Implant-Supported Fixed Dental Prostheses in an Edentulous Patient with Dystrophic Epidermolysis Bullosa

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Purpose: This clinical report describes the use of implant-supported fixed dental prostheses (ISFDPs) in a severe case of dystrophic epidermolysis bullosa. ***Materials and Methods:*** The patient's appearance was characterized by reduced corporal growth and severe mutilation of the hands and feet. He was first examined at age 20. The severely decayed residual dentition had already been extracted by the time of examination. Conventional dentures were not possible due to severe microstomia and the fragility of the denture-bearing tissues. Even a modest manual touch was very painful and detached the epithelium of the oral mucosa. The first treatment was only possible under general anesthesia. To allow some prospect for oral rehabilitation, four implants were inserted in the maxilla and three in the mandible. Several years of steroid treatment had weakened the bony structures. Therefore, the diameter of the last drill used to prepare the bone for implant insertion was smaller than the diameter of the implants to improve primary stability. Complete FDPs with a shortened dental arch design served as superstructures. Several fractures in the screw-designed titanium abutments in the mandible necessitated insertion of three additional implants and an ISFDP with two occlusal units, which was screwed horizontally to a milled bar mesostructure. ***Results:*** Despite multiple fractures of the acrylic resin veneers caused by severe bruxism and the small occlusal surface, this rehabilitation proved successful until the patient died at age 25, as a consequence of his hereditary disease. ***Conclusion:*** This treatment greatly improved the patient's oral function, nutrition, and psychosocial well-being. *Int J Prosthodont 2010;23:42–48.*

Disturbances in dental and craniofacial development are common, and the risk of dental disease is often higher in individuals with a rare diagnosis. Oral rehabilitation in patients suffering from rare disorders is principally described in clinical reports of individual cases. Knowledge of dental implant treatment in cases of rare disorders is still limited since few reports have been published in the literature.[1,2] Despite a low level of evidence, the publication of case reports can help professionals choose more appropriate treatment methods. The present case illustrates the limits between what is advisable and what is possible for the oral rehabilitation of individuals with a severe disability or disease.

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Epidermolysis Bullosa

Epidermolysis bullosa (EB) comprises a group of genetic dermatoses mainly characterized by the occurrence of pressure-induced blisters. Three major types have been distinguished—simplex, junctional, and dystrophic—and numerous subtypes have been identified.[3] The three main forms are differentiated by the location of the ultrastructural cleavage. The most severe

form, the autosomal recessive dystrophic EB, is caused by a collagen defect with an inadequate development of hemidesmosomes in the papillary zone beneath the basal membrane of the skin and oral mucosa. Bullae lead to painful ulcers on rupturing, and healing causes cicatrization with atrophic scarring. Consequently, contractures over joints lead to adhesion between individual fingers and toes, resulting in pseudosyndactyly. Mutilation and the chronic need for care often lead to psychosocial problems in patients suffering from this disease. With an incidence between 1 to 50 cases in 1,000,000 live births, EB is a rare disease.[4] Whereas most junctional EB patients die during infancy or childhood, the majority of dystrophic EB cases live beyond the age of 20.[5]

Involvement of the oral mucosa is commonly found in all three forms of EB. However, oral debilitation from cicatrization of former blisters is most frequently found in the recessive dystrophic form.[6] Microstomia from such scarring and ankyloglossia are often accompanied by severe alveolar bone resorption with atrophy of the maxilla and prognathism of the mandible.[7–10] Severe periodontitis and rampant caries have been described as common clinical findings in young individuals with EB.[11,12] Dental involvement is also found in junctional EB, where abnormal tooth development, hypoplasia of the enamel, or both can occur.[13–16]

