

CLINICAL RESEARCH

Digital scanning for implant-supported fixed complete-arch dental prostheses for patients with epidermolysis bullosa: A case series evaluation



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Epidermolysis bullosa comprises a group of hereditary disorders characterized by mechanical weakness of the skin and mucous membranes, with the recurrent appearance of aphthae, blisters, and vesicles in response to minimum tissue friction.¹⁻³ The incidence of epidermolysis bullosa, also known as “skin of glass” disease, is 5 cases per 100 000 inhabitants.⁴⁻⁷ The disorder is classified into 3 groups (simplex, junctional, and dystrophic) and 25 subtypes, of which the recessive dystrophic epidermolysis bullosa (RDEB) presents with the greatest oral mucosa involvement.¹⁻⁵ RDEB is characterized by the presence of blisters all over the body, particularly in areas exposed to friction such as the mouth, hands, feet, elbows, and knees. It is a rare genetic disorder that affects the skin and mucous membranes, in which blisters form either spontaneously or in response to minimal mechanical trauma.^{1,2,8,9} The most common oral manifestations of epidermolysis bullosa include intraoral

ABSTRACT

Statement of problem. The treatment of patients with recessive dystrophic epidermolysis bullosa has been compromised in the past by the lack of oral therapeutic information and the use of conventional complete dentures.

Purpose. The purpose of this clinical case series study was to describe a digital rehabilitation protocol involving computer-aided design and computer-aided manufacturing (CAD-CAM) techniques for the treatment of patients with recessive dystrophic epidermolysis bullosa and to follow up for 4 years the patients who underwent this treatment.

Material and methods. A case series analyzing implant survival, peri-implant tissue health, and patient satisfaction with the treatment received was made of 4 patients with recessive dystrophic epidermolysis bullosa. Bimaxillary fixed implant-supported complete-arch rehabilitation was carried out by using a digital protocol with CAD-CAM techniques.

Results. The implant survival rate was 100%, with a bleeding rate of 74.2% and an inflammation rate of 58.0%. Gingival stability was achieved in 77.4% of the patients, with gingival displacement in 22.6% of the implants. However, the probing depth was maintained between 1 and 3 mm in 96.7% of the implants placed.

Conclusions. Fixed complete-arch implant-supported rehabilitation is a successful treatment for patients with epidermolysis. Digital intraoral scanning facilitates the treatment of patients with this condition and reduces the clinical complications associated with conventional impression techniques (ulceration, blistering, angular cheilitis, and so forth). (*J Prosthet Dent* 2019;122:364-70)

ulcerations and the formation of bullae, severe periodontal disease and alveolar bone resorption, ankyloglossia, tongue atrophy, elimination of buccal and vestibular sulci, lingual depapillation, atrophy of the palatal folds, and microstomia (Fig. 1). Affected patients typically present syndactyly (Fig. 2) because of the continuous friction to the hands in daily life, together

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Clinical Implications

The digital workflow in the planning and design of the implant-supported prosthetic rehabilitation of patients with recessive dystrophic epidermolysis bullosa simplifies the process and reduces the clinical complications associated with conventional complete dentures.

with esophageal strictures causing digestive tract obstruction and consequently dysphagia.^{1-3,5} An increased incidence of oral carcinoma has also been reported in these patients.^{1,4,6,10}

The treatment of patients with epidermolysis bullosa using conventional removable prostheses frequently results in mucosal blister formation caused by friction of the dentures on the mucosa. In individuals with this rare disease, rehabilitation with fixed implant-supported prostheses should reduce the possibility of mucosal ulcerations, although the prosthodontic management of such patients can be complex.³⁻⁶

In the past, the treatment of these patients with conventional complete dentures has been compromised by the lack of oral therapeutic information. Subsequently, the first set of patients treated with implants was rehabilitated with fixed cemented prostheses due to angulation of the implants and the limited oral aperture that precluded delivery of a screw-retained prosthesis. However, the development of computer-aided design and computer-aided manufacturing (CAD-CAM) technology has facilitated the passive fit and correct angulation of implants placed in difficult sites. Nevertheless, descriptions of the use of such technology in patients with epidermolysis bullosa are sparse.¹¹

Intraoral scanners generating a standard tessellation language (STL) file have made it possible to improve patient management, constituting the first step in the digital workflow of making restorations on teeth and implants.¹²⁻¹⁷ The purpose of this clinical case series study was to describe the treatment of patients with RDEB with bimaxillary fixed implant-supported complete-arch rehabilitation using a digital protocol with CAD-CAM techniques.

