A Retrospective 3- to 5-Year Study of the Reconstruction of Oral Function Using Implant-Supported Prostheses in Patients With Hypohidrotic Ectodermal Dysplasia

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The aim of this study was to evaluate oral function rehabilitation in patients with hypohidrotic ectodermal dysplasia (HED) using implant-supported prostheses based on bone augmentation. From September 2005 and March 2009, 25 HED patients were chosen for clinical data analysis in this study. The criteria for patient selection included the following: the display of clinical features of HED, the number of congenitally missing teeth (>5), the patient age (>16 years), the patient's willingness, and the patient's tolerance for bone graft surgery and implant placement. Follow-up evaluations were initiated from the time of implant prosthetic placement and scheduled annually for 3–5 years. The effects of oral function reconstruction were assessed based on the cumulative survival and success rates of implants, the health of the peri-implant area, and the degree of patient satisfaction. Twenty-five HED patients received 169 conventional implants and 10 zygomatic implants (179 total implants). During 3–5 years of post-loading evaluations, 5 of the 179 implants failed and 3 implants were removed. The 3-year success and cumulative survival rates were 97.2% and 98.3%, respectively. Furthermore, periodontal probing and radiographic assessments showed that the 3-year incidence of peri-implantitis was 4.5%. Finally, HED patients expressed high degrees of satisfaction with their facial contours, masticatory function, pronunciation ability, and comfort with the implant-supported prostheses. The results of this 3- to 5-year retrospective study indicate that the oral function of HED patients can be effectively reconstructed using bone augmentation and implant-supported prostheses; however, longer term results are warranted in the future.

Key Words: hypohidrotic ectodermal dysplasia, hypodontia, dental implant, oral function construction, implant-supported prosthesis, peri-implantitis

INTRODUCTION

Ectodermal dysplasia (ED) is an inherited disorder that is classified into 2 major types: hypohidrotic ectodermal dysplasia (HED) and hidrotic ectodermal dysplasia.[1,2] HED is one of the most common types of genetic ectodermal dysplasia disorders, and an incidence of 0.63 per 10 000 subjects has been reported.[3]

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The typical clinical characteristics of HED patients include sparse hair, a juga forehead and chin, "saddle" nose, thick and protruding lips, missing or misshaped teeth, "bat ears," and wrinkling and pigment sedimentation around the "ear" or "orbital lobe of the ear."[4] Oral findings are often significant and may include multiple tooth abnormalities, particularly anodontia and hypodontia. The congenital absence of teeth results in bone atrophy, therefore placement of endosseous implants in the bone for subsequent restoration is difficult, particularly in patients with anodontia. Bone augmentation is usually necessary via bone grafting or distraction osteogenesis.[1] Previous clinical reports have described the use of dental implants to replace congenitally missing teeth in patients with ED;[5–9] however, there have been few prospective studies on the effects of reconstructed oral function in HED patients of different ages who have missing teeth.

HED often has harmful physiological and psychological effects on patients, but; procedures that include bone augmentation (eg, bone grafting or distraction osteogenesis), implant placement, and prostheses can effectively reconstruct oral function in HED patients. In recent years, there is increasing demand for reconstruction of oral function in HED patients. The cost of such reconstruction is an important consideration for many patients. Moreover, the effective reconstruction of oral function in HED patients remains a challenge for dentists because of the aforementioned insufficiency in jawbone height and width. In the present prospective study, we restored oral function in HED patients according to defined standards and with follow-up evaluations over 3–5 years. The purpose of this study was to assess the effects of such reconstruction in young HED patients.

Materials and Methods

Patient selection

From September 2005 and March 2009, 25 patients with HED were treated at the Department of Oral and Maxillofacial Surgery and the Department of Oral and Craniomaxillofacial Implantology, Ninth People's Hospital Affiliated with Shanghai Jiao Tong University, School of Medicine, Shanghai, China. According to the following criteria, the retrospective data in the present investigation were collected. The criteria for inclusion in the study were (1) patients with a diagnosis of HED, (multiple organ abnormalities, for example, hair, sweat glands, nails, and skin) who were a minimum of 5 congenitally missing permanent teeth; (2) patients over 16 years old who already decided to use implants to reconstruct oral function; and (3) patients who were physically able to tolerate conventional surgical and restorative procedures. Patients who met the above criteria and provided written informed consent were assigned a unique study identification number that provided information on implant sites and test group.