Dental management of EB has been described mainly on the basis of individual case reports.[17–24] Prevention has been proposed as the most effective means to arrest dental disease, although it is difficult to perform the correct preventive procedures without inducing blisters.[25] The use of sucralfate has been proposed to prevent oral blister formation and discomfort.[26] The use of dental implants in the oral rehabilitation of individuals with EB has been described in only a few publications to date. Peñarrocha-Diago et al reported on the placement of 15 implants (7 maxillary and 8 mandibular) to support overdentures in four patients with dystrophic EB.[27] Their preliminary findings after a mean follow-up time of 2.5 years (range: 1 to 4 years) were promising. Implant-supported fixed dental prostheses (ISFDPs) were placed in an edentulous 29-year-old patient with dystrophic EB.[28] They were placed on 4 implants each and had a shortened dental arch design. Eight months postinsertion, the metal framework of the mandibular ISFDP had fractured but could be repaired. Two fractured implant abutments had to be replaced. In 2007, Peñarrocha et al further reported on three dystrophic EB cases who were treated with ISFPDs in the maxilla and mandible; from the 27 implants placed, only 1 maxillary implant was lost.[29] In a more recent article, Peñarrocha and coworkers compared fixed and removable treatment concepts in six patients with dystrophic EB.[30] From the 21 maxillary and 17 mandibular implants placed in the edentulous arches, 97.9% were still in function 5.5 years postinsertion (range: 1 to 9 years). Improvements in comfort and retention, function, esthetics and appearance, taste, speech, and self-esteem were observed for both types or prostheses, yet the level of satisfaction was slightly higher in patients with ISFDPs.

Clinical Case Report

At age 20, the patient suffered from severe symptoms of dystrophic EB. He was the youngest of seven siblings of Turkish origin and the only case of EB in the family. His appearance was characterized by reduced corporal growth and severe mutilation of the hands and feet. Pressure-induced blisters covered his entire body except for his face (Figs 1 and 2). Over the years, he had been treated with high doses of steroids. Most parts of his body were permanently bandaged, except for the head, and he gave the impression of being mentally retarded. When the patient was first seen he was already edentulous, but archives from the Clinic of Dermatology, University of Mainz, Mainz, Germany, yielded information on the history of his decayed dentition (Fig 3). Compliance was difficult at first because even a moderate manual touch was very painful and detached the epithelium of the oral mucosa. Nevertheless, the patient expressed a wish for teeth so that he could gnaw chicken from the bone and eat his favorite food, potato crisps. Further, he desired "gold teeth" but was convinced that a single gold facet on a posterior tooth would be much more attractive.

Clinical examination revealed microstomia and very small edentulous ridges without blistering. The epithelium of the oral mucosa detached painfully on moderate touch. Conventional dentures were not possible because of the microstomia and the fragility of the denture-bearing tissues. ISFDPs were planned for the maxilla and mandible as the only possible oral rehabilitation. Under general anesthesia, four cylindrical screw-shaped IMZ TwinPlus implants (Friadent) were inserted in the maxilla and three in the mandible (Fig 4). Because of the weak bony structures, the diameter of the last drill used to prepare the bone for implant insertion was smaller than the diameter of the implants used to improve primary stability. In the maxilla, two 4.0-mm-diameter distal implants and two 3.3-mm-diameter anterior implants were placed, whereas the contour of the mandible limited the implant diameter to 3.3 mm. After 16 weeks of submerged healing, stage-two surgery was performed under general anesthesia, and conventional impressions and bite registration were made during the operation. ISFDPs with shortened dental arches of one occlusal unit were manufactured using screw-designed titanium abutments

Fig 1 *(left)* The patient suffered from a severe form of dystrophic EB with severely mutilated hands and feet. Blisters were distributed all over the body except for the face. (Courtesy of Prof Dr K. Bork, Mainz, Germany.)

Fig 2 *(top right)* Facial photograph of the patient before treatment. Note the edentulous profile.

Fig 3 *(bottom right)* The decayed dentition had been removed previous to the start of implant treatment. (Courtesy of Prof Dr K. Bork, Mainz, Germany.)

Fig 4 *(left)* Implant placement could only be performed under general anesthesia.

Fig 5 *(right)* The first ISFDP in the mandible was screwed directly to the titanium connectors. The microstomia allowed only vestibular access to the screws.

and a gold alloy framework with acrylic resin denture teeth. Limited mouth opening and vision necessitated access to the retention screws from the vestibule. The lingual position of the mandibular incisors, the result of prognathism, proved a mechanical disadvantage and led to fractures of the mandibular screw-designed titanium abutments four times within the first year of use (Fig 5). In three of four fractures, the remaining part of the titanium abutment could only be removed under general anesthesia. Finally, encouraged by the clinical stability of the implants, three more implants of the same diameter were placed in the mandible. Following insertion, the six mandibular implants were splinted using a resin bar for 6 weeks. The subsequent superstructure consisted of an ISFDP with two occlusal units, which was horizontally screwed onto a milled bar mesostructure because the limited space precluded access with a screwdriver for direct vertical screw retention (Figs 6a to 6c). The occlusal scheme was balanced in both ISFDPs.

