View metadata, citation and similar papers at core.ac.uk

brought to you by CORE

provided by epublications@Marquette

Marquette University
e-Publications@Marquette

School of Dentistry Faculty Research and Publications

Dentistry, School of

6-1-2016

Removable Partial Dentures vs Overdentures in Children with Ectodermal Dysplasia: Two Case Reports

Georgios Maroulakos
Marquette University, georgios.maroulakos@marquette.edu

Ioli I. Artopoulou
National and Kapodistrian University of Athens

Matina V. Angelopoulou
Marquette University, matina.angelopoulou@marquette.edu

Dimitris Emmanouil
National and Kapodistrian University of Athens

Accepted version. *European Archives of Paediatric Dentistry*, Vol. 17, No. 3 (June 2016): 205-210.
DOI. © 2016 Springer International Publishing AG. Part of Springer Nature. Used with permission.

Removable partial dentures vs overdentures in children with ectodermal dysplasia: two case reports

Authors:

G. Maroulakos*, I.I. Artopoulou**, M.V. Angelopoulou*** D. Emmanouil****

* Department of General Dental Sciences, Division of Prosthodontics, Marquette University, School of Dentistry, USA

** Department of Prosthodontics, National and Kapodistrian University of Athens, School of Dentistry, Athens, Greece

*** Department of Developental Sciences, Division of Pediatric Dentistry, Marquette University, School of Dentistry, USA

**** Department of Pediatric Dentistry, National and Kapodistrian University of Athens, School of Dentistry, Athens, Greece

Corresponding author's information:

Dr. Georgios Maroulakos

415 E Vine Str #304

Milwaukee, WI 53212

Tel: +1 214 755 7426

E-mail: gmaroulakos@yahoo.gr

Compliance with Ethical Standards

Conflict of Interest: The authors declare that they have no conflict of interest.

Removable partial dentures vs overdentures in children with ectodermal dysplasia: two case reports

ABSTRACT

Background: Ectodermal dysplasia represents a disorder group characterised by abnormal development of the ectodermal derivatives. Removable partial (RPD), or complete dentures (CD) and overdentures (OD) are most often the treatment of choice for the young affected patients. Prosthetic intervention is of utmost importance in the management of ED patients, as it resolves problems associated with functional, aesthetic, and psychological issues, and ensures a patient's quality of life. However, few studies present the principles and guidelines that can assist in the decision making process of the most appropriate removable prosthesis. The purpose of this study was to suggest a simple treatment decision-making algorithm for selecting an effective and individualised rehabilitative treatment plan, considering different parameters.

Case reports: The cases and treatment of two young ED patients are described and each one was treated with either RPDs or ODs.

Follow-up: Periodic recalls were employed to manage problems, and monitor changes associated with occlusion and fit of the prostheses in relation to each patient's growth. Both patients were followed-up for more than 2 years and reported significant improvement in their appearance, masticatory function, and social behaviour as a result of the prosthetic rehabilitation.

Conclusion: The main factors guiding the decision process towards the choice of a RPD or an OD are the presence of posterior natural teeth, facial aesthetics, lip support, number and size of existing natural teeth, and the occlusal vertical dimension.

Key words: ectodermal dysplasia, prosthetic rehabilitation, removable partial denture, overdenture, hypodontia, children

BACKGROUND

Ectodermal Dysplasia (ED) is a genetic condition characterised by dysplasia of two or more structures related to ectodermal embryologic origin (Pinheiro and Freire-Maia 1994). ED has been related to more than 170 genetic syndromes and occurs in approximately 1 in 100,000 live births (Itin 2013). Clinically, the patients present abnormalities of the skin, hair, nails, teeth, mucus and sudoriferous glands (Pinheiro and Freire-Maia 1994). Hypodontia affects around 80% of ED patients and leads to atrophic alveolar ridges and reduced occlusal vertical dimension. In addition, tooth morphological and structural abnormalities are common. Also, ED patients may present with a significant reduction in salivary gland secretion function (Bergendal 2014).

