Implant-Prosthodontic Treatment for Special Care Patients: A Case Series Study

Ceyda Öczakir, Dr Med Denta/Sacha Balmer, Dr Med Dentb/Regina Mericske-Stern, Prof Dr Med Dentc

Purpose: The aim of the study was to assess implant survival and complications with implants and prostheses in patients exhibiting a variety of systemic diseases and congenital defects. *Materials and Methods:* Patients with specific medical conditions are regularly treated at the Department of Prosthodontics, University of Bern, Switzerland. All those who had received implant-prosthodontic treatment during the past 12 years were reexamined for this study. Among these patients the following diseases were observed: cleft lip/palate (n = 8), Down syndrome (n = 3), Sjögren syndrome and scleroderma (n = 2), ectodermal dysplasia (n = 4), developmental retardation (n = 2), chronic leukemia (n = 2), lichen planus (n = 1), cerebral palsy (n = 1), deaf-muteness (n = 1), amyotrophic lateral sclerosis (n = 1). At the time of the treatment the mean age was 55.6 years. ITI implants had been placed according to a standard protocol with local anesthesia, except for one patient in whom full anesthesia was used. One hundred three implants were loaded and supported a total of 34 fixed or removable prostheses. All patients were appointed to a regular maintenance care program. In the context of the present study, all but 1 patient were reexamined clinically. New radiographs were obtained, and the implants and prostheses assessed. Additional information was obtained from regular records in the patients' charts. Results: Three implants were lost in the healing phase, and 1 implant was replaced. Only 1 patient with 4 implants was lost from the study (she had passed away). The survival rate of the loaded implants was 100%. In 1 patient, peri-implant bony defects were detected around all 3 intraforaminal implants. The prosthetic plan was maintained in all patients, and they continued to wear the originally planned type of prosthesis. Complications included insufficient hygiene, soft tissue hyperplasia, extraction of remaining teeth, and minor maintenance or repair of the prostheses. Conclusions: So far, the mostly unknown implications and possible risks for the process of osseointegration and long-term maintenance in patients with such rare diseases and defects has resulted in a rather restricted application of implants. However, from the present results, it appears that implants can successfully be placed and maintained. This is ascribed in part to a strict maintenance care program provided by the caregivers and to a high compliance of the patients who participated in this program to perform good oral hygiene. Int J Prosthodont 2005;18:383–389.

Correspondence to: Regina Mericske-Stern, Department of Prosthodontics, University of Bern, Freiburgstrasse 7, CH 3010 Bern, Switzerland. Fax: +41-31-632-49-33. E-mail: regina.mericske@ zmk.unibe.ch Although not frequent, a variety of rare diseases, comprising congenital defects, disabilities or special systemic conditions, and local oral pathologies are found in dental patients. These include Down syndrome (chromosome 21 anomalies), syndromic cleft lip/palate, Sjögren syndrome, oral lichen planus, hereditary ectodermal dysplasia, and others. Some directly affect the oral cavity, while others have systemic or behavioral aspects. Patients with these conditions are in need of special management, and clear benefits may

^aGraduate Student, Department of Prosthodontics, University of Bern, Switzerland.

^bAssistant Professor, Department of Prosthodontics, University of Bern, Switzerland.

^cChair, Department of Prosthodontics, University of Bern, Switzerland.

be obtained from placement of implants to replace teeth or support removable prostheses. Implants may be a unique solution to provide these patients with stable prostheses, particularly if they have lost or did not form many or all their teeth.

