

Prosthodontic management of hypohidrotic ectodermal dysplasia: a case report

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ABSTRACT

Introduction: Ectodermal dysplasia (ED) is a hereditary disorder associated with developmental disorders of two or more structures of ectodermal embryonic origin. Hypodontia or anodontia of the primary and permanent dentition, poorly developed alveolar ridges and improper maxillo - mandibular relations, are the most common oral manifestations. Management of patients with ectodermal dysplasia requires a multidisciplinary team approach.

Case presentation: A 6.5 year-old boy with hypohidrotic ectodermal dysplasia (HED) presented in this article, had typical features of HED: hypohidrosis, hypotrichosis, severe hypodontia, atrophic alveolar ridges, old-looking facial expression. According to the patient's age and clinical findings, removable complete over-denture prosthesis in both arches was the treatment of choice.

Conclusion: In patients with ED, it is important to establish correct maxillo - mandibular relations and normal function of the dento-facial system (chewing, swallowing, and speaking). Prosthodontic treatment has a major impact on aesthetics and functions, facilitates psychological development and improves emotional condition and social life of the patient.

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INTRODUCTION

Ectodermal dysplasias (ED) represent a large and heterogeneous group of disorders characterized by developmental disturbances of two or more tissues and structures (skin, hair, nails, sweat glands, teeth enamel, eye lenses, parts of the inner ear and nerves) derived from the embryonic ectodermal layer.^{1,2} More than seven out of 10,000 babies are born with some form of ectodermal dysplasia, regardless of the gender, race or ethnic group.³ Around 170 types of ectodermal dysplasia, clinically different conditions, can be distinguished depending on the

structures that are not properly developed. Most of these conditions are inherited; however there are literature data of children that have been reported as the first case of ectodermal dysplasia in their family.⁴

There are two major categories of ectodermal dysplasia depending on the number and functionality of the sweat glands: hypohidrotic (HED) or anhidrotic, and hidrotic type. Anhidrotic or hypohidrotic type (Christ - Siemens - Touraine syndrome) are the most common types of ectodermal dysplasia where the sweat glands are either absent or significantly reduced in number and function, resulting in heat intolerance.⁵ Hypohidrosis, hypotrichosis (skin, hair and nail anomalies) and hypodontia or anodontia, present a triad of symptoms of this syndrome. As it has an X - linked recessive inheritance pattern, it severely affects males, although females may also display minor symptoms. Hidrotic ectodermal dysplasia (Clouston syndrome) is inherited as an autosomal dominant trait, with properly developed sweat glands and equally affects males and females.^{4,5}

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Children with ED usually have a normal intellectual development and life expectancy, but their facial appearance (result of developmental abnormalities) warrants professional concern for their emotional well-being and social progress.^{6,7} Several functions of the oro-facial system such as mastication, swallowing and phonetics are disrupted due to the improper maxillo-mandibular relations (including reduced vertical dimension)⁶, possible disturbances in the development of jaws/palate, teeth absence (hypodontia or anodontia), and malformation or malposition of existing teeth.

A multidisciplinary team approach^{8,9} is needed to establish the correct functions of oro-facial system. Pedodontist, orthodontist, maxillo-facial surgeon, prosthodontist as well as speech therapist, play a major role in the treatment of patients with ED.

Pedodontist is usually the therapist who has the first contact with child having ED. Trust of the child and an attitude towards dental treatment, usually depends on the pedodontist's ability "to get closer" to the child. Along with orthodontist, they provide guidelines for the course of treatment, taking into account the stage of bone and dental growth and development. Maxillo-facial surgeons solve difficult malformations like cheilognathopalatoschisis or heavier skeletal malocclusions.

