

Dental Communication

Dental Management in a Child with Ectodermal Dysplasia: A Case Report Along with Review

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ABSTRACT

Ectodermal dysplasia (ED) is a rare hereditary syndrome that is categorized by the nonstandard development of certain tissues and structures of ectodermal origin. This disorder is important to dentists because it affects teeth and results in hypodontia or anodontia. Young children who are affected by ED and anodontia face functional and aesthetic difficulties due to the deficiency of teeth. Restoring lost vertical dimension and teeth with a removable prosthesis improves speech, aesthetics, and appearance and thus boosts the patient's self-confidence. We report a case of hypohidrotic ED with an oligodontia (absence of all primary teeth except upper second primary molars) in a 5-year-old female patient who was successfully rehabilitated with conventional removable dentures in the maxillary and mandibular arches using multidisciplinary approach. The goal of the treatments was to improve psychological development in addition to promoting aesthetics, speech, and eating effectiveness, as well as the overall improvement of the stomatognathic system. Previously, most of the ED cases were treated with removable prosthesis, either in form of partial or complete removable prosthesis in both maxillary as well as mandibular arches. These types of prosthesis were allowed for relining and rebasing as most of the ED patients were receiving treatments during young ages. These days, with the improvements of dental materials and equipments, advanced treatments with overdenture implants have been introduced for replacement of missing teeth which are caused by ED. The present case report along with an updated review discusses the successful dental management of the rare disorder.

KEY WORDS: ANODONTIA, ECTODERMAL DYSPLASIA, MOUTH REHABILITATION, PROSTHODONTIC TREATMENT, REMOVABLE PROSTHESIS.

INTRODUCTION

Ectodermal dysplasia (ED) syndrome is a rare heterogeneous group of inherited disorders that share main defects in the development of two or more tissues derived from the ectoderm (Ul Bari and Ber Rahman 2007). Clinically, ED can be broadly classified into hypohidrotic (X-linked recessive) and hidrotic (autosomal inherited) types. Anhidrotic (hypohidrotic) ED, also known as Christ–Siemens–Touraine syndrome, is the utmost common ED (80%). It affects males

and is inherited from female carriers. It is a triad of sparse hair (atrachosis or hypotrichosis), abnormal or missing teeth (anodontia or hypodontia), and inability to sweat due to the lack of sweat glands (anhidrosis or hypohidrosis) (Naveen et al. 2012; Ladda et al. 2013; Mascolo et al. 2018). Defects in materials originating from other embryologic layers are not unusual. These disorders are congenital, diffuse, and nonprogressive (Ul Bari and Ber Rahman 2007). Patients with ED have a normal life expectancy and intelligence (Al-Araimi et al. 2019; Wimalarathna et al. 2020).

However, if ED goes unrecognized in early infancy, hyperthermia due to the lack of sweat glands may lead to brain damage or death. Anodontia, which represents the

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congenital absence of all teeth in the primary dentition and/or the permanent dentition, is a rare dental manifestation that associates with ED (Chugh et al. 2016; Al-Araimi et al. 2019). Vaidya et al. (2013), observed EDs in 11 patients over two generations in one family. Severe teeth abnormalities; mild fingernail deviations; and toenail abnormalities were seen in all the patients and were considered as abnormalities that affected individuals in a one family. Many case reports have been published worldwide. Table 1 provides a summary of the studies, including the following information: author(s) names and year of publication, country and family history, gender, age, arches involved, teeth present (primary or permanent), dental abnormality associated (if any), treatment received by the patient, and other associated clinical manifestations (Al-Araimi et al. 2019).

The lack of teeth and special appearance are reported as the major concerns of patients with ED. The sequence and type of treatments differ from patient to patient. However, it is important to address the complaints of most patients with ED which include restoring function, speech, facial appearance, and tooth aesthetics by normalizing the vertical dimension and supporting the facial soft tissues. This case report describes a multidisciplinary approach for dental management of a child with ED which involved pediatric dentistry and prosthodontic specialties.

the classical triad of hypohidrosis, hypotrichosis, and hypodontia. Apart from these manifestations, the patient presented dry skin, very thin eyebrows, thin brittle nails, saddle nose, thick everted depressed lips, nasal bridge, and sunken cheeks with prominent supraorbital ridges. The lower third of the face was shorter than the middle and upper thirds (Fig 1 A-C). Intraoral examination revealed a large tongue and complete anodontia in both arches (except for the two primary second molars in the maxillary arch) (Fig 1D). Radiographic examination revealed the presence of thin and resorbed bone in both arches and two maxillary primary second molars (Fig 2A).

