

Oral rehabilitation of a hereditary ectodermal dysplasia with implant-supported prostheses: A case report

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Abstract

Prosthodontic rehabilitation of patient with ectodermal dysplasia is challenging due to the oral deficiencies typical in this disorder. Ectodermal dysplasia is heterogeneous group of inherited disorders in which two or more ectodermally derived anatomic structures fail to develop. This case report outlined a clinical course of restoring functions and esthetics for a 21 year old female with hypohydrotic ectodermal dysplasia who was treated with implant supported dental prosthesis. The patient was completely satisfied and showed significant improvement in oral functions and psychosocial activities.

Keywords: Ectodermal, Hypohydrotic, Prosthodontic, Implant, Oral

I. INTRODUCTION

The most frequent developing disorders of hereditary origin in the somatognathic system are: Down syndrome, ectodermal dysplasia, cleft lip and palate. Ectodermal dysplasia primarily affects the structures which are of ectodermal origin such as skin, teeth, glands, nails and hair follicle.^[1] Ectodermal dysplasia can be classified with a wide range of clinical manifestations but presence and absence of sweat gland is the prime criteria that classify it into hypohydrotic and hydrotic ectodermal dysplasia. Hypohydrotic ectodermal dysplasia is an X-linked recessive trait with the absence of, or ample lessening in sweat glands. The acceptable etiology behind ectodermal dysplasia is that it occurs due to the aberrant development of ectodermal derivative in early embryonic life. The typical external features of hypohydrotic ectodermal dysplasia are frontal bossing, saddle shaped nose, everted lips, sunken cheek and hyperpigmentation of skin around eyes etc. The most common oral manifestations are anodontia or oligodontia, conical or pegged teeth, delayed tooth eruption and deficiency of alveolar ridge.^[2] The traditional prosthodontic rehabilitation of an ectodermal dysplasia patient has consisted of numerous combinations of fixed and removable prosthesis and overdentures. But in last decade, placement of osseointegrated implant have been recognized and documented in literatures. The treatment plan of patient with ectodermal dysplasia is built upon a number of factors like patient's age, medical & dental history, alveolar ridge and patient's mental outlook.^[3] This is a case report of a 21 year old adult patient who presented with hydrotic ectodermal dysplasia and severe hypodontia. A multidisciplinary treatment approach was used for full mouth rehabilitation using implant supported prosthetics.

II. CLINICAL REPORT

A 21 year old girl was reported to the department of Prosthodontics of M.B. Kedia Dental College and Teaching Hospital, Birgunj (Nepal) with the chief complaint of un-aesthetic appearance and missing teeth. Family history revealed that her father and younger sister were also suffering from the same syndrome. Extraoral examination showed generalized trichodysplasia (fine sparse hair, scant eyelashes and eyebrows), onchodysplasia (abnormal nails), frontal bossing, depressed nasal bridge, everted lips, hyperpigmented skin around eyes, decreased lower facial height, a prominent chin, and a resultant concave facial profile (**Figure 1**). Intraoral examination revealed dry mucosa with oligodontia, conical shaped canine and premolars, and few retained deciduous teeth in mandible. In maxilla, spacing was present between up to premolars with missing laterals and first premolars. Whereas in mandible