Prosthodontic treatment is the last and very important step in the management of ED patients. It includes removable dentures (complete, partial or over-denture prosthesis), fixed partial dentures, and sometimes insertion of an endosseous implants. The treatment modality depends on the patient's age, developmental stage, clinical findings, but also social status of the patient's parents/guardians. Implant placement and fixed prosthesis with rigid connector crossing the mid line (especially in maxilla), in young patients should be postponed till the craniofacial growth is completed. Thus, removable prosthesis with periodic renewal becomes the treatment of choice in growing patients with ectodermal dysplasia.¹⁰

Very important thing is that the prosthodontic treatment should be started during pre-school age of the child, so it would have enough time to get used to the dentures before going to school. It is important for establishment of a new dietary habits and normal function of oro-facial system, but also has a great impact on the psychological development as well as improved and more rapid social integration of the child in new environment.¹¹

This article presents a case of prosthetic rehabilitation of a child with hypohidrotic ectodermal dysplasia associated with severe hypodontia.

The purpose of this article is to point out that, although the treatment of patients with mild phenotypic expression of ectodermal dysplasia can be treated by general dentists, if they have a good knowledge of the matter (specifics of child growth and development), it is better if such cases are treated in a clinical setting because of the possibility for close consultative

communication and cooperation between pedodontist, orthodontist and prosthodontist (multidisciplinary approach).

CASE REPORT

A 6.5 year-old boy with hypohidrotic ectodermal dysplasia came to the University dental clinical center for the control examination of the previously made (1 year before) dentures. The patient could not get used to the dentures; he had felt uncomfortable. Due to jaw development and increased length of the clinical crown of the existing deciduous teeth, it was decided to devise a new maxillary and mandibular prosthesis.

After the family and medical history were taken, a complete examination was performed, including clinical examination and panoramic radiography. Family history reveal that mother have hypodontia of upper lateral incisors, younger brother with ED, and healthy older sister.

A clinical extraoral examination (Figure 1a,b) revealed typical features of ectodermal dysplasia like square and bossing forehead, prominent supraorbital ridges, slightly pigmented and wrinkled eyelids, prominent and pointed ears. The nose was short and wide with anteverted nostrils and depressed nasal bridge, giving the middle face concave appearance. The patient had large philtrum, protruding lips, small and pointed chin, deep mentolabial and nasolabial folds and lower facial height (vertical dimension) contributed to a senile (old-looking) facial expression. The hair was sparse, very thin, soft, blond and slow-growing (hypotrichosis). Eyebrows and eyelashes appeared one year before. The skin was dry (as result of hypohidrosis), thin with light coloring, rough and shiny, with several wounds caused by scratching. Finger and toe nails appeared short, thick, striated and slow-growing. Because of the hypohidrosis, boy had difficulties with body temperature regulation; boy cannot stand high air temperatures.

Intraoral examination revealed undeveloped maxilla with poorly expressed tubers, flat palatal vault with slightly prominent and wide palatal tori, hypertrophic gingivobuccal plicae. Alveolar ridges were rather atrophic (knife-ridge) except in the areas where teeth were present. The color of alveolar mucosa and gingiva was normal. Severe hypodontia was present with missing most of the primary and buds of the permanent teeth. Only five primary teeth existed in the jaws: central incisors and canines in the maxilla and right canine in the mandible. Teeth were spaced and conically shaped with deep overbite in canine region. Salivary secretion was a little bit reduced, but the child did not experience any problems during the mastication or wearing the dentures. Underdevelopment of alveolar ridges was also confirmed by orthopantomogram (Figure 2) that revealed four deciduous teeth in the maxilla and one in mandible, as well as only two developing permanent teeth in the frontal region of the maxilla.

Figure 1. Features of ectodermal dysplasia in a 6.5-year-old boy.



Figure 1a. Frontal view - senile (old-looking) facial expression.



Figure 1b. Profile view - note deep mentolabial and nasolabial folds and lower facial height (vertical dimension), protruding lips and pointed chin.

The patient attends regular school as well as 40-minute lecture every day at school for children with disabilities (parents observed greater intellectual development of the child).

