CLINICAL REPORTS

Full Mouth Rehabilitation of a Hypohidrotic Ectodermal Dysplasia Patient with Dental Implants: A Clinical Report

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Prosthodontic treatment in patients with ectodermal dysplasia (ED) is difficult to manage because of the oral deficiencies typical in this disorder and because afflicted individuals are quite young when they are evaluated for treatment. This clinical report describes an 18-year-old patient with hypohidrotic ED treated with dental implants. Treatment included a maxillary implant overdenture and a mandibular hybrid prosthesis supported by osseointegrated implants. At the one-year follow-up, the patient presented significant improvements in oral function and psychosocial activities.

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THE ECTODERMAL dysplasias (EDs) are a group of genetic disorders involving congenital defects of two or more ectodermal structures; i.e., skin, hair, nails, nerve cells, sweat glands, and parts of the eye and ear.[1,2] Oral findings often are significant and can include multiple abnormalities of the dentition (such as anodontia, hypodontia, or malformed and widely spaced peg-like teeth), loss of occlusal vertical dimension, protuberant lips, and lack of normal alveolar ridge development.[3,4] With little or no dental support, a hypoplastic maxilla and mandible result in bite collapse and narrowing of the alveolar ridges.

ED is inherited as an x-linked recessive trait, and has two major types: (1) hypohidrotic or anhidrotic (decreased number or total absence of sweat glands or their abnormal function resulting in a reduced level or lack of perspiration) and (2) hidrotic EDs. The most common condition among the EDs is hypohidrotic ectodermal dysplasia (HED).[5,6] HED is the more severe form and is associated with hypodontia or anodontia, hypotrichosis (fine, sparse blond hair, including a decreased density in both eyebrows and eyelashes), and hypohidrosis or anhidrosis.[7,8]

Conventional prosthodontic treatment for an ED patient has consisted of various combinations of overdentures, complete or partial removable prostheses, or fixed prostheses.[9,10] In recent years, endosseous dental implants have been recognized as an important alternative for ED patients to support, stabilize, and retain the prosthesis.[11-13] The placement of endosseous dental implants in locations favorable for supporting subsequent restorations may be difficult, however, and may require bone grafting.[11,14]

The present report describes using osseointegrated dental implants to rehabilitate an HED patient with cleft palate who presented with maxillary oligodontia, mandibular anodontia, and severe atrophy of the residual alveolar crest.

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Figure 1. Intraoral view of maxillary remaining dentition.

Clinical Report

An 18-year-old male patient with HED was referred for dental treatment. The chief complaint was the unesthetic appearance and lack of function of his dentition.

The patient was under medical care for severe dry eye (xerosis), hearing loss, respiratory problems, and abnormal skin. There were no other medical complications for this patient.

Extraoral examination revealed generalized trichodysplasia (fine sparse hair, scant eyelashes and eyebrows), onchodysplasia (abnormal nails), frontal bossing, a depressed nasal bridge, lip thickening, decreased lower facial height, a prominent chin, and a resultant concave facial profile. The intraoral examination in turn showed partial maxillary (Fig 1) and total mandibular edentulism (Fig 2), unilateral open cleft palate and severe maxillary and mandibular atrophy with a small, thin underdeveloped alveolar ridge. Carious lesions, dental plaque, and calculus deposits were observed on the existing maxillary teeth.

Closure of the congenital complete unilateral cleft lip had been performed at 6 months of age and closure of the complete unilateral cleft palate at the age of 6 years. The surgery was unsuccessful and, as a result, secondary repair surgery and a bone graft using free iliac crest was performed at the age of 17 years. Mandibular and maxillary osteoplasty was performed at the same time to correct prognathic appearance. Again, the surgical procedure for the closure of the palatal cleft was considered unsuccessful.

Orthopantomography (Fig 3) and computed spiral tomography revealed extreme maxillary

Figure 2. Intraoral view of mandibular residual ridge.

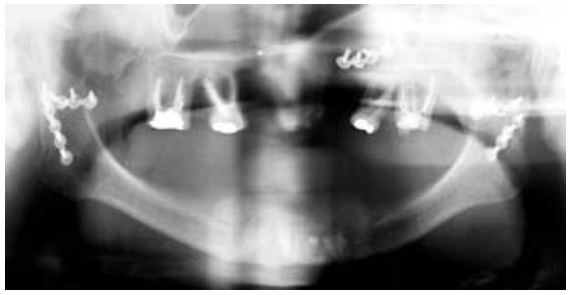

Figure 3. Preoperative panoramic radiograph.

Figure 4. Intraoral view of maxillary implants with cover screws in place.