Treatment options were discussed with prosthodontic colleague and the parents. The preferred treatment option was a removable prosthesis (partial removable denture for the maxillary arch and removable complete denture for the mandibular arch). At this appointment, oral prophylaxis was performed for the two existing maxillary teeth, then maxillary and mandibular alginate primary impressions were taken (Fig 2B). From these impressions, a study cast (Fig 3A) was poured, and then special maxillary and mandibular custom tries were prepared for the final impression (Fig 3B).

Figure 1: Extra-oral lateral views (A-B), Skin of the hand and nail fingers (C), Pre-operative intra-oral view (D).



Figure 2: Panoramic View of the patient (A), an alginate impression for maxillary arch (B).

Figure 3: Primary cast with special tries (A), Max and mand final impression.

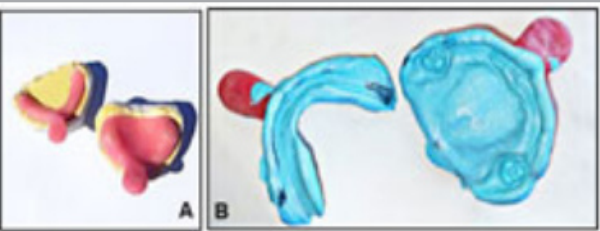


Figure 3; Max and Mand master caste (A), Occlusal rimes on the casts (B), right, left, and frontal views after arrangement of teeth and before to teeth try-in (C-E).



The final impressions of the maxillary and mandibular arches were recorded accurately after border molding by using medium-viscosity elastomeric impression materials. After the beading and boxing of the final impressions, the casts were poured with an improved die stone for increased strength and abrasion resistance (Fig 4A). Then, the maxillary and mandibular occlusal rimes were prepared (Fig 4B). Subsequently, jaw relation was done in the conventional manner, and tooth arrangement was done with acrylic resin teeth and was followed by the

METHODOLOGY

A 5-year-old female patient reported to the Department of Pedodontics and Preventive Dentistry, College of Dentistry, Sanaa University, Yemen. Her chief complaints were the absence of teeth in the oral cavity, the dryness of the mouth, and difficulty during eating. On general and extraoral examinations, the patient presented

final try-in of the waxed dentures (Fig 4C-E). Finally, the wax dentures were processed with a heat-polymerized denture base resin (Fig 5A-B). The complete dentures were delivered, and the patient was instructed on the maintenance of oral hygiene and dentures. Extraoral photographs after the insertion of the dentures were taken (Fig 5 C-E). Recall appointments were done after 1 week and 3 and 6 months for adjustments if needed.

Figure 4: Processed denture is ready for insertion (A-B), extra-oral photographs after insertion of the dentures (C-E)



RESULTS AND DISCUSSION

Our patient is a 5-year-old female Yemeni with oligodontia caused by ED. She was managed by the fabrication of a maxillary Removable partial denture (RPD) and mandibular Complete Denture (CD). With this prosthesis, occlusion, masticatory function, aesthetics, speech, and overall quality of life were restored. The early replacement of missing teeth results in a positive effect on growth and aids in restoring masticatory function and aesthetics; maintains healthy supporting tissues and speech; and boosts the patient's self-esteem, thus improving the patient's overall quality of life (Murthy and Vaze 2010; Al Nuaimi and Mansoor 2019). The use of RPD will allow relining in the future given that the growth of the patient is continuous. It accommodates jaw growth in both arches, reduces the cost and frequency of remaking the prosthesis, and maintains typical oral occupations, thus resulting in an overall improvement in aesthetic outcome (Vieira et al. 2007; Murthy and Vaze 2010; Al Nuaimi and Mansoor 2019).

The clinical expression of ED depends on the structures and organs that are affected during embryonic development. In the present case, a female with ED exhibited several abnormalities that included the skin, scalp, body hair, gland, finger- and toenail, craniofacial, oral, and tooth abnormalities. The facial characteristics of young patients disturb their emotional condition and social life. Therefore, the management of orofacial and teeth abnormalities has a positive effect not only on masticatory and phonetic functions but also greatly influences the aesthetics and self-confidence of the patient and considerably increases their social

confidence, self-esteem, and quality of life (Aragon 2020; Wimalarathna et al. 2020).

A pedodontist, maxillofacial surgeon, orthodontist, prosthodontist, and speech therapist should be involved in the treatment of such patients. Malformed, decayed, or stained teeth should be managed with composite materials (Patel et al. 2010; Prithviraj et al. 2014; Mittal et al. 2015). In patients with ED, establishing the correct maxillo-mandibular relations and the normal function of the dento-facial system (chewing, swallowing, and speaking) is important. Prosthodontic treatment has a major effect on aesthetics and functions, facilitates psychological development, and improves the emotional condition and social life of the patient (Mittal et al. 2015; Mascolo et al. 2018). Alginate hydrocolloid (irreversible hydrocolloid) impression material is used for primary impression and sometimes for final impressions with special trays because it is considered as clean, biocompatible, and pleasant for the patient. In addition, this material has a short setting time and thus causes limited discomfort to the patient and can be easily removed from the undercut area presented by malformed teeth (Wimalarathna et al. 2020).

