

## Prosthodontic treatment of a young patient with hypohidrotic ectodermal dysplasia

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### Abstract

**Background:** Management of clinical manifestations associated with Hypohidrotic Ectodermal Dysplasia (HED) presents a unique challenge for prosthodontists and pedodontists. Oligodontia associated with ectodermal dysplasia is often characterized by underdeveloped alveolar bone with reduced alveolar ridges. This results in reduced bone volume to support conventional prosthetic dentures and for the placement of dental implants. Dental rehabilitation of this kind patients is complicated and interventions may be required throughout the growth period until permanent treatment. Early dental intervention can improve esthetic and functional properties, also emotional and psychological problems can be minimized of patient.

**Case report:** The report highlights the outcomes of tooth-retained overdenture treatment with cast copings in a 20-year-old male with HED. The overdenture treatment, that has an important role in preventive prosthodontics to delay future prosthodontic problems, was planned for patient to provide functional, esthetic and psychological rehabilitation. The patient had no esthetic and functional complaints related to his prostheses eight years later, except for the pain and mobility in right mandibular first premolar tooth.

**Conclusion:** Overdenture may be an important part of the preventive treatment modality among young patients with HED. The aim of this case report was that the esthetic, function and phonation of HED patient who has not indication for implant-supported prostheses were provided with tooth supported overdenture prosthesis.

### Keywords

Hypohidrotic ectodermal dysplasia; oligodontia; overdenture; preventive; young patient.

## Introduction

Ectodermal Dysplasia (ED), which is characterized by abnormal development of two or more ectodermal structures, is a clinical and genetic disorder. Even though by some accounts more than 170 different subtypes of ED can be defined, one of the most commonly encountered type in this group is Hypohidrotic Ectodermal Dysplasia (HED) [1,2]. Hypotrichosis, hypohidrosis, anodontia, and oligodontia are the symptoms which are seen generally [3,4]. Alveolar bone loss, underdeveloped alveolar ridges and reduced sulcular depth are occurred due to oligodontia [1,2]. The skin is dry, scaly and thin, the eyebrows, eyelashes, and hair are absent or sparse [3,5]. In some cases, frontal bossing, saddle nose, prominent supraorbital ridges, unguinal malformation, palmoplantar hyperkeratosis, dyspigmentation, periorbital wrinkles, and chronic rhinitis and pharyngitis may be observed as additional anomalies [6]. In the primary and permanent dentitions, conical roots of posterior teeth, peg-shaped anterior teeth, delayed eruption, impacted teeth, enlarged pulp chambers, and taurodontism may be appeared [7]. Oral rehabilitation of patients with HED usually involves removable dental prostheses [7,8] and also implant-supported dental prostheses [9,10]. HED patients are usually shy and withdrawn. Dental treatment can save their self esteem, and consequently it can enable to socialize of patient and enhance their quality of life. Therefore, the aim of the study was to report a case of maxillar and mandibular oligodontia treated with overdenture in a twenty year-old patient diagnosed with HED.

## Case presentation

In this case report, a 20 year-old male patient was referred for treatment to the Dentistry Department of Bakirkoy Research and Training Hospital for Psychiatry, Neurology and Neurosurgery with a chief complaint of unsatisfactory appearance of his teeth. Extraoral examination revealed sparse and fine hair, frontal bossing, high-set orbits, low-set ears, depressed nasal bridge, sunken cheeks, prominent and reverse lips and pronounced chin. The patient also complained of severe discomfort in hot environment owing to the inability to sweat. His nails were normal but the skin was dry, scaly and rough. Increased pigmentation, perioral wrinkles, and swollen and dry lips were observed. Ethical approval was obtained and prior to examinations, research subject was instructed about the objectives of the study and he agreed to his participation by signing the free and informed consent form. Intraoral examination revealed multiple retained with abnormally shaped permanent teeth 15, 25, 35, 45, 16, 26, 36, 46 which were conical in shape with reduced mesio-distal diameter. Both height and width of posterior alveolar bone were reduced, and additionally sulcular depth was decreased. There was no mucosal defects or pathology on the palate. It was seen that the tongue was enlarged and extended. As a result of clinical and radiological findings, the patient had oligodontia associated with HED. Maxillary and mandibular overdentures were identified as the most appropriate treatment considering its benefits of simplicity and non-invasiveness as well as preservation of alveolar bone, which is typically underdeveloped in these patients. The diagnostic casts were prepared to evaluate the inter-arch space. The remaining teeth were considered as abutments on which individual protective copings were fabricated post tooth preparation. Impressions of the teeth were made with polyvinyl siloxane elastomeric impression material (Elite HD, Zhermack, Badia Polesine, Italy) and metal copings obtained were checked for fit in the patient's mouth and finally were cemented with chemically polymerizing