MATERIAL AND METHODS

Four patients with RDEB were treated with implants in the dental clinic of the University of Valencia between September 2013 and December 2014. A clinical case series study was made, analyzing implant survival, peri-implant tissue health, and patient satisfaction with the treatment received.

The inclusion criteria were patients diagnosed with RDEB, who are edentulous or partially edentulous, and



Figure 1. External view of limited oral opening.

for whom removal of any remaining teeth was indicated. In compliance with the principles of the Declaration of Helsinki, written informed consent was obtained for fixed implant-supported or removable implant-retained rehabilitation and a minimum of 1 year of follow-up after loading.

Panoramic radiographs and maxillary cone beam computed tomography scans (Promax 3D; Planmeca) were obtained (Fig. 3). The surgical protocol has been described in previous publications.^{1,18-20} The lips of the patient and the surgical instruments were lubricated with petroleum jelly before surgery to minimize the risk of blister formation and lesions caused by friction with the mucosa.²¹ Surgery in all 4 patients was performed under sedation, with 1% propofol (Diprivan) administered by an anesthetist. Infiltrating local anesthesia with epinephrine (Ultracain; Normon) was provided in the treatment zone. The implants were placed using a combination of drills and osteotomes to preserve the remaining bone and facilitate primary stability. At the mandibular level, conventional drilling was used with the minimum required saline solution irrigation to minimize aspirator use, which could cause friction damage. Furthermore, when the aspirator was used, it was always placed in contact with bone, not soft tissues. Tissue-level, internal connection implants (TSA; Phibo Dental Solutions) were placed. The bone defects adjacent to the implant created during surgery were filled with autologous bone particles collected from the surgical drills during preparation of the implant beds. Beta-tricalcium phosphate (KeraOs; Keramat) was also used as a bone-grafting material in those patients in whom the autologous bone was insufficient, and resorbable collagen membranes (Lyostypt; B. Braun) were used to protect the bone grafts. Panoramic radiographs were obtained immediately after surgery, and oral antibiotic treatment (amoxicillin 500 mg every 8 hours for 7 days) and a nonsteroidal anti-inflammatory medication were prescribed (ibuprofen 600 mg every 8 hours for 3 days).

The implants remained submerged without loading during an osseointegration period (3 months in the



Figure 2. Clinical view of lesions on hands.

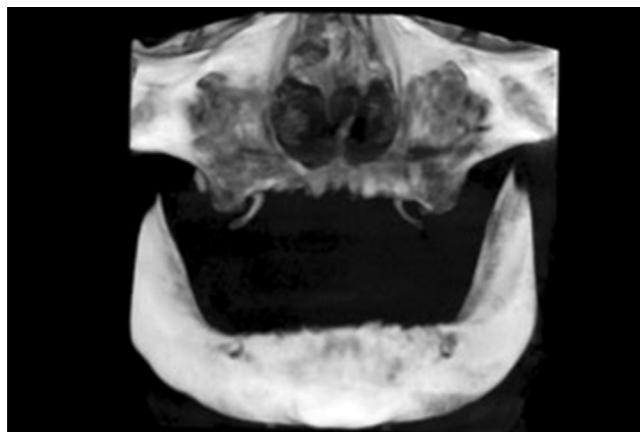


Figure 3. Pretreatment cone beam computed tomography image.



Figure 4. Intraoral scanning of implants.

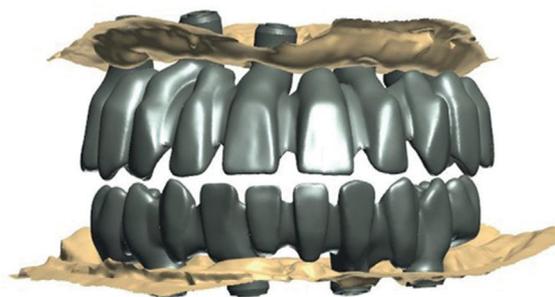


Figure 5. Virtual design of bimaxillary metal-ceramic fixed implant-supported complete-arch prostheses.

mandible and 6 months in the maxilla) before a conventional second stage surgery. During the osseointegration period, a conventional removable prosthesis was provided, but only for esthetic purposes. The immediate removable provisional prosthesis was lined with tissue conditioner (Visco-Gel; Dentsply Sirona) and replaced every 2 weeks to avoid mucosal ulcerations.