Long-term maintenance of a rigid implant-bone interface requires continual bone remodeling and is determined by a complex tissue response. Osseointegration and maintenance of endosseous dental implants are influenced by many factors and are dependent on specific systemic or local oral conditions.^{2–5} A distinction has been made between possible and proven risks for successful osseointegration and stable maintenance of implants.⁶ Patients with various rare diseases are often considered to be at elevated risk of implant failure. Since they do not fulfill strict selection criteria, they are excluded from implant therapy. Today, such conditions are no longer an absolute contraindication for the placement of implants, and case reports provide evidence of successful treatment outcomes during the past 10 years.7-15 However, no comparative studies exist that demonstrate that any specific systemic disease or congenital condition would affect the process of osseointegration and soft tissue integration of the implants. Today it is clear that success or failure of osseointegration is multifactorial, dependent on local anatomic conditions, systemic health, genetic disposition, immune function, and behavioral factors. A recent study gave detailed recommendations on patient selection for implant placement with regard to patients' individual conditions. Further, reporting of changes or special events in patients with such diseases to a central register was suggested to make information better available and to expand knowledge.

With regard to the local oral situation or patients' behavior, prosthodontic treatment may be demanding and requires highly individual solutions. Bone and soft tissue irregularities, including alterations after surgical procedures, are frequent in cleft palate patients. 16 Additionally, a sagittal Angle Class III occlusion is common. Deficiencies in basal and alveolar bone growth result in poor support of prostheses in patients with hypohidrotic ectodermal dysplasia and/or anodontia. Further ankylotic infra-occlusion of the remaining teeth is often observed and may be difficult to compensate with the prostheses.¹⁷ Patients with reduced salivary flow and soft tissue pathologies may also experience problems with removable prostheses. Inability to adapt to complete prostheses is encountered in patients with motor and mental deficiencies. 11,14 Mental incompetence, local abnormalities, and disability compromise and hinder adequate hygiene performance procedures.

The aim of the present case series study was to assess implant and prosthesis survival in special care patients with rare systemic diseases and congenital defects.

Methods

Patients

Patients with specific medical conditions are regularly treated at the Department of Prosthodontics, University of Bern. All those who had received implant-prosthodontic treatment during the past 12 years were reexamined for this study. Altogether, 25 patients were identified with congenital and special systemic diseases from all patients' charts and recall registers of the Department of Prosthodontics, University of Bern. These were hereditary ectodermal dysplasia (HED), HED in conjunction with giant-cell granuloma (GCG), cleft lip and palate (CLP), Down syndrome (DS), Sjögren syndrome (SS) and scleroderma (S), cerebral palsy (CP), developmental retardation (DMR), chronic leukemia (CL), lichen planus of skin and mucosa (LP), amyotrophic lateral sclerosis (ALS), and deaf-muteness (D-M). This single edentulous D-M patient had also had a benign but extended osseous tumor removed from the right alveolar bone of the maxilla. Thus, wearing of a complete prosthesis became impossible for this patient.

The patients had undergone implant surgery and prosthodontic treatment during the last 12 years at the department. The group comprised 17 female and 8 male patients, with a mean age of 55.6 years at the time of implant placement (range, 19 to 89 years). The mean observation time was 5.8 years in the year 2003, with a range from a minimum of 2 years up to 12 years. Implants had not been placed in growing children or adolescents. ^{18,19} Two elderly patients with unoperated cleft palate had a large open oral-nasal defect and were in need of obturator prostheses. Two other patients with cleft palate had undergone bone graft procedures before implants were placed.

Inclusion criteria were:

- Poor stability of prostheses without implants
- Inability to wear prostheses or replace teeth without implants
- Inability of the patient to wear removable prostheses without better support and stable anchorage
- Sufficient bone to place implants using local anesthesia
- Strong wish of the patient for improvement of oral/dental prosthetic situation
- · Communication with patient is possible

Patients with systemic problems, which are typical for a geriatric population, were not included in this study.

Implants and Prostheses

At the time the treatment was performed, 10 patients were completely edentulous and 15 patients had remaining natural teeth in 1 or both jaws. All patients had received ITI implants (Straumann Dental Implant System) according to the same surgical protocol in a one-stage procedure. All but 1 patient received their implants under local anesthesia. This patient with cerebral palsy was operated under full anesthesia at the hospital. A total of 105 implants were placed in 25 patients, with 8 receiving implants in both jaws. Fourteen patients with a total of 53 implants had an observation period with regular recalls of more than 5 years.

