted verifiable longitudinal data to be collected. However, a number of the treatment plans were established several years ago, before cone-beam computed tomography was readily available. All patients underwent a thorough examination, with treatment decisions made by an interdisciplinary team of experienced specialists. Therefore, one must assume that the patients who received dental implants were reasonably suited for this procedure.

A weakness of the study was the relatively few participants, and to conduct the statistical comparisons independently, the patients were required to be divided into even smaller groups. Gathering a large longitudinal sample of patients with severe hypodontia is challenging because of logistics and the rarity of the condition. However, the differences that were discovered were highly significant. The participation rate of 70% must be considered acceptable in light of the relatively long follow-up period, the high propensity of young adults to relocate geographically, and patient demotivation after several years of intensive dental treatment.

CONCLUSIONS

Dental implants in the anterior region proved to be an inadequate treatment modality in patients with severe hypodontia, with visible metal the most notable issue. Mucosal discoloration was evident in the anterior mandible in almost all patients and, in the anterior maxilla, in two thirds of the patients. Orthodontic space closure resulted in superior papilla esthetics but had the drawback of a less optimal crown morphology in the anterior mandible. Dental implants, orthodontic space closure, and retaining deciduous teeth were the most commonly prescribed treatments. Persisting deciduous teeth showed a good survival rate at the follow-up examination.

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