Surgical procedures

Following a comprehensive examination and a detailed treatment plan discussion with the patient and his or her family, the surgical procedures were completed according to the principles of minimal trauma and predictable outcomes. In cases of severe bone atrophy, the first step was bone augmentation using 1 of 3 methods. One treatment option included onlay grafting to horizontally and vertically augment the alveolar ridges in the anterior tooth area with harvesting of autogenous bone blocks from the mandibular oblique ridge, iliac crest, fibula, and calvaria. Alternatively, another option comprised vertical distraction osteogenesis (DO) in the area between the mental foramina of the mandible. As a third option, an artificial bone material (Bio-Oss, Geistlich Bio-Oss, GeistlichPharma AG, Bahnhofstrasse, Switzerland) was used to assist the implants placed in the patients if the existing bone volume was insufficient. Three to 6 months after the completion of bone augmentation, radiographic examination was performed to ensure that the bone volume was sufficient for implant placement. Among the 179 total implants placed in the jaws, 94 were placed in the maxilla and 85 in the mandible. Two implant systems were used to restore oral function: (1) Nobel Biocare (Gothenburg, Sweden) and (2) Institute Straumann AG (Basel, Switzerland). Both conventional implants (CIs) and zygomatic implants (ZIs) were used. CIs were 3.3 to 4.8 mm in diameter and 8 to 12 mm in length for the Straumann system, and 3.5 to 5.0 mm in diameter and 10 to 13 mm in length for the Nobel Biocare system (replace system). ZIs were 4.0 mm in diameter and 40 to 52.5 mm in length for the Nobel Biocare system (Branemark system). Decisions regarding implant length and diameter were based

Table 1

Clinical feature of patients with hypohidrotic ectodermal dysplasia (HED)*

Characteristics	Patients (n = 25)
Age (year)	17–28
Gender (M/F)	13/12
Implants (n)	ZIs = 10; CIs = 169
Length	ZIs = 40–52.5 mm; ITI = 8–10 mm; Replace = 10–13 mm
Diameter	3.3–4.8 mm; 3.5–5.0 mm; 4.0 mm
Bone augmentation in mandibule	DO = 2; fibular graft = 2; GBR = 7
Bone augmentation in maxilla	Iliac graft = 5; fibular graft = 1; GBR = 11
Mean reconstruction period in years (SD)	0.8 (0.5)
The type of restoration	Implant-supported fixed dentures = 24; implant-supported overdenture = 1 patient

*F indicates female; M, male; ZIs, zygomatic implants; CIs, conventional implants; DO, distraction osteogenesis.

on preoperative radiographs and clinical findings at the time of surgical placement. Panoramic radiographs and computerized tomography scans were used to evaluate the mandible and maxilla before surgery. For patients with anodontia, 6 implants (2 ZIs and 4 CIs) were placed in the upper jaw, whereas 4 or 2 CIs were placed in the anterior area of the mandible. For partially edentulous patients, CIs were placed in the toothless portion of the alveolar crest. Clinical and radiographic examinations were used to evaluate the effects of the implants, including location, direction, and safety.

Prosthetic procedures

Three to 6 months after implant placement, prosthetic procedures were implemented. For complex cases involving severe hypodontia and anodontia, the transitional denture was completed with acrylic resin. After 6 months, the final prosthetic procedures were performed using CAD/CAM technology as previously reported.[10] Open-tray impression transfer copings were made using polyether elastomeric impression material for both maxillary and mandibular arches. Master casts were poured and a wax trial was performed for clinical evaluation and patient approval, after which resin patterns were fabricated using the diagnostic denture wax-up as a guide. Adequate cutback was then applied to the resin framework to accommodate conventional full-coverage crown restorations in both the maxillary and mandibular arches. The resin pattern frameworks were scanned using a Procera Forte scanner (Nobel Biocare, Benelux BV, Houten, Utrecht, the Netherlands), were milled from solid pieces of titanium, and were manually finished before clinical evaluation. A framework try-in was performed to check for passivity of fit. Once the fit was found satisfactory, the frameworks were veneered with gingival pink porcelain to resemble the soft tissue in the gingival areas. Ceramic crowns were completed and a screw-retained framework was then secured to the implants at 35 Ncm torque. The screw access holes were sealed with gutta-percha before composite fillings were placed on top of the framework. The crowns were individually cemented over the framework using provisional cement in a conventional

Table 2

Cumulative survival and success rates of implants*

Interval	Implants at Start of Interval	Drop-Out Implants	Failing Implants	Removed Implants	Cumulative Survival Rate, %	Cumulative Success Rate, %
Placement to loading	179	0	0	3 CIs	98.3	98.3
Loading	178	0	1 CIs	0	98.3	97.2
To 1 year	171	5 CIs	2 CIs	1	98.3	97.2
1–2 years	171	0	0	0	98.3	97.2
2–3 years	159	12 CIs	0 CIs	0	98.3	97.2
3–4 years	118	0	0 CIs	0	98.3	97.2
4–5 years	62	0	2 CIs	2	98.3	97.2

*Failing implants: implants with bone resorption >1.5 mm after the first year of loading and >0.2 mm in the following years but fulfilling the other criteria of Albrektsson et al.[12] CI indicates conventional implants.

TABLE 3

Evaluation of sulcus bleeding index (SBI) for implants in 25 patients*

Measurement Time	SBI	Implants, %
Baseline–6 months	0	84.4
	1	13.4
	2	2.2
	3	0
Baseline–1 year	0	84.4
	1	12.1
	2	2.3
	3	1.2
Baseline–2 years	0	84.2
	1	13.5
	2	2.3
	3	0
Baseline–3 years	0	84.3
	1	13.2
	2	2.5
	3	0
Baseline–4 years	0	83.9
	1	13.6
	2	2.5
	3	0
Baseline–5 years	0	82.8
	1	12.5
	2	3.1
	3	1.6

*0 indicates no bleeding; 1, an isolated bleeding spot was visible; 2, blood formed a confluent red line on the mucosal margin; 3, heavy or profuse bleeding.