adhesive resin (Panavia 21, Kuraray Co. Ltd. Osaka, Japan) (Figure 1,2). For final impression of overdenture prosthesis, a custom tray, which was fabricated with autopolymerized acrylic resin, were used and border moulding (Kerr, Orange, CA, USA) was completed. The impression was taken with zinc oxide/eugenol (Horus, Dentsply, Brasil). Master casts were prepared with Type III dental stone (Moldano, Bayer, Leverkusen, Germany). Copings on the master cast were covered with wax, separating material were applied, and the record base was fabricated.

The denture was prepared with anatomically shaped acrylic teeth, which has a cuspal gradient of  $33^\circ$  (Trubyte Biotone. Dentsply, Canada), in centric occlusion with balanced articulation. The final denture was made with heat-cure acrylic resin (Lucitone199, Dentsply, Germany) and was polished (Figure 3,4,5).

The denture bases were checked in the patient's mouth for passive fit on the mucosa and the attachments with the simultaneous accurate contact, and the occlusion was equilibrated (Figure 6,7). Maintenance procedures of the prosthesis, importance of follow-up were explained to the patient. To complete necessary adjustments, a scheduling was done for the next days. It was observed that the patient was adapted to the denture, and physical and emotional state of patient improved positively. Oral hygiene and recall appointments are a crucial factor for long term successful usage of the prosthesis but although the patient was recalled every six months, he did not visit the clinic regularly. His home was too far from the clinic. Through phone calls, his mother reported significant improvements in speech and mastication.

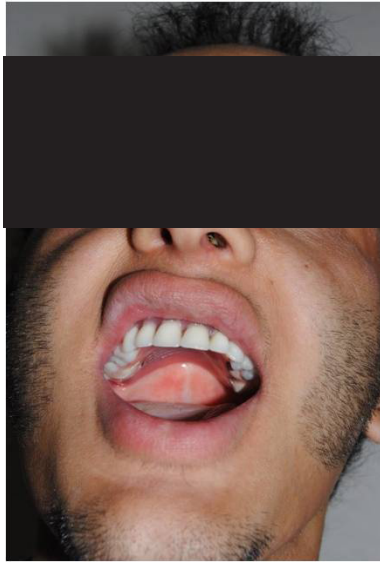
Eight years later, he visited the clinic for dental implant placement. The patient had developed loss of retention in the maxillary denture during those eight years because of tooth extraction 15, 25. Figure 8 shows panoramic radiography of the patient after eight years. The patient had no esthetic and functional complaints related to his prostheses, except for the pain and mobility in right mandibular first premolar tooth. His appearance had changed, and the artificial teeth were abraded. Intraoral examination showed that the patient's oral hygiene was inadequate (Figure 9,10). Dental implant treatment or replacing older prostheses have been delayed because of economic and transportation reasons and he requested that his existing prostheses be modified. After tooth 15 was extracted, with the periodontal and conservative treatment of remaining teeth and the remake of copings, it was decided to replace the prostheses.