Digital scans were obtained 2 weeks after second stage surgery by using an intraoral scanner (TRIOS; 3Shape A/S) according to the protocol of the manufacturer. Both arches were scanned without scan bodies to register the tissues, and then scanning was repeated with scan bodies to define the 3D position of the prosthetic platform of the implants. After scanning the arches, occlusion and vertical dimension were recorded with the following technique. With all the scan bodies screwed into both arches, the patients were instructed to establish occlusion; later, this occlusal position was fixed with occlusal registration material (Occlufast Rock; Zhermack SpA). Posteriorly, this intermaxillary relationship was digitally scanned, and a best-fit function was used to correlate the anteroposterior and apical-coronal relations of the maxillary

and mandibular digital scans. A facebow record was made to transfer the maxillary relationship to a conventional analog articulator (Artex; Amann Girrbach).

CAD-CAM techniques were used to fabricate the fixed complete-arch implant-supported prosthesis. Based on the STL files obtained by the intraoral scan (Fig. 4), a CAD tool (Fig. 5) (3Shape CAD Design Software) was used to design the direct-to-implant screwed structures made of cobalt-chromium alloy (Adhoc; Phibo Dental Solutions) (Fig. 6). Adaptation of the frameworks was evaluated, and the prostheses were completed after placement of the ceramic veneer (IPS d.Sign; Ivoclar Vivadent AG). The prostheses were placed after assessing occlusion and esthetics by inserting the screws from the buccal side because of the limited oral openings of the patients. Angled screws (Axis; Phibo Dental Solutions) were used for this purpose (Fig. 7) and were inserted at an angle of 25 degrees and tightened to 20 to 25 Ncm (Fig. 8).

The patients were monitored 1 and 3 months after surgery and every 6 months after prosthetic loading (Fig. 9). Panoramic radiographs were obtained every 12

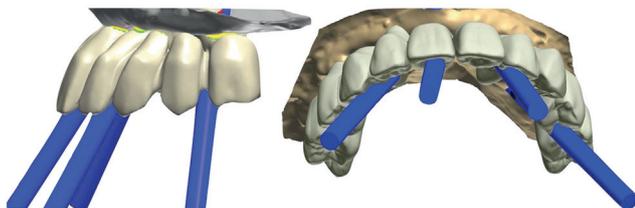


Figure 6. Computer-aided design with screw access to buccal side.

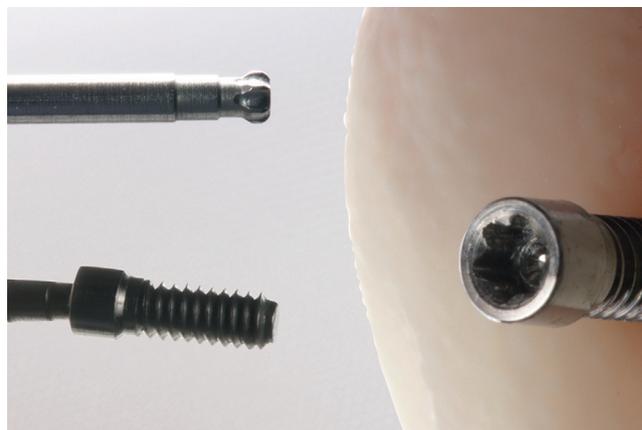


Figure 7. Screwdriver with screw of hexalobular design.



Figure 8. Operator placing screw in anterior implant.



Figure 9. External view of smile.

months to evaluate bone loss (Fig. 10). Intraoral periapical radiographs were contraindicated because of friction between the film and the oral mucosa. Some of the patients were unable to return for all the monitoring visits because of physical limitations or because they lived in other cities or countries.

On the last monitoring visit, the patients completed a general satisfaction questionnaire and a questionnaire assessing the impact of treatment on quality of life. The general satisfaction questionnaire scored parameters such as comfort, esthetics, and speech on a scale from 0 to 10 (0 = “not at all satisfied” and 10 = “totally satisfied”). The quality of life questionnaire comprised questions referring to the impact of treatment on mood and daily activities such as eating, speech, work, and social relations. The possible answers ranged from “not at all” to “a lot.”

Implant survival and peri-implant tissue health corresponding to each patient was also recorded during the final follow-up visit. The assessment of implant survival was based on implant mobility, presence of exudate, persistent inflammation (assessed by visual inspection of redness of the gums), discomfort, bleeding, and periapical radiolucency (determined from panoramic radiographs).



Figure 10. Panoramic radiograph 6 months after treatment.