TABLE 4

Evaluation of probing depth (PD) for implants in 25 patients*

Measurement Time	PD (Changes From BL), mm	Implants, n
Baseline–6 months	0–1	177
	1.5–3	1
	3.5–5	1
	≥5	0
Baseline–1 year	0–1	170
	1.5–3	2
	3.5–5	0
	≥5	1
Baseline–2 years	0–1	169
	1.5–3	2
	3.5-5	0
	≥5	0
Baseline–3 years	0–1	157
	1.5–3	2
	3.5–5	0
	≥5	0
Baseline–4 years	0–1	115
	1.5–3	1
	3.5–5	2
	≥5	0
Baseline–5 years	0–1	61
	1.5–3	1
	3.5–5	0
	≥5	2

*n indicates number of sites where there was a finding of increased probing depth corresponding to intervals. Probing depth intervals (millimeters) are changes from baseline (BL) to the 5-year permanent prosthesis insertion.

manner. For other types of general hypodontia (the deficient number of teeth <5), prosthetic procedures were completed using routine technology for implant-supported dentures. After the prostheses were completed, the patients were placed on a standard oral hygiene protocol using chlorhexidine rinse and periodontal maintenance of periodic 3-month recall visits.

Evaluation criteria

Following the completion of patient prostheses, a series of follow-up visits were performed over 3–5 years, where three categories of data were recorded. Category 1 included monitoring of infection: (a) sulcus bleeding index (SBI) (assessing the bleeding tendency of the marginal peri-implant tissues at the mesial, distal, buccal, and lingual aspects of each implant); (b) probing depths (PD) (of the mesial, distal, buccal, and lingual surfaces); (c) suppuration (yes or no during the probing procedure); (d) measures of overall oral hygiene conditions (plaque index, gingival index); and (e) mobility testing (removing the prosthesis, attaching a post, and applying an opposing force using 2 hand instruments (eg, mirrors). Category 2 included the following radiographic analyses: (a) periapical radiographs; (b) scanning radiographic films and marking crestal bone landmarks; (c) evaluator scoring of 2 marks designating both the mesial and distal aspects of the implant (the apical and coronal intersection of the crestal bone with the implant body); (d) bone height measurements; and (e) scoring of the crestal bone height (the distance between the crestal bone-implant intersection point and the reference line). Statistical significance was tested using analysis of variance. Category 3 included reporting of peri-implantitis: (a) mucositis with a positive finding of bleeding and/or suppuration upon probing; (b) probing depth; (c) crestal bone loss that was progressive, and confirmed by radiography; and (d) patient satisfaction. Patient satisfaction covered (1) the esthetic aspect of the facial contour, (2) the