Patients with ED require early oral and prosthetic rehabilitation in order not only to repair function and aesthetics, but also to address psychological issues and improve their self-confidence (Hickey and Salter 2006). An interdisciplinary approach is essential to achieve correct diagnosis and to provide optimal care for the patients and their families (Artopoulou et al. 2009).

The prosthodontic rehabilitation of ED patients must be on an individual basis, considering each patient's growth and developmental characteristics. Many treatment approaches have been reported and may include single crowns, fixed partial dentures (FPD), complete dentures (CD), removable partial dentures (RPD), overdentures (OD), and implant retained prostheses. A removable prosthesis is often the treatment of choice for young patients with ED (Pigno et al. 1996; Hickey and Vergo 2001), and prosthetic rehabilitation with ODs and RPDs has been reported in numerous ED cases with hypodontia (Bonilla et al. 1997; Della Valle et al. 2004; Ioannidou-Marathiotou et al. 2010; Pae et al. 2011). However, no studies present systematically the factors and guidelines that may assist the decision making process on the most appropriate type of removable prosthesis.

The purpose of this study was to suggest, through two clinical reports, a simple treatment decision-making algorithm for selecting an effective and individualised rehabilitative treatment plan, considering anatomical and developmental parameters.

CASE REPORTS

Case 1

History and diagnosis

A 2.5-year-old boy was referred to the Graduate Paediatric Dentistry Clinic at The National and Kapodistrian University of Athens for dental treatment. The patient presented with: hypohidrotic ED, fine sparse hair, scant eyelashes and eyebrows, soft, thin and dry skin, flattened facial appearance, and an aged profile with increased nasolabial fold and pseudo-class III jaw relationship (Fig. 1a). His medical history also included hypothyroidism and recorded full immunization record. An oral examination revealed hypodontia; four primary maxillary teeth (central incisors and second molars) and six primary mandibular teeth (canines, first and second molars) were present. The maxillary central incisors were conical in shape (Fig. 1b). Carious lesions were detected in all maxillary and mandibular posterior teeth and his oral hygiene was poor. The right maxillary primary canine was slightly hypoplastic. Upon examination, his temporomandibular joint and mandibular movements appeared to be within normal limits for a paediatric patient. He presented with an increased horizontal and vertical overlap, and bilateral telescopic occlusion. Furthermore, the maxillary posterior and mandibular anterior alveolar ridges were underdeveloped. A panoramic radiograph was obtained at the age of three years revealing absence of eight primary teeth and seventeen permanent teeth germs (Fig. 1c).

Treatment

The proposed treatment plan included a prescribed preventive programme appropriate to the patient (oral hygiene instructions, topical fluoride application and dietary modification programme), restoration of carious and malformed teeth, fabrication of maxillary and mandibular removable partial dentures and follow-up every three months for caries control and prosthesis adjustments. After completion of his skeletal and dental growth the definitive treatment plan will aim to include orthodontic treatment to favourably position the remaining teeth, and definitive prosthetic rehabilitation.

Following the application of the preventive programme all carious teeth were restored using composite resin (TPH Spectra Universal Composite, Dentsply International).

Maxillary and mandibular impressions were taken using alginate (BluePrint X-creme, Dentsply International). The diagnostic casts were mounted on a semi-adjustable articulator with the use of facebow and centric relation records. A silicone matrix (Lab Putty, Coltène/Whaledent) was fabricated from the diagnostic wax-up and was used for the aesthetic build-up of the central incisors, utilizing the composite resin layering technique (Sakai et al. 2006). Following the reconstruction of the central incisors, a new maxillary impression using irreversible hydrocolloid was made and a cast was mounted on the same articulator with new centric relation record.

