Case Report

Dental implants for severely atrophied jaws due to ectodermal dysplasia

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ABSTRACT

The aim was to present the successful esthetical and functional rehabilitation of partial anodontia in a case of severe ectodermal dysplasia with complete atrophy of the jaws. A 17-year-old male with Class III malocclusion with partial anodontia sought dental implant treatment. His expectation was that of Class I occlusion. The challenge in the case was to match the expectation, reality, and the clinical possibilities. Ridge augmentation was performed with a combination of rib graft and recombinant human bone morphogenetic protein-2. Simultaneously, 6 implants (Nobel Biocare™ - Tapered Groovy) were placed in maxillary arch and 10 in the mandible. Simultaneous placement ensured faster and better osseointegration though a mild compromise of the primary stability was observed initially. After adequate healing, Customized Zirconia Procera™ system was used to build the framework. Zirconia crown was cemented to the framework. Radiological and clinical evidence of osseointegration was observed in all 16 dental implants. Successful conversion of Class III to Class I occlusion was achieved with the combination of preprosthetic alveolar ridge augmentation, Procera™ Implant Bridge system. Abnormal angulations and or placement of dental implants would result in failure of the implant. Hence conversion of Class III to Class I occlusion needs complete and complex treatment planning so that the entire masticatory apparatus is sufficiently remodeled. Planning should consider the resultant vectors that would otherwise result in failure of framework or compromise the secondary stability of the dental implant during function. A successful case of rehabilitation of complex partial anodontia is presented.

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Ectodermal dysplasia (ED) is a spectrum of a group of syndromes characterized by the anomaly of ectodermal structures. Mutation in the genes that allow interaction between the mesoderm and ectoderm for proper formation of the ectodermal structures is often inherited by the affected individual through various inheritance patterns.[1] The most common feature of ED syndrome is characterized by hypohidrosis, hypotrichosis, and partial anodontia. The head and neck changes include abnormal enamel and dental deposition which predisposes to early loss of dental structures.[2]

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In several instances, due to improper ectodermal-mesodermal interaction, certain growth center fails to develop properly. This often leads to an erroneous maxilla-mandibular relationship. Patients with ED opting for permanent dental rehabilitation presents with major challenges like atrophied maxillary and mandibular alveolar ridges, collapsed bite, and conical shaped teeth. The aim of this report is to present a comprehensive, complete, multidisciplinary approach to rehabilitation of a teenager with ED.

CASE REPORT

A 17-year-old male sought dental implant treatment; his expectation was to achieve a normal smile. The past dental

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and medical history revealed that he had been diagnosed with hereditary hypohidrotic ED since his early childhood. On extraoral examination, he had dry skin, sparse thin hair on the scalp and eyebrows, prominent lips, and mandibular prognathism [Figure 1a]. Intraorally, partial anodontia, decreased vertical height, and Class III malocclusion were noted. The permanent central incisors, lateral incisors, premolars, first molar, and mandibular 2nd molar on the left side were missing. The maxillary and mandibular canines were abnormally conical in shape. The retained maxillary deciduous molars were sound in endodontic and periodontic evaluation. The mandibular canines had compromised periodontal support. On intraoral palpation, the edentulous area had reduced residual alveolar height and residual alveolar width (RAH and RAW, respectively) due to functional alveolar atrophy [Figure 1b].

The radiographic panoramic view was used to assess the requirement of the alveolar ridge augmentation and for the evaluation of the teeth present. An Orthopantomograph (Kodak-Trophy, 77437 Croissy-Beaubourg-France, Eastman KODAK company; Model: KODAK 8000 C, Carestream Health, Inc., Sistema Rayos X Digital Extraoral; Mfg 2006/11) was obtained and in 1:1 magnification, the RAH were measured [Figure 2]. The RAH in the maxillary anterior region, preoperative position of the central incisor region was about 5 mm, at that of the lateral incisor region was about 6 mm, and at premolar region was about 14 mm. In the mandibular anterior region, preoperative position of the incisors, the RAH was about 6 mm, in the canine region it was about 7 mm, and in the premolar and molar region it was about 4 mm. Supra eruption of the mandibular canines was also noted. The RAW was measured with a Vernier Caliper in a diagnostic maxillary and mandibular casts made with an alginate impression (ADA Standard Number 18. ISO 1563:1990).[3] The measurements have been tabulated in Table 1.

The treatment plan envisioned was alveolar ridge augmentation to increase the RAH and RAW, by autogenous bone grafting for both maxillary and mandibular arches followed by implant placement in both the jaws.