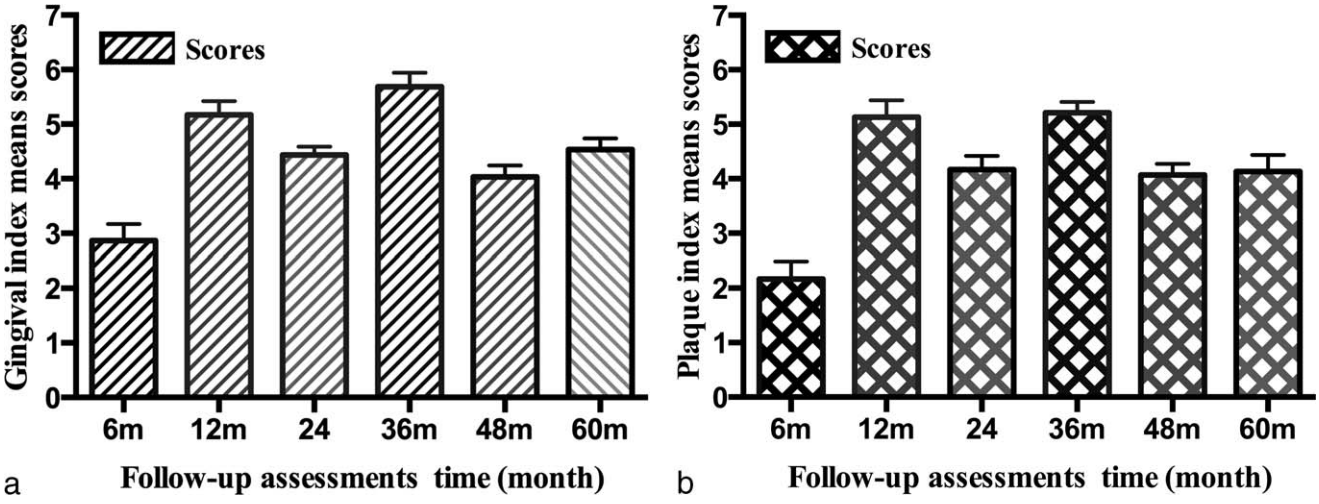


Figure 1. The mean scores for the gingival and plaque indices through 3–5 years of follow-up assessment were <1.0. (a) Gingival index scores: 0 = no inflammation; 1 = mild inflammation, slight change in color, and little change in texture; 2 = moderate inflammation, moderate glazing, redness, edema hypertrophy, and bleeding on probing; and 3 = severe inflammation, marked redness and hypertrophy, tendency to spontaneously bleed, and ulceration. (b) Plaque index scores: 0 = no plaque; 1 = plaque was not visible to the unaided eye, but when the tip of a probe was run across the gingival margin, a thin film of plaque was observed; 2 = moderate accumulation of soft deposits at the gingival margin and/or on the tooth surface visible to the naked eye; and 3 = abundance of soft matter at the gingival margin, in the gingival pocket, and on the surface of the tooth. BL indicates baseline (implant placement).

function of the prosthesis, (3) the comfort level of the prosthesis, and (4) pronunciation ability. As previously described, each score was reported using a scale of 0–2 points.[11]

For statistical analysis, the software program SPSS 10.0 (SPSS Science, Chicago, Ill) was used. Because of the descriptive character of the study, no adjusting of *P* values for multiple testing was performed.

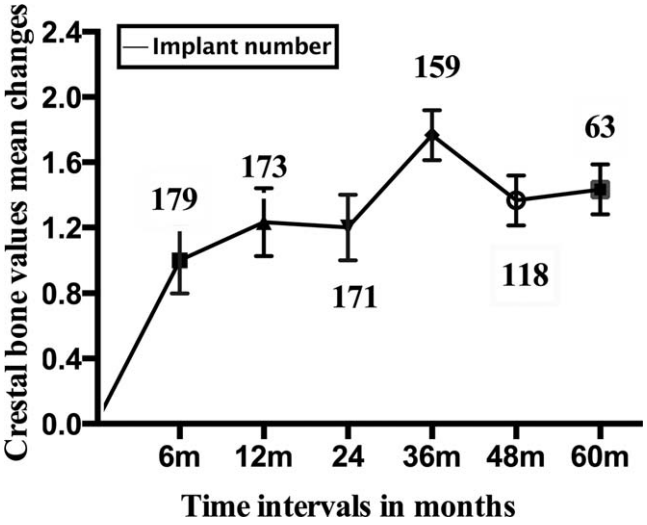


Figure 2. The crestal bone values were the mean changes measured from baseline (in mm) through 3–5 years of follow-up evaluation of implants. Error bars indicate SE values. The value at each error bar indicates the number of implants evaluated at that interval.

Results

Twenty-five HED patients, ranging in age from 17 to 28 years, were treated with implant-supported prostheses between September 2005 and March 2009. Before the implants were placed, bone augmentation was performed using autogeneic bone grafts (from the mandibular oblique ridge, ilium, fibula, or calvaria), DO, or artificial bone (Bio-Oss). Five patients were treated by autogeneic bone graft from the ilium to the maxilla. Three patients received bone grafts from the mandibular oblique ridge to the maxilla and mandible. The bone volume was augmented using bone grafts from the calvaria to the mandible in 1 patient, and from the fibula to the mandible and maxilla in 2 patients. Two patients were treated by DO in the mandible. Local bone augmentation was completed in other patients using Bio-Oss and Bio-Gide. Bone augmentation was successful in all patients, although partial loss of the graft was observed in those patients who received iliac grafts.

One hundred seventy-nine implants were placed in support of prostheses, of which 10 implants were

