Perceptions of Outcomes of Implant Therapy in Patients with Ectodermal Dysplasia Syndromes

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Purpose: The purpose of this study was to evaluate patient-specific outcomes and satisfaction using dental implants in a population affected with ectodermal dysplasia. ***Materials and Methods:*** Patient-based data were collected using a self-reported survey instrument sent to patients belonging to a private patient foundation and/or treated previously at a government clinic. A standardized survey instrument was developed to evaluate patient satisfaction, outcomes, and potential complications using dental implants. ***Results:*** The survey instrument was mailed to 253 affected individuals self-reported to have various forms of ectodermal dysplasia and who were voluntarily participants in the National Foundation for Ectodermal Dysplasias and/or were participants in the US National Institute for Dental Craniofacial Research Intramural Ectodermal Dysplasia clinical research program. A total of 109 responses were obtained (43% response rate). The duration following completion of implant therapy ranged from to 1 to 23 years. Of the 109 participants, 50% reported either an implant or prosthetic complication with implant treatment, and 24% reported some form of failure with implant therapy. However, 91% of participants reported being either satisfied or very satisfied with dental implants, and 95% reported that the treatment was worth the time and cost. ***Conclusions:*** Affected individuals receiving tooth replacement therapy with dental implants reported satisfaction with the outcome. A higher level of complications, including infection, mechanical problems, and implant loss, relative to the unaffected population was reported. *Int J Prosthodont 2008;21:195–200.*

Ectodermal dysplasia (ED) syndromes are a group of heritable disorders affecting the teeth, hair, nails, and glands and are clinically diagnosed when 2 or more structures are involved.[1] According to the Compendium of Birth Defects, as few as 1 to 7 in 10,000 births results in a dysplasia of an ectodermally derived structure.[2] More than 170 different genetic syndromes have been identified with mild to severe manifestations in the ED syndromic family. Individuals affected may have a combination of symptoms including sparse hair, brittle nails, dental malformations, skin problems, absence of sweat glands, and complications of swallowing, vision, and hearing.[3]

Individuals affected by ED syndromes often experience a significant need for oral health care. Hypodontia and anodontia are frequently reported and create complex restorative issues.[1,4–8] The resulting lack of bone formation and normal oral and craniofacial development leads to significant diagnostic and treatment challenges.[5] Besides hypodontia and anodontia, other oral craniofacial concerns include abnormal patterns of tooth eruption, widely spaced teeth, poorly shaped teeth, and salivary hypofunction.[9] A functional dentition is important for proper diet, facial esthetics, speech, and emotional development.[10] The treatment plans to meet the needs of these individuals can be extensive, including tissue-supported removable prostheses, implant-supported removable prostheses, and implant-supported fixed prostheses of various designs.[8] As is common with many special needs patients, those affected with ED syndromes often travel long distances at considerable expense in time and money to obtain multi-specialist care at a regional, often university-based, clinical care center.

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In the late 1980s, a National Institutes of Health (NIH) intramural clinical research project was initiated to evaluate the safety and efficacy of dental implants in individuals affected by ED syndromes. This project was conducted at the outpatient clinic at the National Institute of Dental and Craniofacial Research (NIDCR) in Bethesda, Maryland.[11] The active phase of implant treatment occurred between 1986 and 1994. No systematic attempt to provide long-term follow-up of outcomes in this population was performed. To understand long-term outcomes of this care, a patient-based survey instrument was created with a core of the Oral Health Impact Profile (OHIP-14) instrument. This instrument was sent to all individuals treated at the NIDCR clinical study and all individuals in the National Foundation for Ectodermal Dysplasias (NFED) (Mascoutah, Illinois) database who reported having had dental implant therapy as part of their dental treatment plan. The survey was designed to capture information about the implant therapy itself, complications, impact on quality of life (QOL), care providers, and the health and economic impact of implant therapy in this population.