Although no absolute time for the start of treatment exists, Pigno et al. (1994), stated that an initial prosthesis should be provided before the patient reaches school age. Kupietzky and Houpt (1995), reported that a Removable Denture RD can be fabricated for a patient as young as 3 years old. As the child grows, the RD has to be modified and substituted because longitudinal studies on anodontia have indicated that the growth of both jaws is independent of the presence of teeth. At least three replacements are needed between the period of early and late mixed and permanent dentition. When the patient is in the last period, the RD may be replaced with a fixed-type restoration by using osseointegrated implants. Guckes et al. (1991), recommended that this approach should be postponed till the age of 13 because of possible implant movement caused by jaw growth (Guckes et al. 1991; Pigno et al. 1994; Kupietzky and Houpt 1995; Wimalarathna et al. 2020).

This clinical report describes the types, characteristic features, and treatment options for a young female patient with ED. With proper multidisciplinary treatment, the patient can enjoy a relatively normal life. The options for a definitive treatment plan may include implant-supported fixed or removable prosthesis singly or in combination (Murthy and Vaze, 2010). New alternatives, such as the use of implants, for the rehabilitation for children with hypohidrotic ED must be carefully considered given the presence of underdeveloped, thin alveolar bones, and age (Vieira et al. 2007; Mascolo et al. 2018; Al-Araimi et al. 2019).

CONCLUSION

The findings of this study suggests that a multidisciplinary management of ED should be considered as this multisystemic disorder affects numerous parts of the

body. Early analysis is vital for taking essential preventive actions and enhancing the patient's quality of life. This review and case report contributed to the information on ED and to the education of undergraduate students and general dentists. The treatment was supported in

accordance with the patient's requirements and support (symptomatic management depending on the affected ectodermal structures). Dental professionals play a vital role in contributing to the overall appearance and wellbeing of the affected patients.