TABLE 5

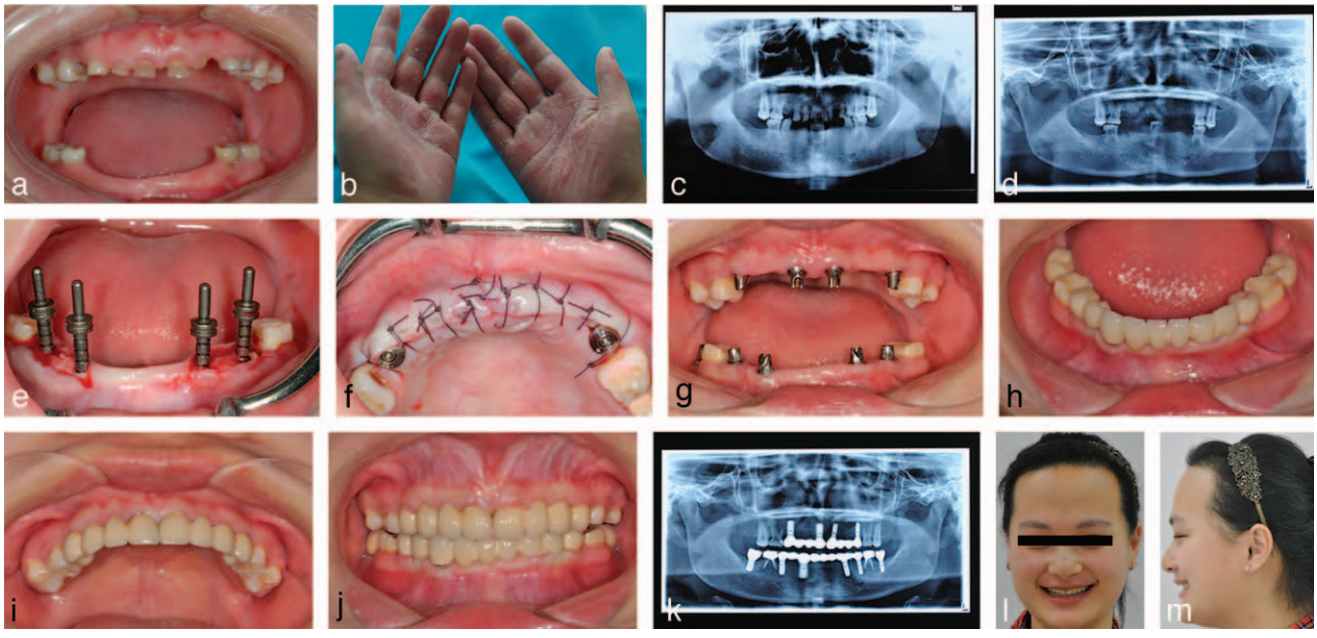
Patients' satisfaction*

Facial Contour	Prosthesis Esthetics	Pronunciation	Prosthesis Function
19 patients = 2, 33 patients = 1	20 patients = 2, 2 patients = 1	21 patients = 2, 1 patient = 1	20 patients = 2, 2 patients = 1

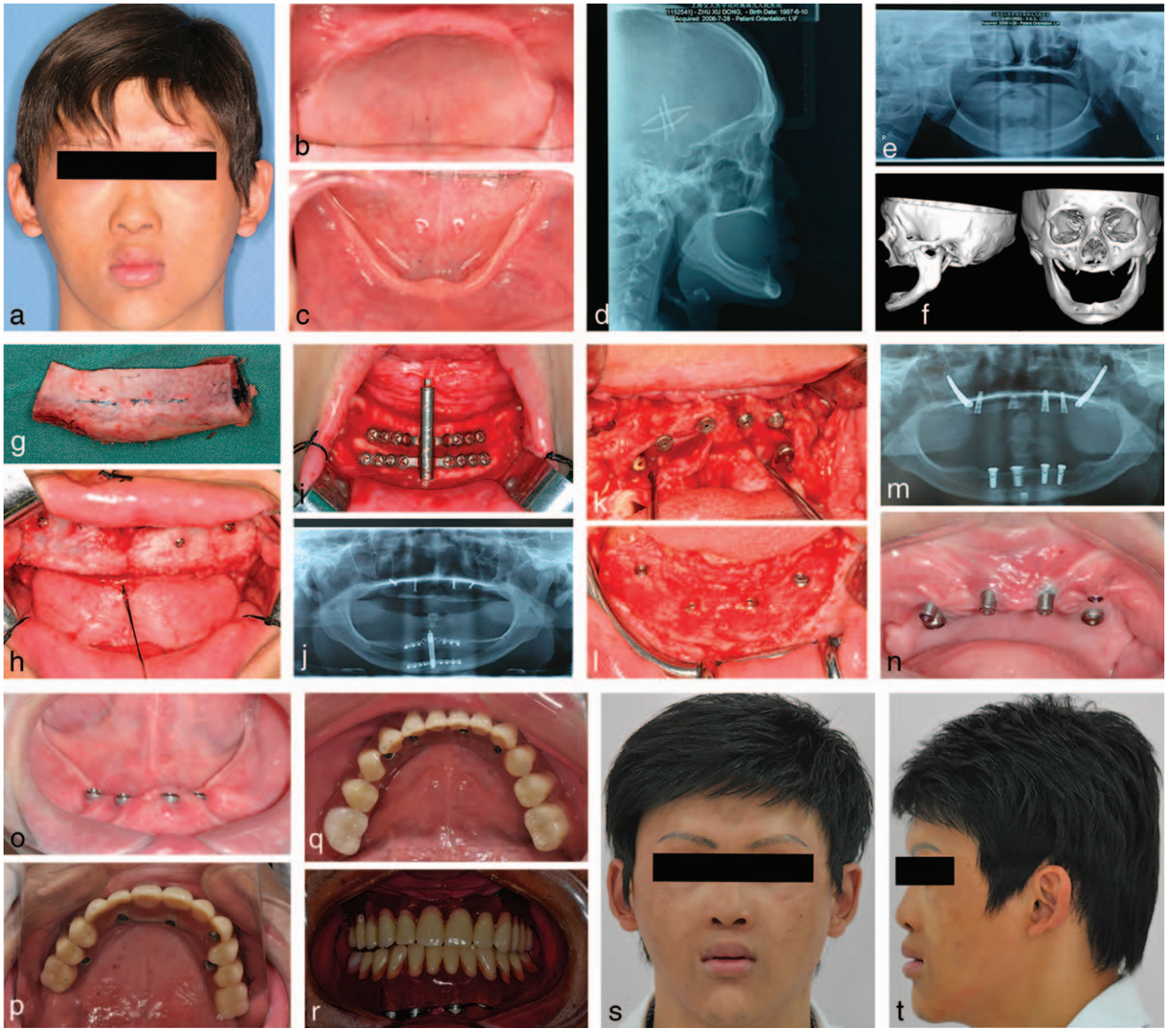
*0 = unsatisfied; 1 = partially satisfied; 2 = fully satisfied. Three patients were not included because they were lost to follow-up.