Social behavior of the child has been improved since the first pair of dentures had been made one year before, as well as nutrition. That resulted in prompt growing and development of the child.

Treatment procedure: Since patient was only 6.5 years old, with undeveloped alveolar ridges, making of new maxillary and mandibular mobile dentures could be considered as a treatment. The choice has been made to produce tooth supported overdentures, preserving the existing deciduous teeth as long as

possible and preserving of the alveolar bone as well. Routine procedures followed for the construction of the complete overdentures: preliminary impressions were made with appropriate stock trays and irreversible hydrocolloid material (Hydrogum soft, Zhermack, Italy). Casts were prepared using dental stone and custom trays (Plaque Photo Light - curing hybride composite resin for making individual trays, Dentabiz, Sweden) were fabricated respectively. Border molding was done with a thermoplastic material (Hoffmann's Impression Compound green, Germany) while the final (functional) impressions were made with light body polyvinyl siloxane impression material (Low viscosity C - Silicone Oranwash L, Zhermack, Italy). Final casts were made using hard dental stone and temporary bases (Hoffmann's Shellac Base Plates, Germany) with wax rim (Modeling wax, Dentaurum, Germany) were made respectively. Maxillo - mandibular relations were established, vertical dimension of occlusion and centric relation were recorded. Then the casts were mounted on a semi adjustable articulator and artificial teeth (NT Ünay acrylic resin teeth, Toros Dental, Turkey), reshaped considering the child's age, were arranged according to a balanced occlusion. Final trial was taken to verify vertical and centric relations, occlusion, phonetics and aesthetics. The maxillary and mandibular prosthesis (Figure 3) were fabricated in the conventional heat cure acrylic resin (SR Triplex Hot, Heat - curing denture base material, Ivoclar Vivadent, Schaan Liechtenstein). The dentures were then inserted in the patient's mouth and adjusted carefully. The facial frontal and profile expression improved significantly with complete dentures (Figure 4 a,b,c) resulting in patient's satisfaction with his facial appearance. The patient and his mother were instructed about maintaining the denture hygiene and wearing them all the time except while brushing and sleeping.

Prosthetic rehabilitation of the patient was followed by numerous challenges: his age (generally, children are impatient) and several delicate stages of dentures fabrication. Cooperation with the patient at the beginning was difficult, but after acquisition of mutual trust, further stages were performed without major problems; it was possible to implement all the necessary procedures.

Because of the delicacy of the procedures for taking the impressions, especially border molding and taking functional impressions (causing an urge to vomit), the patient was scheduled in the morning, before any meal taken, as the child was most relaxed at that time. The child performed all the required (muscle) lips and tongue movements, needed to determine the borders of the dentures. Because of the absence of most of the primary teeth, the patient had no sense of centered positioning of the mandible in relation to the maxilla; it took time to establish proper maxillo - mandibular relations (vertical dimension of occlusion and centric relation).

Due to lack of children's artificial teeth, standard teeth that were selected for this case, were reduced in their dimensions and reshaped, so they impose as primary teeth. Considering the

Figure 2. Orthopantomogram reveals undeveloped alveolar ridges and severe hypodontia.



Figure 3. The maxillary and the mandibular over-denture prosthesis made of heat cure acrylic resin.



Figure 4. Improved facial appearance after insertion of over-denture prosthesis.



Figure 4a. Frontal view.

patients' age, the dental arches ended with first molars that were set in dentures without reduction. Somewhat reduced secretion of saliva did not affect the implementation of the therapeutic procedures.

Recall was done after 24 hours to make necessary corrections. After a week, the patient got used to the dentures very well. Four weeks later, there was a need for minimal adjustment of the dentures. One month was enough for the child to accept the dentures. They helped him to talk properly, and allowed him establish normal dietary habits. His appearance improved, as well as his social life and his relationship with the classmates. Subsequent visits were scheduled at 2-month intervals to monitor bone growth and development, as well as oral hygiene.