Materials and Methods

The survey instrument used to assess patient demographics, dentition, interventions, time of care, and family impact was designed during a 2-day workshop held by the investigative team. The QOL component was based on the OHIP-14 instrument.[12] The survey instrument was pilot tested on a small group of subjects followed by a mass mailing to all members of the NFED and the NIDCR study population that had previously self-disclosed undergoing dental implant therapy. A total of 253 surveys was distributed, with 30 surveys returned because of out-of-date addresses. Twelve surveys were not completed because no implants were placed (treatment planned but not performed). Of the remaining surveys, 109 were completed and returned (43%). Of the 109 completed surveys, 91 were completed with sufficient detail to be extracted for this analysis, resulting in a 36% final rate of evaluation. Of the 109 participants, 26 had participated in the NIDCR study. The remaining 83 participants were NFED members who had dental implants as part of their dental treatment plans.

To improve the response rate, the instrument was mailed 3 times. Reminder postcards were sent 1 month after the initial mailing, followed by redacting mailings to individuals who had not returned the survey. A commercial internet-based "people finder" service (www.peoplefinders.com) was used along with contacting city-by-city telephone directory assistance to locate as many of the NIDCR study participants as possible. Phone calls and personalized notes were sent to improve the response rate.

The 26 NIDCR respondents represented 49% of the 53 individuals originally treated in the intramural study. In the NIDCR population, there were 264 implants placed (4.0 or 3.75 mm diameter; Brånemark, Nobel Biocare) in 39 male and 14 female patients (aged 8 to 68 years at the time of implant placement) with 242 implants in the mandible and 28 in the maxilla.[11] Two of the 53 individuals self-reported never receiving dental implants and were excluded from this study. Of the 27 individuals located, 1 individual failed to return the survey.

Of the 26 individuals from the NIDCR study, there were 21 males and 5 females. Twenty individuals completing the survey were between the ages of 18 and 34 years old at the time of treatment. Ten individuals lived in southern states, 7 in the Midwest, 6 in the Northeast, 2 in western states, and 1 outside the USA.

Two hundred twenty surveys were mailed to individuals in the NFED database who reported having dental implant treatment. Eighty-three individuals (38%; 36 males and 42 females) returned the survey and were included in the analysis. The participants were evenly distributed across the USA. All data were tabulated using SPSS 13.0 software (SPSS).

Results

Dental implant therapy was performed between 1981 and 2004. The median patient age for implant placement in the maxilla was 18 years of age (range: 12 to 70 years); for the mandible, it was 17 years of age (range: 5 to 72 years). A short health-risk survey was included in the survey instrument. Five individuals reported smoking cigarettes, 27 individuals reported chronic health problems, and 7 reported taking corticosteroids routinely. Retrospectively, when asked about the reasons for seeking implant therapy, individuals reported that denture stability and retention was the primary reason (NIDCR population: 88%; non-NIDCR: 83%), followed by chewing (NIDCR: 89%; non-NIDCR: 77%), appearance (NIDCR: 89%; non-NIDCR: 75%), and speech (NIDCR: 81%; non NIDCR: 47%).

Dental Care Providers

All individuals who responded to this survey indicated that they had seen more than one dental specialist when planning implant treatment (Table 1). Most individuals saw more than 4 different specialists during the formulation of the dental treatment plan. Twenty-one percent of the NIDCR population and 51% of the non-NIDCR population received orthodontic care as part of their dental treatment plan. Of the non-NIDCR population who received orthodontic care, 16% had the orthodontic care repeated. Thirty-five percent of the

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Table 1 Types of Dental Clinician Seen by Patients During Implant Treatment

Type	No. of patients	% of patients
General dentist	38	35
Pediatric dentist	2	2
Orthodontist	1	1
Prosthodontist	43	39
Oral surgeon	4	4
Periodontist	3	3
Maxillofacial prosthodontist	1	1
Dental student	1	1
Unsure	2	2
Total answered	95	88
Unanswered	14	12
Total	109	100

Table 2 Maxillary Teeth Present at the Start and Conclusion of Implant Therapy

	Before treatment		After treatment	
No. of teeth	No. of patients	% of patients	No. of patients	%. of patients
0	10	9	22	20
1–5	43	39	46	42
6–10	37	34	22	20
11–16	10	9	5	5
Total answered	100	92	95	87
Unanswered	9	8	14	13
Total	109	100	109	100