ZIs and 169 were CIs. One patient chose removable prostheses, whereas 24 patients underwent reconstruction using fixed prostheses (Table 1). Over the 3- to 5-year period of follow-up observations, 3 patients (12%) did not return for annual evaluations; these cases were designated as lost to follow-up. Five CIs failed and 3 CIs were removed during the follow-up period. The 3-year success and cumulative survival rates were 97.2% and 98.3%, respectively (Table 2). The data collected at each follow-up assessment for the SBI and PD measurements from each implant are presented in Tables 3 and 4. The probing data showed that SBI scores of 0 were observed in 84% (range 82.8% to 84.4%) of implants over the 3–5 years of follow-up. The PD values were measured as increases from baseline values obtained at the prosthesis insertion visit 3–5 years after the implant placement surgery. The majority of the PD values for implants were between 0 and 1 mm, and only a few values were $\geq$3.5 mm.

During the course of the study, peri-implantitis was observed in 8 cases, 3 of which required implant removal (incidence 4.5%). Measurements of general oral health and hygiene (eg, gingival and plaque indices) revealed a slight downward trend during the follow-up period (Figure 1). Four incidences of radiolucency occurred, as detected by a baseline radiograph for CIs. On the subsequent 1-year radiographs, the radiolucency disappeared. The data from implant radiographs showed crestal bone change outcomes as illustrated in Figure 2. The number of implants in this figure represents the radiographs qualified for this analysis at each study interval, whereas all radiographs for all patients were inspected for progressive bone loss. Bone loss values of 0–3 mm during 12 months were observed



FIGURES 3. Clinical procedure for restoring oral function in hypohidrotic ectodermal dysplasia patients with hypodontia using conventional implant-supported fixed prosthetic replacement. (a–c) Examination of patient characteristics using oral, hand, and panoramic X rays. (d–f) Placement of implants. (g–k) Implant-supported fixed prosthetic rehabilitation. (l–m) Front and side pictures following reconstruction.



Figures 4. Clinical procedure for restoring oral function in hypohidrotic ectodermal dysplasia patients with anodontia using zygomatic implants and conventional implant–supported fixed prosthetic replacement. (a–f) Evaluation of patient characteristics using oral, body surface, and panoramic X rays and computerized tomography. (g–j) Bone augmentation using autogenous bone grafting from the ilium and distraction osteogenesis. (k–o) Placement of implants. (p–r) Implant-supported fixed prosthetic rehabilitation. (s–t) Front and side pictures following reconstruction.

for 98.3% of the implants. Cases of severe bone loss occurred during the first year after the implants were placed, and bone loss was maintained at a relatively constant level afterward. During the 3–5 years of follow-up, crestal bone loss exceeded 5.0 mm in 3 cases (Table 4) and was associated with severe peri-implantitis.

Except for a small number of cases of screw loosening, the prosthetic reconstructions were successful during the follow-up period. Nineteen patients were fully satisfied and 3 were partially satisfied with their facial contours. Of the 22 patients (excluding the 3 lost to follow-up), 20 were fully satisfied with the esthetic aspect of their prostheses. One patient who was partially satisfied with the function of her prostheses had peri-implant infections. All patients were fully satisfied with their pronunciation ability, except for the patient who chose removable prosthetic restorations. One patient was only partially satisfied with the function of his prostheses because of the insufficient stability of the ceramic crown. The

patient satisfaction scores are summarized in Table 5. The restoration outcomes of two cases are illustrated in Figures 3 and 4.

Discussion

The results of this study confirm that the oral function of patients with HED can be reconstructed using bone augmentation and implant-supported prostheses. It is necessary to apply multidisciplinary treatment planning concepts for the successful rehabilitation of oral function in these patients.

Patients with HED, particularly those with anodontia, often display maxillary hypoplasia, mandibular prognathia, bone atrophy of the jaw, and/or facial concavity.[13] The reconstruction of oral function in these patients is challenging because of the severe bone deficiencies. Bone augmentation is a prerequisite step before the restoration of oral function with implants. In this study, we used 3 methods to augment bone volume in the jaw: artificial bone (Bio-Oss), autografts (mandibular oblique ridge, ilium, or calvaria), and DO. For minor bone insufficiency at the horizontal or vertical level, artificial bone is an effective means to increase bone volume.[14] Sixteen of 25 patients with hypodontia were given Bio-Oss and Bio-Gide to augment bone. Onlay grafting is also regarded as an effective approach for implant placement,[11] and autografts were taken for 10 patients. DO was also used to augment vertical bone volume in the mandible for 2 patients. In contrast to autogenous bone grafts, DO accomplishes bone augmentation without the disadvantages of damaging healthy tissue or producing morbidity at the donor site. However, successful DO requires close cooperation between the patient and the doctor because the distraction is activated in the mandible at a rate of 4 times per day (0.25 mm each time) and lasts 15 days.[1] When patients present with unfavorable intermaxillary relationships, orthognathic surgery may also be indicated. Five of 25 patients underwent orthognathic surgery in the maxilla, and the unfavorable intermaxillary relationships were corrected. All of the patients experienced successful bone grafts, although the autogenous bone or artificial bone grafts displayed a slight resorption in the area of bone augmentation, particularly in the iliac grafts. Sufficient augmentation of bone volume provided not only adequate facial contours, but also adequate support for the subsequent phases of implant placement and for the restoration of oral biological functions.