Figure 4b. Profile view.



Figure 4c. Smile.

DISCUSSION

Clinical expression of hypohidrotic ectodermal dysplasia depends on structures and organs that are affected during the embryonal development. In this case a boy with HED is presented with several symptoms that include skin, scalp and body hair abnormalities, gland, finger and toe nails abnormalities, cranio - facial and oral abnormalities as well as teeth abnormalities.

Although the intelligence is not affected, facial appearance of young patients affects their emotional condition as well as their social life. That is why the treatment of oro - facial and teeth abnormalities has positive impact not only on masticatory and phonetic functions, but great influence on aesthetics and self - confidence of the patient.

Pedodontist, orthodontist, maxillo - facial surgeon, prosthodontist as well as speech therapist should be included in the treatment of patients with ED. Decayed teeth, if any, should be restored with composite materials and malformed teeth reshaped.⁹⁻¹²

Sometimes prior to a prosthodontic treatment, there is need of inserting an orthodontic appliance, no matter if there are impacted,¹³ misaligned teeth or inappropriate (improper) maxillo - mandibular relations (malocclusion).¹⁴ According to Suri et al.,⁷ there should be "simultaneous use of functional and fixed appliances to modify the pattern of dentofacial development". The objectives of orthodontic treatment during period of growing and development, are improving the sagittal and vertical skeletal relations that could be achieved with modification of the functional appliances simultaneously with eruption of the maxillary and mandibular molars and dental arch expansion. The aim of a second phase of orthodontic treatment (closer to the age of skeletal maturation) would be definitive tooth alignment and space management using fixed orthodontic appliances as preparation for dental implant - supported restorations.⁶ Orthodontic treatment resulted in closure of diastema and spaces when agenesis is moderate. In cases with severe agenesis, orthodontic appliances enable retention of spaces for future prosthodontic treatment.¹⁵

Prosthodontic treatment may include conventional removable prosthesis, implant - supported prosthesis,¹⁶ fixed prosthesis,¹⁷ as well as flexible dentures¹⁸ depending upon the severity of disease and patient's demand.¹⁹ These approaches are used either individually or in combination to provide optimal results.

Fixed prosthodontic treatment is seldom used exclusively in the treatment of ED because of the presence of a minimal number of teeth and large pulp cavity. Fixed prosthesis, if it crosses the midline, interferences with maxillary growth in young patients due to rigid connectors.²⁰ El Osta Chaiban et al. reported successful outcome of multidisciplinary approach in solving the

teeth abnormalities in young girl with ED, with orthodontic appliance for teeth alignment and removable prosthesis during the growing period, and fixed porcelain fused to metal bridges when the girl was 17 years old.²¹

Dental implants can be used to support, retain and stabilize the prosthesis. Success of the implant therapy depends on the age of the patients. Ectodermal dysplasia doesn't have negative effect on survival rate of implants when inserted on adult patients. Guckes et al.²² presented 91% survival rate of endosseous dental implants that were placed in anterior mandible and 76% of those placed in anterior maxilla in a group of ED patients aged 8-68. Same authors described successful placement of implants in 3 year - old patient with ED but only in mandible.¹⁶ As the lateral growth of the anterior mandible is usually completed by 3 years of age, implant placement can be considered if anodontia exists in this area. Implants' relative position within the anterior mandible remains unchanged as growth takes place in the rami and condyles.²² In any case, it is not recommended to place implants in growing children as a routine practice, especially in maxilla.²³ According to the guidelines of National Foundation for ED, implants are recommended for the anterior portion of mandible in children older than preschool age (7 years or older).²⁴ Kramer et al.²⁵ successfully inserted and functionally loaded implants into the anterior mandible in 8 year - old children with severe hypodontia. According to Bector et al.,²⁶ dental implants inserted at 8 years of age can be successfully integrated following maxillary and mandibular growth displacement; minor impaction of the maxillary implants and change in mandibular implants' inclination were detected 12 years after insertion.