Figs 6a to 6c **(a)** For the final restoration, four maxillary and six mandibular implants were used. **(b)** The mandibular ISFDP was screwed horizontally on a milled bar. **(c)** The patient had wanted a gold facet on the maxillary left first premolar.

Fig 7 *(left)* Oral hygiene could not be carried out by the patient due to severe mutilation of his hands.

Fig 8 *(top right)* The ISFDPs in situ. The incisal abrasion was caused by stress on the reconstruction due to severe bruxism on the limited occlusal surfaces.

Fig 9 *(bottom right)* Radiograph showing the restorations in place and the severe radiolucency of the bone due to years of cortisone treatment.

Although the tongue showed no severe ankyloglossia, it was not sufficiently mobile to wipe clean the mandibular vestibular sulcus. At irregular biannual intervals, the ISFDPs and titanium abutments were removed and cleaned (Fig 7). The considerable occlusal load resulting from the small occlusal surface and severe bruxism activity wore down the acrylic resin teeth and necessitated multiple repairs (Fig 8). Nevertheless, the maxillary ISFDP was successful for nearly 5 years, and the mandibular ISFDP for almost 4 years. All implants were clinically consolidated and radiographically osseointegrated (Fig 9). Periotest (Siemens) values after 3 years of use ranged from –5 to 1 (Table 1). Despite a Plaque Index of 100% and the absence of lingual and vestibular attached gingiva around the mandibular implants, the probing depth was 0. No peri-implant blisters were observed. The severely shortened dental arch did not lead to any symptoms of temporomandibular disorder and continued to satisfy the patient's initial desire to chew his favorite food.

The patient considered the ISFDP treatment to be a great success, with an essential gain in perceived quality of life, notwithstanding numerous treatment sessions and operations (Fig 10). He passed away at the age of 25 as a consequence of his hereditary disease.

Table 1 Periotest Values of Implants After 3 Years In Situ

	Periotest Value						
Implant site*	−5	−4	−3	−2	−1	0	1
14			X				
12							X
22							X
24					X		
45		X					
43			X				
41	X						
31			X				
33		X					
35							X

*FDI tooth-numbering system.

Fig 10 The gain in quality of life outweighed the costs and effort of this extensive, invasive treatment.

Discussion

High doses of steroids reduce the density of mandibular bone and have been shown to induce osteoporotic-like conditions in animal experiments.[31] Daily injections of glucocorticoids resulted in cortical thinning, irregular trabecular patterns, and impaired extracellular matrix formation and mineralization. These osteoporosis-like conditions significantly decrease the amount of bone contact with the implant surface.[32] Nevertheless, in this case, all implants were osseointegrated after 3 years and had mostly negative Periotest values, confirming good clinical stability. No translucencies or peri-implant bone loss were visible on the radiographs. Despite the abundant plaque due to the compromised oral hygiene measures, no clinical peri-implant inflammation or pockets were present. A particular saliva composition might have precluded the formation of calculus; previous systemic EB medication may have helped to avoid a severe inflammatory response.

Drilling undersized holes for the implants allowed primary stability to be achieved despite the poor bone quality. Lack of subsequent complications provokes interest in exemptions from the generally accepted aim of creating a passive fit of an implant to cut bone surface.

Fixed reconstructions were chosen to avoid any contact with the alveolar mucosa. Peñarrocha-Diago et al[27] chose to provide their four EB patients with implant-supported overdentures to facilitate cleaning, to keep their options open in case of implant loss, and because of a lack of alveolar bone. However, they reported frequent mucosal ulcerations in the areas in contact with the overdentures. Later, this group of authors also successfully applied fixed implant-supported treatment concepts and reported slightly higher patient satisfaction.[29,30] In the present case, removable dentures were not an option. Severe microstomia would have precluded the insertion of a full palate maxillary denture and the oral tissues seemed much too delicate mechanically. However, the patient would have benefited from a full dental arch with sufficient occlusal surfaces to allow normal masticatory function—especially since swallowing problems with esophageal stenoses and ulcers are commonly reported phenomena in dystrophic EB. The first version of the ISFDP had one premolar occlusal unit and the second version had two premolar occlusal units. The ISFDPs were not cantilevered further distally because of a lack of experience with the reduced bone density. The prognathism of the mandible was already proving to be a mechanical disadvantage for the mandibular ISFDP because the incisors were placed lingual to the implants. This nonaxial load presumably contributed principally to the frequent fractures of the screw-designed titanium abutments. Lee et al were equally limited to the insertion of ISFDPs with only four implants and premolar occlusion due to limited access and posterior occlusal space.[28] Abutment fractures and a fracture of the metal framework also occurred in their reported case.