To minimize the dangers of implants becoming embedded, relocated, or displaced as the jaws grow or inhibited maxillofacial tissue growth by a rigid implant-supported prosthesis, we chose the patients near adulthood or in the lower portion of the declining adolescent growth curve (age $>$ 16) using the diagnosis of hand/wrist radiographs.[15] One hundred seventy-nine implants were placed in the 25 patients, including 10 ZIs and 169 CIs. During the 3-year follow-up, the success and survival rates were 97.2% and 98.3%, respectively. Five of the 179 implants failed, and 3 implants were removed. The implant failures occurred mainly at the position of bone absorption of the maxilla for the iliac grafts. Following significant bone absorption, the periodontal pockets became deeper, the thickness of the mucosa increased, and the incidence rate of peri-implantitis increased. All of the ZIs were successful, and complications such as peri-implantitis, infections, and maxillary sinusitis were not detected. Peri-implantitis is often difficult to treat using local or systemic antimicrobial approaches, or even with surgical interventions.[16–18] Oral hygiene played a critical role in the prevention of peri-implantitis and in the successful treatment of the patients in the present study. Patients with HED should use topical fluoride daily for prophylaxis against the high risk of caries resulting from dry mouth, as well as chlorhexidine in combination with daily use of a soft toothbrush to combat the inflammatory response.[19] The general oral health and hygiene of patients in the present study were measured by the gingival and plaque indices. These data indicated that (1) all patients maintained good oral health and hygiene except for the 3 patients lost to follow-up, and that (2) a slight downward trend in the indices occurred during the follow-up period.

Success in detecting the signs of mucositis and peri-implantitis and in initiating proper intervention can prevent progressive bone loss that causes implant failure. The early clinical detection of mucositis and peri-implantitis is best accomplished by using systematic mucosal probing techniques to measure SBI and PD. There was a low incidence of possible signs of peri-implantitis based on the results from the SBI scores: 84% of implants had a

score of 0. Two detected cases of peri-implantitis were successfully treated by surgical debridement and systemic antibiotics, and 3 peri-implantitis cases were maintained in a controlled state. In 3 other peri-implantitis cases, the development of inflammation and resulting bone loss could not be controlled, requiring implant removal.

The diagnosis of peri-implantitis requires a loss of crestal bone in conjunction with a probing depth $\geq$5 mm.[20] In the present study, radiographic evidence of bone loss was initially obtained at a follow-up appointment. The pattern of regression was notable in the first year; subsequently however, no progressive bone resorption was reflected by the mean values following the insertion of the permanent prosthesis 1 year after the implant placement. Notable bone resorption was observed in the cases of uncontrolled peri-implantitis. The radiographic results were generally consistent with the SBI and PD results. The data regarding patient satisfaction indicated that $>$90% of the patients were fully satisfied with the reconstruction of oral function including facial contours, esthetic aspect of the prosthesis, function, and pronunciation ability. One patient chose removable prostheses for the mandible for economic reasons and was partially satisfied.

Many previous reports have shown that the oral function of patients with ED can be restored using dental implants.[5,20–23] However, there have been few prospective studies on the restoration of oral function in patients with ED using systematic patient selection and a long-term follow-up. The main objective of the present study was to determine the clinical efficacy of oral function reconstruction in a prospective 3–5 year study. In many cases, we used ZIs to restore oral function in patients with anodontia. Peri-implantitis was carefully monitored during the 3-to 5-year follow-up period. Of course, longer observation is necessary to evaluate the final efficacy of the reconstruction of oral function in HED patients.

The clinical data studied included the level of crestal bone resorption and SBI and PD (using 3 scores) in implants. Our findings did not show a significant difference between ZIs and CIs, mainly because the number of ZIs was much lower. We expect that future studies using larger numbers of ZIs will reveal a significant difference. Additionally, comparing the results with other types of edentulous patients and detecting the differing genetic mutations in patients with various missing teeth will be our future focus for ED research.

Conclusions

The results of this study indicate that (1) the bone volume in the jaws of HED patients can be augmented using artificial bone, autografts, or DO; (2) the oral function of HED patients can be effectively reconstructed using implant-supported prostheses based on bone augmentation; and (3) the patients expressed high satisfaction with the restoration of their oral function.