Cronin et al.²⁴ have concluded that implants placed after age 15 years for girls and 18 years for boys, provided the most predictable prognosis. Yet, Sweeney et al.²⁷ found 20% failure in implants placed in anterior maxilla and only 7.8% failure in anterior mandible.

There are different consequences of oral implants, if placed before dental and skeletal maturation:^{16,22,25} multidimensional restrictions of craniofacial skeletal growth, unpredictable implant dislocations¹⁶ as they do not participate in the maxillary growth process of drift and displacement, implant exposure because of bone resorption, limitation of maxillary growth if implants are connected by a rigid prosthesis that crosses the midline, infraocclusal position of inserted implants compared with adjacent natural dentition,^{16,24,28} as well as trauma to adjacent teeth buds leading to abnormalities in teeth development.

Removable prosthesis made by acrylic resin (complete dentures, partial dentures or over - dentures)¹³ are the most frequently reported treatment modality for the dental management of ED in childhood; these are cost effective,²⁹ and can be easily readapted

and modified (relaying) during periods of rapid growth.

The objectives of prosthetic treatment in young patients with ectodermal dysplasia are bone preservation, early development of correct patterns of chewing, swallowing, speaking and restoring good facial support with improved temporomandibular joint function facilitating development of normal emotional and psychological profile.³⁰⁻³² There is no definitive time to begin dental treatment, but some authors suggest that an initial prosthesis should be provided before school age of the patient, so as to adapt and get used to the prosthesis.²⁰ With regard to child cooperation, dentures can also be fabricated as early as 3 to 4 years of age thus enabling a normal function of masticatory and perioral muscles, and consequently the growth pattern of basal bones. According to Bidra et al.³³ it is critical that children are covered with prosthodontic rehabilitation at an early age. Children rejected by their peer groups are more likely to become aggressive, delinquent, and may experience mental health problems in adulthood. The possibility to look like their peers is paramount for proper psychological development of ED patients.²¹

In this case report, prosthodontic treatment of 6.5 - year - old boy is presented. Treatment consisted of fabrication of over - dentures made of acrylic resin. Preservation of the primary teeth as long as possible, would prevent reduction of alveolar ridge i.e. bone loss, (that could be expected after teeth extraction), and promote a larger area for distribution of the chewing forces and better retention of the prosthesis.

Making over - dentures was possible because the vertical dimension allowed that. On the other hand, partial prosthesis in maxilla was not made because of the teeth positioning: primary incisors have erupted in the middle of the distance between

midline and canines, consequently there was not enough room to insert lateral incisors. Also, inserting only one tooth at the midline would interfere with aesthetics. That was the reason for production of tooth supported over-dentures, with space for deciduous teeth inside them, thus preserving sensory input from periodontal receptors, preserving the alveolar bone, better retention, stability and masticatory performance of the dentures, as well as psychological advantages for the patient.

During treatment procedures the child showed great cooperation. New pair of prosthesis showed better retention than previously made, so mastication can be easily performed. The patient was more satisfied with his face appearance and aesthetics, as correction of vertical dimensions has been made (result of cranio - facial development). An implant - retained prosthesis could be taken under consideration later on, when the jaw development is completed.

CONCLUSION

This case highlights the positive effect of oral rehabilitation on the physical, emotional and social life of the patients with ED. Considering an age when patients should be dentally treated, making removable dentures is a rational, reasonable, acceptable and cost effective option. It is necessary to check and adjust the dentures regularly (at least every two months), to enable the normal growth and development of craniofacial structures, as well as disabling any potential discomfort of the patient. Maxillo - facial surgery, implant placement and fixed prosthesis with rigid connector crossing the mid line, as a more comfortable and aesthetic solution for patients with ectodermal dysplasia, could be considered once the skeletal growth is completed.

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