After the surgical and prosthodontic treatment was completed, all patients were appointed for regular maintenance care and service. The patients participated at least once per year-typically 2 or 3 times-in recall appointments, which was organized by the dental hygienist. Under supervision of the clinician, the hygienist organized the patients visits, performed hygiene procedures, and if necessary, obtained radiographs. If any particular problems were registered, the patients were also seen by the clinician or received an additional appointment with the clinician. Radiographs, however, were not taken annually; therefore, mean annual bone loss could not be measured and is not reported in the present study. No patient included in this review dropped out permanently or was lost for unknown reasons from the recall system.

In 1 female patient with amyotrophic lateral sclerosis, both implants failed during the healing phase. She did not wish to have implants placed again. This patient was not included in the present report and is not presented in the tabular data, except for the survival analysis of the implants. Therefore, the present study reports on the results of 24 patients.

Data Collection

In the context of the present data collection, all 24 patients underwent a clinical examination by 1 clinician, and new radiographs were obtained in 2003. All patients were treated according to identical protocols, and treatment information was registered in the patients' charts and a computer. The present retrospective data collection comprises:

- Number and location of implants placed in the maxilla and mandible
- Failure of implants during the healing phase
- · Survival of loaded implants
- Type of prosthesis delivered to the patients in the maxilla and mandible
- · Survival of prosthesis

Table 1 Patients and Loaded Implants

Disease/ defects	No. of patients	No. of implants	
Ectodermal dysplasia	3	12	
Giant-cell granuloma	1	7	
Cleft lip/palate	8†	43	
Down syndrome	3	8	
Sjögren syndrome (SS)	1	4	
SS and scleroderma	1	8	
Mental retardation	2	5	
Cerebral palsy	1	2	
Chronic leukemia	2	4	
Lichen planus	1	4	
Deaf-muteness	1 [‡]	7	
Totals	24	103	

[†]In one patient, this was combined with mental retardation.

Table 2 Types of Prostheses Placed in Patients

	OD/RPD	FPP/SC	FCP	
Maxilla	9	4	0	
Mandible	14	4	3	
Total	23	8	3	

OD = overdenture; RPD = removable partial denture; FPP = fixed partial prosthesis; SC = single crown; FCP = fixed complete prosthesis.

- Biologic or technical complications related to the implants or prostheses
- Events related to the specific diseases and patients' conditions during the observation period

Prosthetic complications were classified according to previous reports^{21–23} as: (1) complications with the anchorage system related to the implant components, (2) repair of the prosthesis, or (3) adaptation of the prosthesis.

Statistics

Descriptive statistics were used for patients' demographics and diseases, number of implants and prostheses, and analysis of complications. A life table analysis with cumulative survival rate was calculated for all implants.

Results

Systemic Diseases, Abnormalities, and Congenital Defects

The congenital defects, genetic diseases, and special systemic diseases are summarized in Table 1 with the corresponding number of patients and implants placed. Table 2 summarizes all types of prostheses identified in the 24 patients. Table 3 gives detailed information on each patient who had their implants loaded. This

[‡]This patient had had a benign tumor removed from the maxilla.