Table 1: Summary of worldwide published case reports arranged alphabetically

Author(s)/ Year	Country, Family History	Gender /Age/ Arches	Teeth Present	Treatment Received	Dental Up-normality Associated
Current case	Yemen No	Female/5-Years/ Max & Mand	2 Max primary 2 nd molars	Maxillary RPD & Mandibular CD	Abscess of most primary teeth and all permeant teeth
DI LANARO et al., 2017	Brazil No	Male/ 6-Years/ Max & Mand	Most of teeth were present	Reshaping of Max anteriors with composite & Mand RPD	Absence of permanent teeth Peg shape of all anteriors
de QUEIROZ et al. 2017	Brazil. Yes	Female/10-year-old 000000000000 female patient,	13, 15, 17, 18, 23, 25, 27, 28, 31, 35, 37, 38, 41, 45, 47, 48.	Deciduous teeth 55, 65 and 75	Maxillary and mandibular canines, maxillary premolars Symmetry of present teeth
Velazque and da Silva, 2015	Brazil NO	Male/ 11-Years/ Max & Mand	All Maxillary and Mandibular anteriors	No treatment	Impacted Max & Mand canines
Vieira et al., 2007	Brazil No	Male /5-Years/	No teeth present	Maxillary CD & Mandibular CD	Maxillary and Mandibular with supposed 2 nd primary molar
Nakayama et al., 2015	Japan Yes	Male/14-Years/ 5 Female; age range, 12.7-27.2 years)	Most of teeth present	Referred to ortho, and FPD, Implant treatment	Most effected anterior teeth Present of most permanent teeth Premolar most absent teeth
		4 Female/18-27-Yeras	5-14 teeth present	Referred to ortho, and FPD, Implant treatment	-----
Chugh et al., 2016	India	Male/ 5.6-Years/ Max & Mand	No teeth present	Maxillary & Mandibular CD	-----
Mittal et al., 2015	India Yes	Male/5-years/ Max & Mand	Maxillary 3 teeth	Maxillary RPD Mandibular CD	Conical shape of maxillary canine Absence of permeant teeth
	India Yes	Male /9-Years/ Max & Mand	No teeth	Maxillary RPD and Mandibular Implant Over Denture	-----
Shivastava VK, 2011	India	Male/11-Years/ Max & Mand	Maxillary 1 tooth conical canine	Maxillary RPD & Mandibular CD	-----
Moothedath, et al., 2018	India	Male/20-Years/ Max & Mand	Maxillary canine & Posterior primary teeth	Maxillary RPD & Mandibular CD	Conical shape of Maxillary canines
Sood & Mishra, 2020	India	Male/ 11-Years/ Max & Mand	Maxillary bilateral canines and 1 st Molars	Maxillary & Mandibular RPD	Maxillary peg shape and bilateral primary molars
Kumar et al., 2012	India	Male/ 14-Years/ Max & Mand	Maxillary bilateral canines	No treatment	Absence of all permanent teeth
Hekmatfar et al. 2012	Iran No	Male/3-Years/ Max & Mand	4-peg-shaped anterior in Max Complete edentulism in Mand	Maxillary & Mandibular RPD	2-unerupted molar in Max 2-unerupted incisors in Mand Presence of permanent 1 st molar
	Iran No	Female/10-Years/ Max & Mand	teeth maxillary anterior 4 primary molars, 5 permanent molars, 3 incisors,	Maxillary & Mandibular RPD	Absentees of 3 rd molar teeth ↓ vertical height of facial lower 1/3 & prominent chin lips Unilateral Max contraction Semi-erupted molar, 3 Impacted premolars
Emilija et al., 2015	Macedonia	Male/6.5-Years/ Max & Mand	Maxillary and Mandibular Anterior teeth	Maxillary & Mandibular RPD	Sever bone loss of both arches Absent of all permanent teeth
Emilija et al., 2015	Macedonia	Male/6.5-Years/ Max & Mand	Maxillary and Mandibular Anterior teeth	Maxillary & Mandibular RPD	Sever bone loss of both arches Absent of all permanent teeth
Mascolo et al., 2018	Malta	Female /20 years/ Max & Mand	Most of teeth are present	Maxillary FPD, Mandibular CD	Absent of all permanent teeth
AlNuaimi & Mansoor, 2019	Middle Eastern	Male/ 5-Years/ Max & Mand	All Maxillary and Mandibular anterior with left unilateral molar	Maxillary/ Functional space maintainer Mandibular Fixed PD	Absent of all permanent teeth
Al-Araimi et al., 2020	Oman Yes	Male/ 5-Years/ Max & Mand	-----	No Treatments	Mandibular peg shape canine
Ul Bari and Ber Rahman, 2007	Pakistan No	Male/ 9-Years/ Max & Mand	Centrals and canines are malformed	Refer to Orthodontic	-----
Wimalarathna et al. 2020	Sri Lanka No	Male/11-years/ Max & Mand	-----	Orthodontic Direct and Indirect restorative methods Max and Mand RPD	Absentees of Max, third molar maxillary and mandibular canines Crowded Mand teeth, Peg shaped Mand laterals
Koyuncuoglu et al., 2014	Turkey	Male/12-Years/Max & Mand	2 Max anteriores & 1 Mand	Maxillary FPD & Casted RPD, Mandibular Implant Over Denture	Presented 3 teeth were peg shape Abscess of most primary teeth and all permeant teeth
Yildirim et al., 2012	Turkey	11 Males/5-40 Years/Max & Mand 12 Females	Carious teeth in Maxilla	Depend upon the case	Early diagnosis is vital, patient's quality of life can be improved.
Aragon C, 2020	Turkey	Male/18-Years/Max & Mand	Maxillary 4 anterior & 2 Molar teeth	Maxillary FPD and Casted RPD, Mandibular Implant Over Denture	Semi erupted Maxillary canines No mandibular teeth

Abbreviations: Max-Maxillary; Mand- Mandibular; RPD-Removable partial denture; CD- Complete denture; FPD-Fixed partial denture; 1st – First; 2nd – Second; 3rd – Third

From the physical, emotional, social life, aesthetic, speech, and psychosocial standpoint, early prosthodontic treatment is of great importance to patients with the systemic condition of missing teeth. The psychological environments of the parents and the patient were highly improved particularly with simple and conservative prosthodontic therapy being the rational, reasonable, acceptable, and cost-effective option. Maxillofacial surgery, implant placement, and fixed prosthesis, which will provide results with increased comfort and aesthetics, could be considered later on when the patient have completed skeletal growth.

Conflict of Interest: Authors declares no conflicts of interests to disclose.

Case Report (Human Studies) Ethical Clearance Statement: The Current Case Report/ Studies were Conducted as Per the Guidelines of SCARE. Riaz A Agha et al., (2020). The SCARE 2020 Guideline: Updating Consensus Surgical Case Report (SCARE) Guidelines. doi: 10.1016/j.ijisu.2020.10.034. Epub 2020 Nov 9.

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