Figure 5. Intraoral view of implants in the mandibular arch.

atrophy with little remnant bone. The permanent central incisors and the first and second molars were judged to have a poor prognosis due to severe alveolar bone loss.

The remaining maxillary teeth were considered for extraction because of their poor prognosis and advanced periodontal disease. Complete dentures were fabricated as a provisional restoration.

The definitive treatment plan included fabrication of a maxillary implant-supported overdenture (RP-4 according to Misch) and a mandibular implant-supported fixed prosthesis (FP-3 according to Misch).[15]

Preliminary impressions were made using irreversible hydrocolloid impression material (Alginoplast, Heraeus Kulzer, South Bend, IN), and final impressions were made with vinyl polysiloxane impression material (Affinis, Coltène AG, Feldwiesenstrasse Altstätten, Switzerland). Casts were mounted using an arbitrary facebow (Dentatus, Dentatus USA Ltd., New York, NY), and a centric relation record obtained with polyether bite registration material (Ramitec, 3M ESPE, St. Paul, MN) on a semiadjustable articulator (Dentatus ARH, Dentatus USA Ltd.). Tooth set-up was accomplished on the mounted casts from which a radiographic template with metallic balls and a surgical template were fabricated.

Orthopantomography with these radiographic templates was performed to determine proper implant locations. No metallic balls were considered for the alveolar cleft region.

At the appointment for delivery of the complete dentures, the patient was instructed to wear the prosthesis during healing, following extraction of the teeth.

The surgical procedure was performed under general anesthesia. Eight ITI SLA implants (Institute Straumann AG, Waldenburg, Switzerland) with regular platforms were placed in the maxilla, and the same number were placed in the mandibular arch according to the locations of the metallic balls on the radiographic template and the positions of sleeves in the surgical template. Ten days following the surgical procedure, sutures were removed, and the interim complete dentures relined with soft liner (UFI Gel P, Voco, Cuxhaven, Germany).

Six months after the procedure, clinical and radiographic examinations confirmed the osseointegration of all 16 implants (Figs 4-6). This observation has been made in other reports of patients with ED who received edentulous implants.[8,11,12]

Figure 6. Postoperative panoramic radiograph of osseointegrated implants.

Preliminary impressions for implant-supported restorations were made with silicone impression material (Speedex, Coltène AG), and new diagnostic casts were obtained. Custom trays for making final impressions were fabricated on these casts. Screw-retained impression copings (Institute Straumann AG) were placed into the implant bodies, and final impressions were made with polyether (Impregum F, 3M ESPE) using an open tray technique.[16]

To obtain accurate centric relation records, screw-retained record bases were fabricated on the master casts.[16] Casts were mounted on a semiadjustable articulator (Denar Mark II, Teledyne Water Pik, Fort Collins, CO) using centric relation and an arbitrary facebow record (Denar Slidematic, Teledyne Water Pik).

The existence of an open cleft palate and oral hygiene maintenance problems, as well as speech and esthetic difficulties, contraindicated fixed prosthesis treatment planning. As a result, an implant-supported overdenture was selected as the optimal treatment option for the maxillary arch. A hybrid prosthesis was the treatment of choice for the mandibular arch.

Abutment selection was performed on the master cast, and superstructures were fabricated according to the diagnostic set-up. Eight ball attachments (Rhein 83, Bologna, Italy) were placed on the maxillary superstructure to retain the overdenture. Superstructures were tried-in to ensure passive fitness (Figs 7 and 8). Periapical radiographs confirmed complete adaptation of the superstructures over the implant abutments.

Tooth set-up was completed with the superstructures in place. After esthetic try-in,

Figure 7. Maxillary overdenture superstructure with ball attachments for overdenture retention.

confirmation of centric relation and making protrusive and lateral check bite records, the implant fixed prosthodontic occlusion concept (i.e., mutually protected occlusion) was applied.[17] At delivery, abutment screws and superstructure screws were tightened with controlled torque (35 and 15 N/cm^2, respectively), and a clinical remount was performed to refine the occlusion (Fig 9). The access holes were filled with a light-cured composite resin (Tetric Ceram, Ivoclar Vivadent AG, Schaan, Leichtenstein). Oral hygiene instructions were provided to the patient.

Follow-ups were performed after the first week, first month, sixth month, and first year following prostheses insertion. No complications such as speech impairment, esthetic problems, screw loosening and/or fracture, or implant mobility were detected. The patient reported improvements in oral function and self-confidence.

Discussion

ED presents a group of patients with severe congenital and developmental anomalies. Hypodontia and anodontia are the most common oral characteristics of ED. These anomalies affect esthetic and functional activities, which in turn can bring about psychosocial problems for the patient.