On the other hand, the 3.3-mm diameter of the mandibular implants was a weak point in the reconstruction. The first attempt with the mandibular ISFDP aimed to provide an esthetic and minimally functional reconstruction, given that no alternative prosthetic means were to be considered. However, the occlusal forces on the ISFDPs were initially underestimated; they were only disclosed postinsertion by the substantial incisal abrasion in the second ISFDP shown in Fig 8. Dystrophic EB is often associated with muscular dystrophy, and the physiognomy of the patient did not imply marked bruxism. At decision making, no information on wear and facets on a former dentition were available. However, daytime bruxism and parafunctional activities have been reported to be common in individuals with cognitive disabilities.[33,34]

Meticulous care was taken not to introduce any sharp angles into the oral cavity that might cause blistering or ablation of the oral epithelium. During the osseointegration period, even the rounded healing caps of the maxillary 4-mm IMZ TwinPlus implants had caused corresponding ulcers on the tongue. However, despite the sharpened incisal edges, only minor corresponding blisters or lesions were observed during the wearing period.

Implant placement and the removal of the fractured screw remnants from the fractured titanium abutments were performed under general anesthesia. Intubation of patients with dystrophic EB is very difficult and presents considerable risks because of common esophageal stenoses and ulcers.[35,36] The patient's mental retardation, as well as many years of hospital experience, did not allow for treatment under local anesthesia at first. However, over years of treatment, the patient gained confidence and was much more accepting to even painful treatment procedures. On the other hand, the administration of local anesthesia is also associated with risks. If not deposited deeply enough in the vestibule, the injection could create blisters and, by subsequent scarring, accelerate the elimination of the vestibular sulci.

Finally, some ethical issues must be addressed. Certainly this case describes a borderline indication for ISFDPs. The cost of the six operations and three precious-alloy ISFDPs on 10 implants was fully covered by public resources. The treatment was invasive, drawn out, and at times painful for the patient. *Primum nil nocere* is one of the paradigms in medicine. Considering the lower life expectancy of individuals with severe dystrophic EB, did the benefit for the patient outweigh the costs, pain, time, and labor? In his view, yes! As described by Peñarrocha-Diago et al,[27] the ISFDPs allowed the patient to progress from eating mashed and liquid food to solid foods. Further, he liked the appearance of the ISFDPs and was particularly proud of the gold facet on the maxillary left first premolar.

Conclusion

The oral rehabilitation of this patient with dystrophic EB presented a considerable challenge. With the ISFDPs, the patient perceived an essential gain in quality of life by improving both esthetic and functional aspects, and thus allowing mastication for the first time since the loss of his natural teeth. Despite the poor bone quality, clinical difficulties, poor oral hygiene, and frequent repairs, these ISFDPs were functionally successful until the patient passed away as a consequence of his hereditary disease.

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Literature Abstract

Strain development in 3-unit implant-supported CAD/CAM restorations

This study evaluated whether fixed partial dentures (FPDs) made from optical impressions lead to less strain development than conventionally fabricated FPDs. A measurement model with two solid Straumann implants was set up and strain gauges were attached to the model material adjacent to the implants. Two groups of conventional cementable FPDs (repositioning and pick-up impressions) and a group of computer-aided design/computer-assisted manufacture (CAD/CAM)-generated FPDs (optical impressions) were fabricated (n = 10). Strain development during FPD fixation was recorded. Multivariate analysis of variance was performed. None of the FPDs revealed a true passive fit. The mean strain development at the different strain gauge locations ranged from 80.38 µm/m to 437.11 µm/m. The two groups of conventionally fabricated FPDs showed no significant difference. The CAD/CAM group revealed a significantly lower strain development than those made from the pick-up impressions. No significant difference could be detected between the FPDs manufactured from the repositioning technique impressions and the CAD/CAM. This study showed that restorations fabricated with optical impressions demonstrate a level of fit that is at least as passive as that of conventional FPDs. Nonetheless, this is an in vitro study that allowed for optimum scanning of the models. Moreover, these were cement-retained samples, where passive fit is of less importance than with a screw-retained restoration.

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