Table 3 Overview of Patients

		Observation	No. of implants		Prosthesis type			
Disease	Sex	Age (y)	period (y)	Maxilla	Mandible	Maxillary	Mandibular	Complications
HED	F	37	12	0	4	FCP	FCP	Soft tissue hyperplasia
HED	M	33	12	0	3	RPD	OD-bar	Bony defects, hygiene, caries
HED	M	33	11	0	2	RPD	OD-bar	None
HED/GCG	M	23	3	6	4	FPP	FPP	Porcelain crack
CLP	F	55	7	2	4	OD-ball	FCP	None
CLP	M	54	7	6	0	OD-bar	Natural teeth	Screw loosening, bar fracture
CLP	F	63	6	4	2	OD-bar	OD-bar	Bar fracture, stomatitis
CLP	F	93	6	2	2	OD-ball	OD-bar	None
CLP	F	62	3	4	3	FPP	OD-bar	None
CLP	F	59	2	3	0	FPP	Natural teeth	Gingivitis
CLP	M	46	2	4	1	OD-bar	Single crown	None
CLP	F	54	2	5	0	OD-bar	RPD	None
DS	M	46	9	2	2	RPD-ball	OD-bar	Hygiene, gingivitis
DS	F	53	11	0	2	CD	OD-bar	Hygiene, gingivitis
DS	F	39	2	0	2	RPD	OD-bar	Hygiene
SS	F	63	2	3	1	FPP	Single crown	Caries in remaining teeth
SS/S	F	64	5	4	4	OD-bar	FPP	None
DMR	F	61	8	0	2	CD	OD-ball	Hygiene
DMR	F	59	2	0	3	OD-bar	OD-bar	None
CP	M	58	5	0	2	RPD	OD-bar	Hygiene
CL	F	78	5	0	2	CD	OD-bar	None
CL	F	74	2	0	2	CD	OD-bar	None
LP	F	74	6	0	4	CD	FCP	None
D-M	M	77	4	4	3	OD-bar	OD-bar/ball	Stomatitis, hyperplasia

HED = ectodermal dysplasia; GCG = giant-cell granuloma; CLP = cleft lip/palate; DS = Down syndrome; SS = Sjögren syndrome; S = scleroderma; DMR = mental retardation; CP = cerebral palsy; CL = chronic leukemia; LP = lichen planus; D-M = deaf-muteness; OD = overdenture; RPD = removable partial denture; FPP = fixed partial prosthesis; SC = single crown; FCP = fixed complete prosthesis.

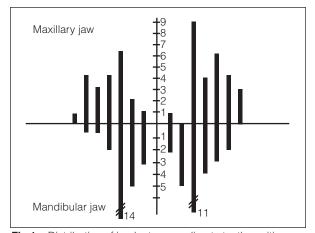


Fig 1 Distribution of implants according to tooth position.

Table 4 Life Table Analysis

Interval	No. of implants	No. of failures	No. replaced	No. of dropouts	SR (%)	CSR (%)
Healing	105	3	1	0	97.2	97.3
0-1 y	103	-	-	0	100	97.3
1-2 y	103	-	-	0	100	97.3
2-3 y	80	-	-	0	100	97.3
3-4 y	63	_	-	0	100	97.3
4-5 y	63	_	_	4*	100	93.4
> 5 y	53	-	-	0	100	93.4

SR = survival rate; CSR = cumulative survival rate. *One patient with 4 implants passed away.

overview includes the disease and defects, age of the patients, years of observation, number of implants, type of prosthesis in both jaws, and the most frequent complications registered during the entire observation period.

Survival of Implants

The survival rate of all loaded implants was 100%. The survival rate during the healing phase was 97.2%, including the patient with ALS. One implant exhibited

slight mobility 3 weeks after surgery in a patient with CLP. There were no clinical signs of inflammation or infection. This implant was removed and replaced in the same position immediately by a new one of a larger diameter to enhance primary stability. The implant healed uneventfully and remained stable also after loading by the overdenture. The location of all successfully 103 loaded implants is shown in Fig 1. One patient with a CLP dropped out from maintenance, since she passed away at the age of 93 years. However, radiographs were available from 2002 and all necessary informa-