Alveolar ridge augmentation and implant placement

Under nasotracheal intubation for general anesthesia, through an inframammary incision the superior portion of the pectoralis major was dissected, the muscle was divided and the costochondral graft (6th rib) was harvested. The surgical site was filled with saline solution and positive pressure ventilation was performed to rule out any leak. The incision was closed in layers and the skin layer was sutured. Through an incision on the maxillary and mandibular alveolar ridge, the mandibular canines were extracted followed by alveoloplasty of the ridge with a round bur. The costochondral graft was used in an onlay grafting technique which was fixed with 8 mm titanium screws. The simultaneous placement of 16 titanium dental implants was performed (6 in the maxillary arch and 10 in the mandibular arch) [Figure 3a and b]. To ensure better and faster osseointegration recombinant human bone morphogenetic protein (rhBMP-2) was used in the grafted site before closure of the incisions [Figure 3c]. The implant torque level was approximately 30 N/cm for all fixtures. The implant measurements have been tabulated in Table 2. Closure of flap was done in layers. Standard antibiotics (ciprofloxacin 500 mg, metronidazole 400 mg) and appropriate nonsteroidal anti-inflammatory drugs (diclofenac sodium 50 mg) were given to control infection and pain. Standard oral hygiene instructions were provided.

Healing phase

A temporary removable denture was rehabilitated for the healing phase of the onlay bone grafting and for osseointegration. After 6 months of healing phase the patient was reviewed, the patient had not suffered symptoms like pain, swelling or fluid discharge, during the phase of healing. Re-measurement of the alveolar ridges was done and a good take up of the graft and the implants was illustrious in the 6th month postoperative panoramic radiograph [Figure 4].

Prosthodontic rehabilitation

An incision on the alveolar ridge was made, to expose the implant. The healing abutments were inserted into the implants to facilitate the formation of the gingival cuff. An acrylic open tray was fabricated to make the final impression. The impression coping was firmly fixed to the implants, stabilized with dental floss and pattern resin, to prevent distortion during the removal of the impression tray. The

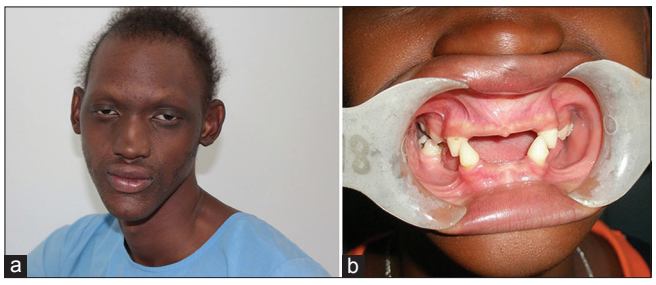

Figure 1: (a) Preoperative profile of the patient. (b) Preoperative intraoral view showing partial anodontia and cone shaped canines

Figure 2: Preoperative orthopantomograph atrophic maxillary anterior alveolus and mandibular anterior and posterior alveolus

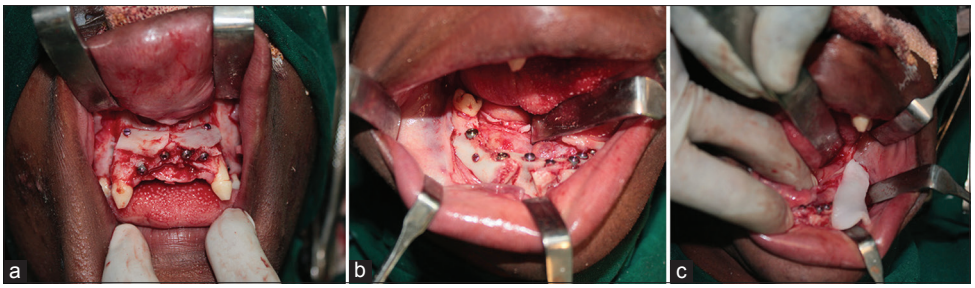

Figure 3: (a and b) Intraoperative view of maxillary and mandibular simultaneous alveolar ridge augmentation with costochondral graft and placement of titanium dental implants. (c) The use of recombinant human bone morphogenetic protein-2 shown in the mandibular ridge

Table 1: Preoperative and 6th postoperative month measurements of the RAW and RAH