Figure 8. Mandibular superstructure for FP-3 prosthesis tried-in the mouth.

Figure 9. Clinical view of maxillary implant-supported overdenture and mandibular fixed prosthesis.

The dentist may be the first person to identify ED in young patients. Rapid growth in early life dictates the use of removable partial or complete dentures for these patients. When full growth is reached, treatment planning may include dental implants to retain, support, and stabilize prostheses.[6,11,13,18] Osseointegrated implants offer an alternative that will provide major improvement in the long-term prognosis for oral rehabilitation.

In treatment planning for implant dentistry in these patients, extra care must be taken to determine whether adequate bone level to receive the implants is present and whether there is adequate vertical dimension of bone to support the implants. Diminished bone volume may limit the success of implants, especially in the maxilla.[11]

This report describes full mouth rehabilitation of an ED patient using osseointegrated dental implants. A maximum of eight osseointegrated implants were used for each arch to compensate for diminished alveolar ridge height, and the resultant reduced length of implant bodies. This would be especially advisable because some of the implants may fail over time. Further, because of the necessity of using general anesthesia and the fact that further such surgeries would be very difficult to be carried out in future, an increased number of osseointegrated implants (eight) were used. A maxillary overdenture was fabricated to facilitate the hygiene maintenance of the open cleft palate region. Prosthodontic treatment improved the patient's esthetic and functional condition and increased his psychosocial confidence and activities.

In conclusion, this case suggests that the use of dental implants in the rehabilitation of ED patients can provide excellent support for dental rehabilitation, both functionally and esthetically.

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References

1. Hickey A, Vergo JR TJ: Prosthetic treatments for patients with ectodermal dysplasia. J Prosthet Dent 2001;86:364-368
2. National Foundation for Ectodermal dysplasia. http://www.nfed.org/
3. Guckes AD, Scurria MS, King TS, et al: Prospective clinical trial of dental implants in persons with ectodermal dysplasia. J Prosthet Dent 2002;88:21-25
4. Dhanrajani PJ, Jiffry AO: Management of ectodermal dysplasia: a literature review. Dent Update 1998;25:73-75
5. NaBadalung DP: Prosthodontic rehabilitation of an anhidrotic ectodermal dysplasia patient: a clinical report. J Prosthet Dent 1999;81:499-502
6. Pigno MA, Blackman RB, Cronin RJ, et al: Prosthodontic management of ectodermal dysplasia: a review of the literature. J Prosthet Dent 1996;76:541-545
7. Blattner RJ: Hereditary ectodermal dysplasia. J Pediatr 1968;73:444-447
8. Davarpanah M, Moon JW, Yang LR, et al: Dental implants in the oral rehabilitation of a teenager with hypohidrotic ectodermal dysplasia: report of a case. Int J Oral Maxillofac Implants 1997;12:252-258
9. Bolender CL, Law DB, Austin LB: Prosthodontic treatment of ectodermal dysplasia. A case report. J Prosthet Dent 1964;14:317-325
10. Snawder KD: Considerations in dental treatment of children with ectodermal dysplasia. J Am Dent Assoc 1976;93:1177-1179
11. Guckes AD, Brahim JS, McCarthy GR, et al: Using endosseous implants for patients with ectodermal dysplasia. J Am Dent Assoc 1991;122:59-62
12. Smith R, Vargervik K, Kearns G, et al: Placement of an endosseous implant in a growing child with ectodermal dysplasia. Oral Surg Oral Med Oral Pathol 1993;75:669-673
13. Rashedi B: Prosthodontic Treatment with implant fixed prosthesis for a patient with ectodermal dysplasia: a clinical report. J Prosthodont 2003;12:198-201
14. Graber LW: Congenital absence of teeth: a review with emphasis on inheritance pattern. J Am Dent Assoc 1978;96:266-275
15. Misch CE: Prosthetic option in implant dentistry. In Misch CE (ed): Dental Implant Prosthetics. St. Louis, MO, Mosby, 2005, pp 43-52
16. Hobo S, Ichida E, Garcia LT: Osseointegration and Occlusal Rehabilitation. Chicago, Quintessence, 1989, pp. 153-55
17. Misch CE: Maxillary denture opposing an implant prosthesis: Hydroxyapatite augmentation and modified occlusal concepts. in Misch CE (ed): Contemporary Implant Dentistry (ed 2). St. Louis, MO, Mosby, 1999, pp 629-645
18. Penarrocha-Diago M, Uribe-Origone R, Rambla-Ferrer J, et al: Fixed rehabilitation of a patient with hypohidrotic ectodermal dysplasia using zygomatic implant. Oral Surg Oral Med Oral Pathol 2004;98:161-165