Peri-implant tissue health was evaluated based on the presence of gingival displacement measured in millimeters by using a periodontal probe (PCPUNC156; Hu-Friedy), the presence of keratinized mucosa (determined as present or absent by visual inspection), the presence of plaque (determined as present or absent by visual inspection), the

Table 1. Implant survival and outcomes (%)

Location	N	Mobility	Exudate	Inflammation	Discomfort	Bleeding	Radiolucency
Maxilla	17	0	0	32.2	0	38.7	0
Mandible	14	0	0	25.8	0	35.5	0
Total	31	0	0	58.0	0	74.2	0

Table 2. Peri-implant soft tissue health: gingival displacement and keratinized gingival tissue

Location	N	Gingival Displacement (%)			Keratinized Gingival Tissue (%)	
		0 mm	1-3 mm	>3 mm	No	Yes
Maxilla	17	38.7	12.9	3.2	32.2	22.6
Mandible	14	38.7	6.4	0	29.	16.1
Total	31	77.4	19.3	3.2	61.2	38.7

presence of bleeding (determined as present or absent by visual inspection), and maximum probing depth measured in millimeters by using a periodontal probe (PCPUNC156; Hu-Friedy).

RESULTS

A total of 31 implants were placed (17 in the maxilla and 14 in the mandible) in 4 patients (3 women and 1 man) diagnosed with RDEB who were of age between 20 and 52 years. Implant rehabilitation was based on a delayed protocol for all patients. Eight fixed screwed implant-supported prostheses were made.

The implant survival rate after 4 years was 100%. High bleeding (74.2%) and inflammation (58.0%) rates were recorded (Table 1). For the peri-implant soft tissue outcomes, 77.4% of the implants showed no gingival displacement. The remaining 22.6% presented gingival displacement (Table 2).

A total of 51.6% of the implants presented plaque at the time of the evaluation. The percentage was greater in the maxilla than in the mandible. The incidence of bleeding was also high (74.2%). Probing depth was maintained in the range of 1 to 3 mm in 96.8% of the implants placed (Table 3).

For the general satisfaction questionnaire, the mean score was over 9 for all the parameters evaluated (eating, speech, esthetics, comfort, and self-esteem) except hygiene (with a score of 6 to 8) (Table 4).

Positive results were recorded from the questionnaire exploring quality of life after treatment. None of the patients felt ashamed of their mouth after treatment, and 3 of the 4 patients did not experience problems carrying out their daily activities (Table 5).

DISCUSSION

Treatment with fixed prostheses supported by implants in edentulous patients with RDEB restored masticatory

Table 3. Peri-implant soft tissue health: presence of plaque, bleeding, probing depth

Location	N	Plaque (%)		Bleeding (%)		Probing Depth (%)	
		Absence	Presence	Absence	Presence	1-3 mm	>3 mm
Maxilla	17	25.8	29.0	16.1	38.7	51.6	3.2
Mandible	14	22.6	22.6	9.7	35.5	45.2	0
Total	31	48.4	51.6	25.8	74.2	96.8	3.2

Table 4. Patient satisfaction with treatment received

Score	General						
	Satisfaction	Eating	Speech	Esthetics	Hygiene	Comfort	Self-esteem
0-4							
5			1				
6					2	1	
7	1				1	1	1
8	1			1	1		1
9	1	1	1	1			
10	1	3	2	2		2	2

function and avoided or delayed the development of esophageal strictures. The patients were able to swallow solid food masticated by using the occlusal surfaces of the fixed implant-supported prostheses.¹⁻⁵ Because the prostheses were fixed to the implants and did not undergo vertical, anteroposterior transfer or rotational movements, induction of pressure and ulceration of the oral mucosa were avoided—this being one of the complications of the treatment of these patients with removable mucosa-supported dentures.^{1,2} Rehabilitation with fixed prostheses improved the quality of life of the patients, who were now able to swallow solid foods. When masticatory function is lost, ingestion is limited to liquids, with the possible risk of poor nutrition and impaired general health. Chronic intake of liquid foods and the consequent lack of stimulation of the digestive tract can lead to esophageal stricture.^{3,4} This in turn precludes the ingestion of any food and may prove life-threatening if surgery is not quickly performed to increase the diameter of the esophagus.¹⁻⁵

As general anesthesia poses the problem of possible ulcerations resulting from intubation, the implants were placed under local anesthesia and intravenous sedation. When intraoral local anesthesia is administered, the anesthetic solution should be injected deeply into the tissues at a rate slow enough to prevent tissue distortion, which might cause tissue separation and blistering in patients with epidermolysis bullosa.^{6,7}

Table 5. Patient quality of life

Quality of life questionnaire topics	Not at All	Very Little	Occasional	Quite a Lot	A Lot
Speech problems	1	3	-	-	-
Unpleasant taste in mouth	3	1	-	-	-
Pain	2	-	2	-	-
Eating difficulties	4	-	-	-	-
Concern about mouth	2	-	1	1	-
Tenseness because of mouth	1	1	1	-	1
Dissatisfaction on eating	4	-	-	-	-
Meal interruptions	4	-	-	-	-
Nervousness about mouth	3	-	1	-	-
Feeling ashamed about mouth	4	-	-	-	-
Irritable because of mouth	1	3	-	-	-
Difficulties at work	3	1	-	-	-
Dissatisfaction with life	3	1	-	-	-
Unable to lead normal life	3	-	1	-	-

Implant surgery poses an additional problem of considerable tissue irritation and resulting bullae formation because of contact between the aspirator and oral mucosa. Wright et al^{3,4} indicated that lubricating the lips of the patient and any tissue susceptible to contact also decreases the likelihood of shear forces and tissue damage. The lips were therefore abundantly and repeatedly lubricated with petroleum jelly.