Site	Side	Tooth region	Preoperative		6th postoperative month	
			RAW (in mm)	RAH (in mm)	RAW (in mm)	RAH (in mm)
Maxilla	Right	Central incisor	2	11.5	6	14
		Lateral incisor	2	16	6	19
		Premolar	2	13	5	15
	Left	Central incisor	1.5	11.5	5.5	14
		Lateral incisor	2.5	13	6.5	17
		Premolar	1.5	11.5	5.5	13.5
Mandible	Right	Central incisor	1.5	10	5.5	13
		Lateral incisor	2	10	5.5	13
		Canine	3	11.5	6	14.5
		Premolar	4	11.5	8	13.5
		First molar	3	10	7	12
	Left	Central incisor	3.5	10	5.5	12
		Lateral incisor	3.5	11.5	7	13.5
		Canine	4	11.5	7.5	14
		Premolar	4	11.5	7.5	14
		First molar	5	11.5	8	14

RAW=Residual alveolar width, RAH=Residual alveolar height

Table 2: Measurements and site of implant placement

Site	Side	Tooth region	Implant width (in mm)	Implant height (in mm)
Maxilla	Right	Central incisor	3.5	11.5
		Lateral incisor	3.5	16
		Premolar	3.5	13
	Left	Central incisor	3.5	11.5
		Lateral incisor	3.5	13
		Premolar	3.5	11.5
Mandible	Right	Central incisor	3.5	10
		Lateral incisor	3.5	10
		Canine	3.5	11.5
		Premolar	4.3	11.5
		First molar	3.5	10
	Left	Central incisor	3.5	10
		Lateral incisor	4.3	11.5
		Canine	4.3	11.5
		Premolar	4.3	11.5
		First molar	4.3	11.5

impression was taken with the open tray by injecting the light body addition silicone material around the copings and identifying the tips of the guide pins. The appropriate implant replicas were attached to the impression copings to fabricate the master cast, using standard methods and type 4 dental stone (ANSI/ADA Specification No. 25, ISO 6873:1998).[3] A gingival mask was used to ensure that the emergence profile of the crown was optimally contoured. Temporary titanium nonengaging abutments were fixed to the master cast and they were stabilized using dental floss and pattern resin, it was tried in the oral cavity to ensure that there was no displacement of the positions. The occlusal rims were fabricated to employ the maxillomandibular relationship. To obtain the relationship of the maxillary dentition to the horizontal reference plane, face bow transfer was established. Articulation of the maxillary and mandibular cast to fully adjustable articulator (STRATOS 300, Individual Articulator, Ivoclar Vivadent) permitted the check for lateral movements. The patient was satisfied with the esthetics and the occlusion created in the trial denture base; after which the putty based index was made over the wax trial.

Procera® Zirconia Implant Bridge system

Scanning of the model locators using a situation manager-OPTIMET Nobel Procera® Scanner 1.3, Optimet Optical Metrology LTD, Jerusalem, 91450 Israel, was done [Figure 5a]. The Procera Implant Bridge (PIB) was designed and scanned with CAD spray (Telescan, DFS-DIAMON GmbH; D-93339 Riedenburg) for clear details. The zirconia framework was framed and a trial was done to prevent errors [Figure 5b]. The customized abutments were telescopic in shape, this enabled a

Class I relationship [Figures 5c and 6b]. The crowns were constructed and the zirconia ceramic layering and firing was completed. The zirconia crowns were cemented to the framework and the screw access was blocked with gingival colored composite resin. To enhance the esthetics, the conical shaped canines in the maxillary arch they were subjected to intentional root canal treatment and zirconia ceramic crowns were placed.

RESULTS

In spite of the alveolar ridge augmentation and the placement of titanium dental implants simultaneously as a single stage procedure there was no loss in the stability of the implants. Radiological and clinical evidence of osseointegration was accomplished in all 16 implants by the 6th month of placement. The patient was satisfied in all facets as his chewing ability, phonation, and esthetic were rejuvenated [Figure 6a and c]. Most importantly he has transposed to be calmer and confident individual after attaining a normal smile as he had desired.

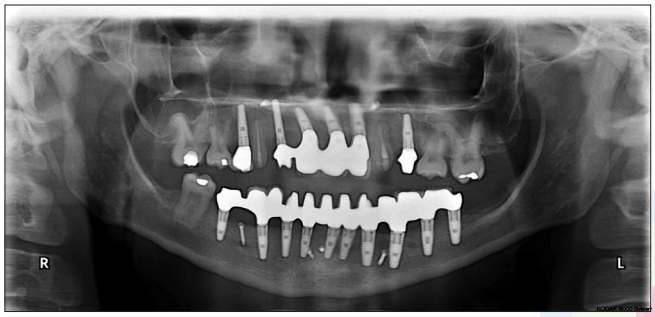

Figure 4: Orthopantomograph showing osseointegration and implant success in the 6th month review after the healing phase

DISCUSSION

Abnormal angulations in the placement of dental implants would result in the failure of the implant.[4] The treatment planning was complex and many factors were to be considered; of which the alveolar ridge, where the RAH and RAW reflects the appropriate treatment choice. However, the quality of the alveolar ridge is almost always compensated in longstanding edentulism due to partial anodontia. Hence, the maxillary and mandibular alveolar ridges with reduced RAH and RAW were indicated for alveolar ridge augmentation. The choice of onlay autogenous bone grafting was performed as the RAW was reduced and the fixation of the graft with rhBMP-2 enabled a good osseointegration at the 6th month follow-up.