Abbreviations

CI: conventional implant
DO: distraction osteogenesis
ED: ectodermal dysplasia
HED: hypohidrotic ectodermal dysplasia
PD: probing depth
SBI: sulcus bleeding index
ZI: zygomatic implant

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References

1. Li D, Liu Y, Ma W, Song Y. Review of ectodermal dysplasia: case report on treatment planning and surgical management of oligodontia with implant restorations. *Implant Dent*. 2011;5:328–330.

2. Kramer FJ, Baethge C, Tschernitschek H. Implants in children with ectodermal dysplasia: a case report and literature review. *Clin Oral Implants Res*. 2007;1:140–146.

3. Guckes AD, Scurria MS, King TS, et al. Prospective clinical trial of dental implants in persons with ectodermal dysplasia. *J Prosthet Dent*. 2002;1:21–25.

4. Liu Y, Yu X, Wang L, et al. Mutation p.Leu354Pro in EDA causes severe hypohidrotic ectodermal dysplasia in a Chinese family. *Gene*. 2012;2:246–250.

5. Al-Ibrahim HA, Al-Hadlaq SM, Abduljabbar TS, et al. Surgical and implant-supported fixed prosthetic treatment of a

patient with ectodermal dysplasia: a case report. *Spec Care Dentist*. 2012;1:1–5.

6. Van Sickels JE, Raybould TP, Hicks EP. Interdisciplinary management of patients with ectodermal dysplasia. *J Oral Implantol*. 2010;3:239–245.

7. Sclar AG, Kannikal J, Ferreira CF, et al. Treatment planning and surgical considerations in implant therapy for patients with agenesis, oligodontia, and ectodermal dysplasia: review and case presentation. *J Oral Maxillofac Surg*. 2009;11:2–12.

8. Bayat M, Khobyari MM, Dalband M, Momen-Heravi F. Full mouth implant rehabilitation of a patient with ectodermal dysplasia after orthognathic surgery, sinus and ridge augmentation: a clinical report. *J Adv Prosthodont*. 2011;2:96–100.

9. Garagiola U, Maiorana C, Ghiglione V, et al. Osseointegration and guided bone regeneration in ectodermal dysplasia patients. *J Craniofac Surg*. 2007;6:1296–1304.

10. Baig MR, Rajan G, Rajan M. Edentulous arch treatment with a CAD/CAM screw-retained framework and cemented crowns: a clinical case report. *J Oral Implantol*. 2009;6:295–299.

11. Chiapasco M, Casentini P, Zaniboni M. Bone augmentation procedures in implant dentistry. *Int J Oral Maxillofac Surg*. 2009;24: 237–259.

12. Albrektsson T, Zarb G, Worthington P, et al. The long-term efficacy of currently used dental implants: a review and proposed criteria of success. *Int J Oral Maxillofac Implants*. 1986;1:11–25.

13. Worsaae N, Jensen BN, Holm B, Holsko J. Treatment of severe hypodontia-oligodontia—an interdisciplinary concept. *Int J Oral Maxillofac Surg*. 2007;6:473–480.

14. Jensen T, Schou S, Svendsen PA, et al. Volumetric changes of the graft after maxillary sinus floor augmentation with Bio-Oss and autogenous bone in different ratios: a radiographic study in minipigs. *Clin Oral Implants Res*. 2012;8:902–910.

15. Oesterle LJ. Implant considerations in the growing child. In: Higuchi KW, ed. *Orthodontic Applications of Osseointegrated Implants*. Chicago, Ill: Quintessence Publishing Co; 2000:133–159.

16. Bach G, Neckel C, Mall C, Krekeler G. Conventional versus laser-assisted therapy of periimplantitis: a five-year comparative study. *Implant Dent*. 2000;3:247–251.

17. Schwarz F, Sculean A, Bieling K, et al. Two-year clinical results following treatment of peri-implantitis lesions using a nanocrystalline hydroxyapatite or a natural bone mineral in combination with a collagen membrane. *J Clin Periodontol*. 2008; 1:80–87.

18. Claffey N, Clarke E, Polyzois I, Renvert S. Surgical treatment of peri-implantitis. *J Clin Periodontol*. 2008;8:316–332.

19. Salvi GE, Persson GR, Heitz-Mayfield LJ, et al. Adjunctive local antibiotic therapy in the treatment of peri-implantitis II: clinical and radiographic outcomes. *Clin Oral Implants Res*. 2007;3: 281–285.

20. Mombelli A. Microbiology and antimicrobial therapy of peri-implantitis. *Periodontology 2000*. 2002;28:177–189.

21. D'Ambrosio A, Cioffi A, Cirillo A, Sammartino G. Immediate loading in ectodermal dysplasia: clinical case. *J Oral Maxillofac Surg*. 2011;9:2351–2356.

22. Grecchi F, Zingari F, Bianco R, et al. Implant rehabilitation in grafted and native bone in patients affected by ectodermal dysplasia: evaluation of 78 implants inserted in 8 patients. *Implant Dent*. 2010;5:400–408.

23. Yap AK, Klineberg I. Dental implants in patients with ectodermal dysplasia and tooth agenesis: a critical review of the literature. *Int J Prosthodont*. 2009;3:268–276.