Table 3 Mandibular Teeth Present at the Start and Conclusion of Implant Therapy

	Before treatment		After treatment	
No. of teeth	No. of patients	% of patients	No. of patients	%. of patients
0	26	24	47	43
1–5	38	35	37	34
6–10	25	23	16	15
11–16	13	12	6	5
Total answered	102	94	106	97
Unanswered	7	6	3	3
Total	109	100	109	100

NIDCR study participants indicated that they had a difficult time finding a dental practitioner to provide follow-up care after the completion of implant therapy, and 41% of the non-NIDCR group reported the same. Of the NIDCR population who had difficulty finding follow-up care, 67% reported that dental practitioners were either unwilling or too inexperienced to provide follow-up care. In the NFED population, 70% reported similar problems (33% of clinicians unwilling to treat and 66% inexperienced or uncomfortable providing care). The majority of both populations (77%) were currently seeing either a general dentist or a prosthodontist for maintenance dental care.

Number of Teeth Present at the Start and Completion of Implant Therapy

The number of teeth present can significantly influence the implant treatment plan; therefore, respondents were asked to indicate the number of primary and adult teeth they developed and the number of natural teeth they had at the time of dental implant therapy (Tables 2 and 3). The majority of patients (39%) reported 1 to 5 teeth in the maxilla and 35% reported 1 to 5 teeth in the mandible at the start of implant therapy. In a subanalysis of the dataset, 21% of the NIDCR population reported having no natural teeth in the maxilla at the start of treatment, 46% reported 1 to 5 natural teeth at the start of treatment, and 33% reported 6 to 10 natural teeth at the start of treatment. The distribution of teeth in the NIDCR study population has previously been presented.[1] In the subset analysis of the mandible, 63% of the NIDCR population reported they developed no adult teeth, 29% developed 1 to 5 adult teeth, and 8% developed 6 to 10 adult teeth. Seventy-two percent of the total population reported having teeth extracted at the start of dental implant treatment. Following implant treatment in the maxilla, 12 patients had all remaining natural teeth extracted as a part of the treatment plan (Table 2). There was a similar trend in the mandibular arch, with an increase from 24% (26 patients) to 43% (47 patients) reporting being edentulous at the conclusion of implant therapy. Prior to implant therapy, patients reported an average of 12.9 $\pm$ 12.5 years wearing complete or partial dentures.

Implant Therapy

All NIDCR participants received care at the Bethesda NIH campus (1986 to 1994). Fifty-four percent of the non-NIDCR population reported dental implant care provided by private practitioners and 41% reported receiving care at university dental schools. Ninety-two percent of these individuals traveled more than 60 miles for each appointment. An oral and maxillofacial surgeon placed the implants in the NIDCR study, with a prosthodontist or general dentist providing the prosthesis. In the non-NIDCR population, oral and maxillofacial surgeons placed the majority of implants (in 54% of the patients), with prosthodontists completing the restorative phase in 47% of the individuals. There was no direct fee paid by participants in the NIDCR study, whereas the non-NIDCR population reported an average cost of $39,170 USD (median: $25,000; range: $2,740 to $110,000).

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At the start of the NIDCR study (1986), the protocol for predictable implant placement in the maxilla was still being developed. The location of the sinus cavities in patients with advanced atrophy introduces complications such as the need for bone grafting and/or use of shorter dental implants. Overall, 54% of patients reported having no implant therapy in the maxilla, and those that did had a median number of 4 implants placed (Table 4). In both populations, a median number of 4 implants was placed in the mandible. The non-NIDCR population reported an average of 2.7 (SD: 3.0) implants in the maxilla and an average of 4.1 (SD: 2.4) implants in the mandible. Thirty-five percent of the NIDCR population had bone augmentation as part of the treatment plan (80% autogenous graft material, 75% derived from the iliac crest, and 25% from intraoral sites). Fifty-two percent of the non-NIDCR population reported that bone augmentation was a part of the treatment plan (61% autogenous graft material, 69% derived from the iliac crest, and 31% from intraoral sites). Two individuals in the non-NIDCR group reported failed bone grafts. Definitive prosthesis designs were either a fixed complete denture or a bar-retained overdenture. The majority of the NIDCR population (79%) and a smaller percentage of the non-NIDCR population (41%) reported they had screw-retained definitive prostheses.