Though orthognathic surgery was considered for the correction of the mandibular prognathism initially, it was objectionable in the interest of the vascularization and stabilization of the onlay autogenous bone graft, atrophied jaw and the proximity of the mandibular canal. However, Class I occlusion was compassed prosthetically [Figure 5c].

The sequence and choice of procedure during rehabilitation reflects on the quality of the prosthesis. To avoid uncertainty

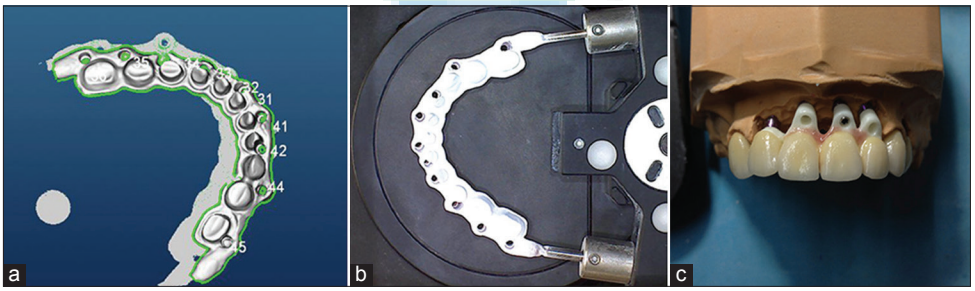

Figure 5: (a) The computer aided designing done with situation manager - Procera® Software Upgrade, version 1.6. (b) The constructed mandibular zirconia framework. (c) The final framework with the prosthetic crowns. The visible maxillary framework is telescopic, this enabled the change of Class III to Class I relationship

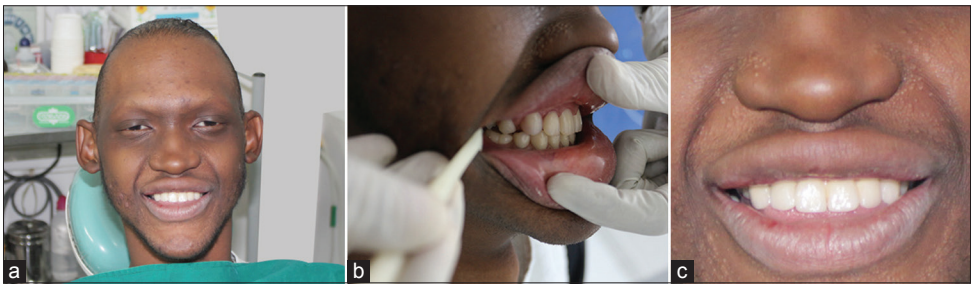

Figure 6: (a-c) Postoperative profile of the patient showing the occlusion and smile

of re-seating of the coping to the impression and for implants with greater divergence than 25° the open tray technique was chosen.[5] The advantages of Zirconia PIB are that it has a good mechanical resistance, biocompatibility, and higher fracture strength.[6] By only grafting on the maxillary arch and implant angulations perpendicular to the basal bone for the mandibular arch the skeletal profile was brought to an end on the relationship. The customized telescope shaped framework[7] of the maxillary and mandibular implant bridge system conceptualized to convert borderline and mild Class III relationship to Class I. Considering the age, using this system in zirconia guaranteed the life span of the implant and the prosthesis. As the prosthesis was a screw retained type the access for retrievability of the framework and the vectors of masticatory forces during lateral movements were considered. The number of interfaces and possibility of peri-implantitis due to subgingival cement is lower than that of cement-retained for prosthesis.[8] All of this had a direct and indirect effect in the success of the customized Zirconia PIB system. The state of partial anodontia had affected the patient's social and psychological development and his behavior and confidence level had grown simultaneously with the functional treatment.

CONCLUSION

A successful case of rehabilitation of complex partial anodontia has been presented. The multidisciplinary approach to the successful treatment planning for the patient with hereditary hypohidrotic ED proved to be necessary. Further, the 1-year follow-up data has proved that dental implants with Zirconia PIB system seem to be a permanent rehabilitation.

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Conflict of interest

There are no conflict of interest.

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