The patients in the case series presented limited oral opening. An intraoral scanner can be used in such situations to obtain a digital scan. This procedure is associated with increased patient satisfaction.¹⁴ The use of these devices in the first phase of rehabilitation treatment can improve the management of these patients because the conventional impression procedure produces friction with the mucosa and generates intraoral ulcerations. With the intraoral scanner, the operator avoids friction of the intraoral mucosa in a more efficient way than with conventional impression techniques.

The scans of both arches were made by using a powder-free intraoral digital scanner based on ultrafast optical scanning technology. The accuracy of the scanning in patients with this condition depends on different factors, including the type of intraoral scanner used, the distance between implants, their angulation, the depth of the implants, the experience of the operator, the size of the scanbodies, and the type of software used.^{12-14,16} In these patients, because the implants were positioned in the anterior sector, the distances between implants did not exceed 10 mm, thereby making it possible to reduce the error introduced in each best-fit alignment.

The prosthesis used in these treatments was a screw-retained direct-to-implant complete-arch rehabilitation designed and fabricated with CAD-CAM technologies. A screwed prosthesis was used because it allows better maintenance of implants, as disassembly is easy. The first treatments of implant-based rehabilitations in patients with epidermolysis bullosa involved the use of cemented

fixed prostheses as it was not possible to adopt a screwed prosthesis design because of the limited oral opening (the driver could not be inserted vertically into the screw access chimney). At present, CAD-CAM technology is widely applied to design and manufacture of implant frameworks, facilitating the access channel with dynamic solutions using hexalobular screws (Axis; Phibo dental Solutions). In this way, it is possible to screw the prosthetic structure from a vertical position, correcting the screw emergence angle up to 25 degrees.

The reported implant success rate in patients with RDEB varies between 97% and 100%.^{9,20,22} Descriptions are sparse regarding the peri-implant tissue outcomes in patients with this condition. In this clinical case series study, the peri-implant soft tissues were evaluated by measuring gingival displacement, the presence or absence of keratinized gingival tissue, the presence or absence of plaque and bleeding, and the assessment of probing depth after 4 years of follow-up. The results obtained were positive; however, the plaque and bleeding rates were high (51.6% for plaque and 74.2% for bleeding) and could be explained by the difficulty in oral hygiene maintenance that these patients face because of their microstomia, syndactyly, ankyloglossia, and the lesions caused by brushing.^{10,19,21} The use of small soft brushes adapted for patients with physical limitations and chlorhexidine rinses is recommended for these patients.²¹

For patient satisfaction and quality of life, high scores between 9 and 10 were recorded for most of the items of the questionnaire, except for satisfaction with oral hygiene because the physical limitations caused by the disease and the mucosal sequelae of brushing greatly complicate maintenance of adequate hygiene. These observations are consistent with those of other studies.^{19,21}

The limitations of this clinical case series study are the small sample size (4 patients) and the lack of homogeneity in complying with the monitoring and maintenance visits; some of the patients lived in other cities or even other countries. Furthermore, their physical limitations (wheelchair or treatment in the form of dialysis) also represented a problem in this regard.

CONCLUSIONS

Based on the findings of this case series, the following conclusions were drawn:

1. Complete-arch implant-supported rehabilitation is a good option for patients with epidermolysis bullosa because of the high success rate (100%) and correct maintenance and the improvement of patients' self-esteem and quality of life (general satisfaction mean score of 9 and positive results in the quality of life questionnaire).

- The digital workflow in the planning and design of the implant-supported prosthetic rehabilitation of patients with RDEB simplifies the process and reduces the clinical complications associated with the conventional methodology (friction aphthae caused by the impression cuvettes, lacerations, lip angle tearing, and detachment of the oral mucosa). Thanks to CAD-CAM designing of the prosthesis, positioning the access channels of the fixation screws can be planned with the screws angled from a buccal and extraoral position, thereby facilitating the screw-retained rehabilitation of these patients with limited oral openings.

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