Table 5 Prosthetic Complications

	Year and no. of patients per interval								
Complications	1 (n = 24)	2 (n = 24)	3 (n = 17)	4 (n = 15) 5	(n = 13)	6 (n = 11)	7 (n = 8)	8 (n = 6)	≥9 (n = 5)
Anchorage system									
Occlusal screw loosened	3	1	1	1	1	1			
Abutment loosening								2	
Abutment fracture									
Female part tightening	1		2			1			
Female part fracture				1			1		1
Female part replaced		2	1			1			1
Fracture of bar/extension	2			1				1	
Denture repair									
Framework fracture									
Porcelain cracks	1	2							
Denture fracture									
Teeth fracture	2								
New prosthesis			1						
New denture									
Denture adaptation									
Relining	1	3							
Occlusal adjustment	1		1						
Esthetic correction									
Sore spots	4						2		
Total	15	8	6	3	1	5	1	3	2

tion was obtained from the patient's chart. Taking into account that implants were lost in the healing phase, and including the patient who passed away, the cumulative survival rate after 5 years was 93.4%, as shown by Table 4.

Survival of Prostheses

The prosthesis plan was maintained in all patients, and they continued to regularly wear their original prostheses with the exception of 1 short-span fixed partial denture, which was remade. No prosthesis had to be changed or converted into another type of prosthesis as a result of implant loss or other problems, and the complication rate was rather low. Survival of prostheses, therefore, was 100%. Table 5 gives a brief overview of all prosthetic complications registered during the entire observation period and maintenance service performed. Relining of the overdentures became necessary for CLP patients for better closure of the obturator and to improve speech. In 2 of the CLP patients, a bar fracture was observed, twice in the same patient. Tightening of occlusal screws and female parts was more frequently performed than other maintenance services.

Special Complications

In spite of regular recall attendance, insufficient hygiene was found repeatedly in patients with DS, CP,

and DMR. Therefore, they often exhibited gingivitis and mucositis. This was also a problem in 1 patient with HED. This same patient had 2 teeth extracted in the course of the observation period, and he was also in need of endodontic treatment of a maxillary canine tooth. The most recent radiographs revealed angular defects around 3 intraforaminal implants after an observation period of 10 years. Supportive interceptive therapy with flap surgery, membrane placement, and filling of the defects with bone chips and bone substitute was planned. However, the patient did not consent to it. This behavior was in keeping with his rather poor compliance during the entire observation period.

Another patient with HED and GCG exhibited a porcelain crack on the central incisor of a fixed partial denture supported by 3 implants. This fixed prosthesis was replaced after 2 years of function. The radiograph of this same patient showed some progression of a GCG in the anterior mandibular bone that was very near an implant supporting a fixed partial denture. Recession and increases in probing depths were registered at the mesial side of the implant. It was decided—taking into consideration the young age of the patient—that he should remain under a strict observation protocol. There was a consensus between the patient, the family physician, and the dental clinician that a surgical revision of the anterior mandible may be necessary but should be postponed until a later time.

One patient with CLP and implant-supported overdentures in the maxilla and mandible repeatedly exhibited a maxillary denture stomatitis in spite of the denture being supported by 4 implants with a horse-shoe design. This means that most of the mucosa was not covered by the overdenture base. A fracture of the long anterior bar segment was further observed in this same patient. The fracture was not re-soldered and the bar was maintained in 2 separate lateral pieces, each with a short anterior cantilever segment.

One female patient with SS exhibited stable performance of all 4 implants supporting fixed partial prostheses and 2 single crowns. However, some of her teeth had to be filled and endodontic treatment provided, following progression of caries and enamel loss.

Discussion

The survival rate of the implants and prostheses was high in the present study. This may be ascribed to the regular maintenance service provided. The patients appeared to be aware of the beneficial effect of implant-prosthodontic treatment and therefore were very motivated to participate in the regular recall. No loaded implants were removed, and the cumulative survival rate after 5 years was reduced because of 4 implants that were lost in the patient who passed away. Thus, survival of ITI implants in this very mixed group was comparable to other reports on patients without special diseases.²⁴ However, from this study, final conclusions cannot yet be drawn regarding the process of osseointegration, bone remodeling, long-term implant stability, and specific diseases. Because of the small number of patients and implants, a comparison of the disease entities is not possible.