Table 4 No. of Patients Relative to the No. of Implants Placed

No. of implants	Maxilla		Mandible	
	No.	%	No.	%
0	54	50	6	6
1	2	2	2	2
2	5	5	13	12
3	7	6	6	6
4	9	8	34	31
5	5	5	22	20
6	11	10	8	7
7	2	2	2	2
8	5	5	4	4
10–14	1	1	3	3
Total answered	101	93	100	93
Unsure	8	7	9	7
Total	109	100	109	100

Complications Following Implant Therapy

Half of the responders perceived some form of postoperative complication (52 of 105 responders) (Table 5). When asked if they had "implant failures," 23 of 96 responders indicated they had some form of implant failure (24%). Of the 52 responders reporting complications, 23% indicated infections (12 of 52 responders) and 19% (10 of 52 responders) related some form of prosthetic complications (loose or broken screws, loose overdentures, etc).

In a subset analysis, 11 individuals (44%) in the NIDCR population experienced complications following dental implant placement. Three individuals (27%) experienced infection and 10 individuals experienced mechanical complications. Other reported complications included mobile or loose implants (18%) and painful, swelling, and inflamed periodontal tissues (18%). Eight individuals (33%) in the NIDCR population had 1 or more dental implants fail.[11]

Twenty-three patients (33%) in the non-NIDCR population experienced complications. Of those responders, 9 individuals (39%) experienced infection and 20

Table 5 Rate of Complications with Implant Therapy in Children (< 18 Years) Versus Adults (≥ 18 Years)

Complications	Children (n = 12)		Adults (n = 85)		
	No.	%	No.	%	*P**
Complications with implants	4	33.3	28	32.9	1.00
Infection	1	8.3	10	11.8	1.00
Loose/mobile implant	2	16.7	7	8.2	0.31
Broken FPD	1	8.3	4	4.7	0.49
Loose denture	0	0.0	3	3.5	1.00
Denture remake	0	0.0	3	3.5	1.00
Denture reline	0	0.0	2	2.4	1.00
Fractured denture	1	8.3	4	4.7	0.49
Speech problems	0	0.0	2	2.4	1.00
Esthetics	0	0.0	6	7.1	1.00
Unsuccessful bone graft	0	0.0	2	2.4	1.00
Gingival recession	0	0.0	2	2.4	1.00
Pain, bleeding, or swelling	0	0.0	3	3.5	1.00
Loose FPD	0	0.0	1	1.2	1.00
Broken screws	0	0.0	3	3.5	1.00

*Two-sided Fisher exact test. No significant differences were found in terms of age ($P < .05$).
FPD = fixed partial denture.

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(87%) reported mechanical complications such as denture fracture, denture reline, loose dentures, or broken screws. Other complications included mobile or loose implants at some point following placement (35%) and painful, swelling, or inflamed periodontal tissues (4%). Fifteen individuals (20.8%) had 1 or more dental implants fail (ie, removal from the mouth).

Since there was a range of ages treated, a subset analysis was performed to separate complications in children (< 18 years of age) from adults (≥ 18 years of age). The rate of reported complications was not significantly different for any form of reported implant or prosthetic complication (Table 5).

Satisfaction with Implant Therapy

When asked about the overall cost/burden in terms of morbidity, time, resources, etc, 95% of all respondents reported that implant therapy was worthwhile. Ninety-one percent reported that they were either very satisfied (70%; 70 of 100 respondents) or satisfied (21%; 21 of 100 respondents) with implant therapy. In regard to satisfaction with the implant prosthesis, 90% of respondents were either very satisfied (59%; 58 of 98 respondents) or satisfied (32%; 31 of 98 respondents), and 92% of the respondents stated they would repeat the method of treatment provided.

Discussion

This study reports the outcomes of a retrospective patient-based survey of patients affected with various forms of ED syndromes. A primary reason for performing this study was to gain a better understanding from a patient-oriented perspective regarding the outcomes and impact of one form of tooth replacement therapy.