retained deciduous lateral incisor and second deciduous molars on both sides were present and rest all anterior teeth were absent (**Figure 2**). The patient also showed up with loss of vertical dimension of occlusion (VDO) with underdeveloped alveolar ridges. Panoramic radiographic examination revealed the entire picture of reduced bone level in lower anterior region of mandible and it also showed retained deciduous and permanent teeth (**Figure 3**). After thorough clinical and radiographic examination, treatment was planned with consultations from orthodontics and oral & maxillofacial departments. The aim of the treatment was the conservation of bone, establishment of normal mechanism of chewing, speech and swallowing and provision of a functional prosthesis with sufficient retention and support. To achieve this goal, treatment plan was outlined in two phases. The first phase included the orthodontic correction in maxillary arch to close the midline diastema, to create space for pontic in 24 region, and to create space for the extraction of retained 25. During orthodontic treatment placement of 3 implants in mandibular arch with extraction of retained 82 was planned (Adin two piece implants). The second phase contained re-establishment of dentition with PFM bridge in 11, 13, 14, and 21, 23, 24 region that also included non-rigid connector in 23, 24 region. Implant supported prosthesis in mandibular arch was completed 8 months after extraction and implant placement surgery. Diagnostic impressions were made using irreversible hydrocolloid impression material (Alginoplast, Heraeus Kulzer, South Bend, IN) and diagnostic models were made with dental stone. Bracket placement was carried out and simultaneously implant placement was done in mandibular arch in 33, 42, 43 region after extraction of retained 82. Post implant placement orthopantomogram was taken. After completing orthodontic treatment, tooth preparation was done in maxillary arch with respect to teeth 11, 13, 15, 21, 23, and 24. Final impression was taken with putty wash technique with vinyl polysiloxane impression material (Affinis, Coltène AG, Feldwiesenstrasse Altstätten, Switzerland). In the meantime, patient was given removable partial denture in mandibular arch. Following this, temporization was done. Later on coping trial was done followed by final cementation using Glass Ionomer Cement. After completing maxillary arch, we placed abutments in mandibular arch, and final impressions were taken by close tray technique with vinyl polysiloxane impression material (Affinis, Coltène AG, Feldwiesenstrasse Altstätten, Switzerland) with gingival mask. Coping trial was done and final prosthesis cementation was carried out (**Figure 4**). Postoperative instructions were given to the patient and post-operative orthopantomogram was taken (**Figure 5**). Patient was quite happy and satisfied with the appearance and function of the prosthesis (**Figure 6**). Patient was put on a 3 month regular follow up schedule.



Figure 1: Extra oral photograph showing concave facial profile (lateral view)



Figure 2: Intra oral photograph before treatment showing oligodontia, generalized spacing in maxillary arch and retained deciduous teeth in mandibular arch



Figure 3: Diagnostic OPG showing retained deciduous and un-erupted permanent teeth



Figure 4: Final cementation of upper maxillary bridge on both sides and coping trial on implants after adjusting abutment



Figure 5: Post-operative orthopantomogram.



Figure 6: Portrait of patient after completion of treatment showing improved aesthetics and facial profile (front view).

III. DISCUSSION

The prosthodontic treatment of ectodermal dysplasia required an extensive knowledge and well organized team work. The primary problem of patient with hypohydrotic ectodermal dysplasia is inefficiency to sweat, leading to irritability and fever in warm environment.^[4]Oligodontia or anodontia are the most typical oral characteristic feature of ectodermal dysplasia patient. These problems not only alter aesthetic and functional activities but also affect the proper psychological and emotional development of patient.^[5]Removable prosthodontics is the most common treatment modality used for the treatment of ectodermal dysplasia patients. Complete denture/partial denture, over denture are the treatment options available for it. These can bring about acceptable aesthetic and functional results but due to less developed alveolar ridges it limits the retention and support. And if the number and distribution of the teeth available seems to be optimal, a fixed partial denture is generally the treatment of choice.^[6]An Osseo-integrated implant proves to be a promising substitute for available conventional treatment options. The success rate of these implants have been mentioned in several published reports but multiple concerns like patient's age, volume of alveolar bone available and maxilla-mandibular relation of remaining teeth in each arch should be taken into consideration.^[7]As we know, in ectodermal dysplasia patients the alveolar ridge is under developed due to lack of tooth development. Because of the bone preserving capacity and increased retention of prosthesis, implants are well chosen for oral rehabilitation of ectodermal dysplasia patient.^[8]It has been reported in the literature that implantsupportedprosthesis showed a significant progress in the emotional and psychosocial development of patients when compared with their condition before the implantswere placed.^[9-10]

IV. CONCLUSION

The treatment with implant supported prosthesis can resolve long term difficulties in ectodermal dysplasia patients. The application of fixed and implant supported prosthesis not only improve the patient aesthetics, phonetics, chewing and deglutition but also showed improvement infunctional, social and psychological activities.

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