A variety of prosthetic reconstructions were placed in the patients of the present report, with a prevalence of overdentures. In the present study, the largest group with congenital diseases were the CLP patients, and treatment concepts for these patients have been previously discussed.²⁵ Maxillary overdentures were the most frequent indication for these patients. Maxillary overdentures can be stabilized by 4 implants, and in planned overdenture cases, they are rather successful.^{21,26} They compensate for anatomic and morphologic deficits and assist in the resolution of phonetic and/or esthetic impairment.27 Studies that compared maxillary fixed prostheses and overdentures in normal patients have shown equal or greater preference for removable prostheses by the patients.^{28,29} The oral situation is also complicated in patients with agenesis of teeth, aplasia of the jawbone, reduced quality of enamel, and malformed teeth, all of which are found in patients with HED and DS. 13,14,30 Similar to CLP patients, conventional prostheses had often been worn by these patients since early childhood. Therefore, these patients are at a high risk of losing their few remaining teeth by caries and periodontal disease. Mandibular 2-implant

overdentures were delivered to 2 of these patients and to 2 older women with chronic leukemia.³¹ One young HED patient with total agenesis of the primary teeth received a fixed prosthesis supported by 4 intraforaminal implants. A recent radiograph obtained 11 years after loading exhibited excellent bone stability around small-diameter implants. None of the HED patients had undergone bone grafting procedures, since they were still quite young, and it seemed more appropriate to postpone such invasive procedures to a later age.

The benefits and advantages of implant placement in conjunction with fixed prostheses are clear in patients with SS, who suffer from dry mouth, rapid development of caries, tooth decay, and sore soft tissue.32,33 Although it appears that the implant survival rate is lower, particularly during the healing phase, it was also demonstrated that crestal bone around the successfully loaded implants did not exhibit accelerated resorption, but remained stable after the first year.³³ The patient with SS and S in the present study received an implant-supported maxillary overdenture with a horseshoe design and extended bar support. Thus, this prosthesis was mostly implant-supported and not mucosasupported. Soreness of the mucosa in conjunction with removable prostheses and dry mouth sensation is also a problem in patients with erosive oral LP and S. While 1 study gave evidence of successful overdenture therapy with 2 ball attachments in the mandible in 2 edentulous patients with oral LP, for the single patient in the present study, comfortable and pain-free wearing of the prosthesis was achieved only when a fixed reconstruction was placed.15

Wearing complete dentures, particularly in the mandible, is difficult or impossible for patients with motor impairment (eg, CP) or reduced oral motor skills, as is often observed in patients with DS (large tongue) or DMR.11 Intraforaminal implants could provide the desired stability for the patients of the present study. Further, communication with these patients required special efforts and empathy.34 This was also the case with regard to the patient with D-M. When a good personal relationship was established between the clinician and these patients, they became highly motivated for treatment and regularly participated at the maintenance recall. In fact, this was important since the most frequent biologic complication was peri-implant mucositis and gingival inflammation resulting from insufficient home care. By means of professional support, this problem was controlled.

No typical pattern of prosthetic complication was observed, with the exception that service was more frequently necessary in the first year. Altogether, maintenance service that had to be provided for these patients was at a rather low level; prosthetic complications encountered corresponded to previous findings.^{21–23}

Conclusions

Unknown implications and possible risks for the process of osseointegration and remodeling in patients with such rare diseases and defects has led to a restricted application of implant treatment. However, from the present report it can be seen that implant therapy is highly beneficial for patients with specific diseases and defects. Implants can be successful if these patients are given continuous professional support. Reports on treatment outcomes in such patients may facilitate the process of patient selection in the future and can be helpful to expand the application of implants.

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