EDs are a complex heterogeneous grouping of anomalies defined by having a dysplasia in 2 or more ectodermally derived structures. The current clinical grouping of manifestations suggests that more than 200 distinct disorders comprise the various EDs, though the most commonly reported form, hypohidrotic ED (HED, or Christ-Siemens-Touraine Syndrome) is typically described in various prosthodontic case reports.[13–27] The oral clinical phenotype of HED results in multiple missing or deformed teeth, lack of alveolar ridge development, and hyposalivation.[14,16–18,27] These conditions can result in complications for the placement of dental implants used to retain a removable overdenture or fixed prosthesis.[17] The US Public Health Service NIDCR intramural clinical research program in Bethesda, Maryland, undertook a clinical research project in the mid-1980s to the mid-1990s to evaluate the safety and efficacy of dental implants in individuals affected by ED.[11,28] Forty patients over the age of 13 and a smaller number of children between the ages of 7 and 10 were provided with implants and a prosthetic reconstruction. Affected individuals receiving dental implants through this study reported success and satisfaction with their care, suggesting that dental implants may be a viable and successful treatment option for individuals affected by ED syndromes.[11] Since the closure of the NIDCR project in the mid-1990s, there has been little follow-up on outcomes in this population. This survey project was therefore undertaken to follow the original cohort of affected individuals and to survey the active database of patient participants who belonged to a national patient-based research foundation.

The outcomes of this study suggest that in the sample of responders, there was a high rate of postoperative complications, with more than half reporting some form of patient-perceived complication during a period between 1981 and 2004. Of these complications, 23% reported "infections" around the implants at some period of time, with a smaller number reporting prosthetic complications (19%). Nonetheless, it is important to note that, when asked if they would repeat whatever form of implant therapy they had received, 92% indicated they would.

In the development of this survey instrument, the investigative team reviewed existing patient-based instruments and created a panel of questions targeted to address the follow-up outcomes of implant therapy in this affected population. The questions were reviewed by an outside advisory board, but no formal test-retest validation was performed. Thus, a limitation of this study is the potential bias induced by some of the terms used. For instance, the high level of "infections" reported may be unclear since exactly what constitutes an infection is open to patient interpretation (pain, swelling, clinician pronouncements of "peri-implant mucositis," "peri-implantitis with bone loss," "frank acute infection," etc). Further, the complication rates may be biased because of the low percentage of responders that completed the complication section of the instrument. For instance, in the complication section, only 52 of the 109 respondents completed the questions regarding the presence or absence of a complication (yes/no). This may have been a result of the length of the instrument, although great attention was paid to creating an instrument that could be completed within 15 to 20 minutes. As with any retrospective patient-based study, the outcomes of this study may be complicated by recall bias given the duration over which respondents completed implant therapy (1981 to 2004). There is also the potential responder bias induced by either overly positive or overly negative responses skewing the outcomes, which is a common condition with retrospective studies. Nevertheless, the outcomes are compelling since ED is a rare condition that demonstrates challenging dental and oral craniofacial conditions.

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Over the past few years, there have been a series of case reports on implant therapy in young subjects with oligodontia or anodontia. These reports often provide short-term technical or procedural approaches, which may demonstrate their technical capabilities but gives little regard to the long-term commitment made by the clinician. When implant therapy is performed, especially in a young adult, the osseointegrated implants placed are intended to be present for the rest of the patient's life. This creates an ethical commitment on the part of the surgical and restorative team to provide clear information as to the reasonable outcomes of care. This study provides patient-specific outcomes that suggest that while the patient-perceived benefits may be high, the complication rates suggest the need to balance the sophistication of the prosthetic interventions with the costs of long-term maintenance, re-restoration, and access to care.

The diagnostic and treatment challenges associated with multiple missing teeth, the resulting lack of bone formation, and oral and craniofacial malformation may be beyond the capabilities of some dental clinicians in general practice. To provide reconstructive services, teams of prosthodontists, orthodontists, oral and maxillofacial surgeons, pediatric dentists, general dentists, geneticists, and other related specialties often work together in regional and academic centers. Consequently, ectodermal dysplasia–affected patients often travel long distances at great expense to receive treatment. Nonetheless, the benefits of care appear to outweigh the various costs, and thus dental implant therapy provides an invaluable means to provide dental restorative and reconstructive care to patients with various forms of ectodermal dysplasia.

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