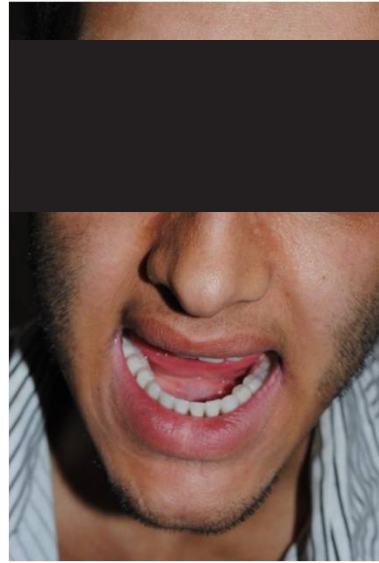
**Figure 1:** Metal coping cemented over prepared abutments on maxilla.



**Figure 2:** Metal coping cemented over prepared abutments on mandibula.



**Figure 3:** Maxillary prosthesis in-situ.



**Figure 4:** Mandibular prosthesis in-situ.



**Figure 5:** Completed maxillary and mandibular overdentures.



**Figure 6:** Facial view showing the completed maxillary and mandibular overdentures.



**Figure 7:** Facial profile view after denture installation.

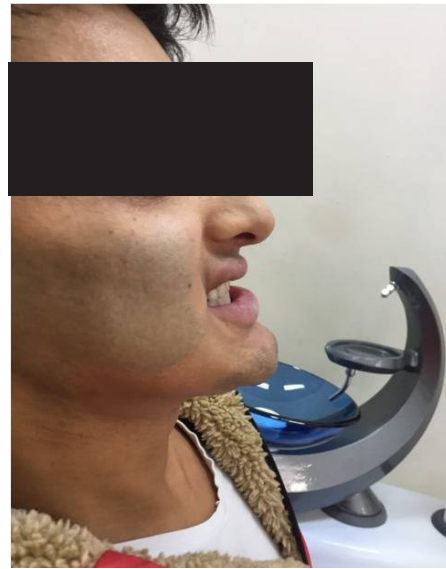


**Figure 8:** Eight-year follow-up findings: Panoramic radiograph.





**Figure 9:** Eight-year follow-up intra oral view.



**Figure 10:** Eight-year follow-up profile view.

## Discussion

Early intervention is an important aspect of dental treatment in patients with HED and gives the patients an opportunity to develop normal oral functions like mastication, speech and swallowing with improved facial support. There is no recommended age at which treatment should commence [3]. However, dental treatments should be completed to provide functional, phonational, psychological, and esthetic before school age [1,5]. In the present case, removable tooth-supported overdenture prostheses were fabricated for the patient. The overdenture prosthesis is a better option than removable complete denture prosthesis due to properties such as to delay the resorption process of the bone, to increase masticatory efficiency and to improve denture foundation area [11]. The proper attachment must be selected carefully for success of the tooth-supported overdenture treatment according to the case. Available buccolingual and inter arch space, the amount of bone support opposing dentition, clinical experience, maintenance problems, cost and motivation of patient are the factors to be considered for attachment selection. Before planning of attachments, the teeth, which are used for overdenture abutments, should be determined. In the case report described above, customized small primary coping with seconder coping was selected for a case with limited inter-arch space. In cases with limited interarch space, denture base can be reinforced with metal framework on top of the coping area to reduce from overdenture fracture.

## Conclusion

This report highlights the outcomes of conventional prosthodontic treatment in a young patient with HED. Such treatment has beneficial psychological effects and improves stomatognathic functioning, esthetics and phonation in young patients who are not candidates for implant-supported dental prostheses. The concept of conventional tooth-retained overdentures is a simple and more cost effective treatment than the implant overdentures. When few firm teeth are present in an otherwise compromised dentition, they can be retained and used as abutments for overdenture fabrication. This helps improve the retention and stability of the final prosthesis significantly. The tooth supported overdenture is a positive treatment

procedure to delay the process of complete edentulism and to preserve bone. To top it all, it gives the patient the satisfaction of having prosthesis with his natural teeth still present. Besides, the patients should be careful for maintenance of oral tissues and prosthesis hygiene, and should be keep recall appointments.

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