Acrylic denture teeth (Bambino Tooth, Major Prodotti Dentari S.p.A.) were set for proper lip support and proper plane of occlusion. Six wrought wire clasps were placed on the following teeth for retention: maxillary incisors, maxillary second molars and mandibular first molars. Both mandibular and maxillary RPDs were processed in thermally activated acrylic resin (Lucitone 199, Dentsply International). The RPDs were inserted and adjustments were made as needed (Fig. 1d). The patient was seen again in 24 hours and 1 week for post-insertion follow-ups.

Follow-up

Prosthetic rehabilitation significantly improved the patient's appearance, masticatory efficiency, speech, and swallowing. The young boy tolerated the RPDs very well. Maintenance, aspiration precautions and oral hygiene instructions were given to the patient and his parents. The patient has been followed up for 3 years with fluoride varnish application every 4 months and adjustments of the RPDs. A new maxillary RPD was fabricated at the age of 6 to accommodate the eruption of the first permanent molars.

Case 2

History and diagnosis

A 4-year-old boy was referred to the Graduate Paediatric Dentistry Clinic, National and Kapodistrian University of Athens for oral rehabilitation. The chief complaint, as stated by the parents, was significant difficulty in chewing and eating due to missing teeth. The patient presented with ED and had a full immunization record.

Extraoral examination showed fine sparse brown hair, slight eyelashes and eyebrows, thin and dry skin, short lower face height, and aged profile. Intraoral clinical

examination indicated the presence of all four primary canines and significantly underdeveloped alveolar ridges (Fig. 2a). All teeth had a conical shape and his oral hygiene was inadequate. Radiographic examination revealed an absence of all other primary teeth and permanent tooth buds.

Treatment

The treatment plan included a preventive programme, consisting of: oral hygiene, topical fluoride application and dietary modification, rehabilitation of dentition using maxillary and mandibular ODs and follow-up every three months for caries control and denture adjustments. After the completion of his growth a definitive treatment with implants and definitive prostheses will be considered.

Following the application of the preventive programme, diagnostic casts were fabricated to determine the available space for restorative materials. Endodontic treatment was necessary to provide adequate space and was performed with calcium hydroxide (Vitapex, Neo Dental International). Subsequently, the clinical crowns were domed leaving 2-3 mm above the gingival level, whereas the root canals were filled with amalgam up to 3mm in the canal (Fig. 2b).

New maxillary and mandibular diagnostic impressions were made using alginate (BluePrint X-creme, Dentsply International) and custom trays were fabricated using light-activated acrylic resin (Triad TruTray, Dentsply International). Final impressions were made using green stick modelling plastic (Impression compound, Kerr) and medium body VPS material (Aquasil Monophase, Dentsply Caulk). Record bases with wax rims were fabricated on the master casts and tried intraorally. The master casts were mounted on a semi-adjustable articulator with the use of facebow and centric relation records.

The setting-up of acrylic denture teeth (VITAPAN, Vita) was completed and the mandibular and maxillary ODs were processed in thermally activated acrylic resin (Lucitone 199, Dentsply International). After processing, a laboratory remounting procedure was performed to evaluate and refine the occlusion and the ODs were polished. The ODs were inserted and adjustments were made accordingly, using pressure indicating paste (PIP, Mizzy) (Fig. 2c). The patient was seen again for 24 hrs and 1 week post-insertion follow-ups. Minor adjustments were made and prostheses'

maintenance and oral hygiene instructions were emphasised. Recall schedule was set with the patient's mother at 3-month intervals.

Follow-up

During the follow-up visits the patient's mother reported significant improvement in the patient's appearance, mastication, and social behaviour as a result of the prosthetic rehabilitation. The patient has been further followed up for 2 years for adjustments of the ODs and preventive application of fluoride varnish on the abutment teeth.

DISCUSSION

Treating the paediatric patient with ED is a challenging task. It has been stated that the clinician managing these patients should, not only be knowledgeable in the various restorative and prosthodontic techniques, but also in the growth and development and behavioural management of these patients (Nowak 1988). In a well calibrated inter-disciplinary team these characteristics do not need to exist in a specific individual but collaboratively between and within the members of the team.

In the cases presented herein, successful treatment was achieved by following a simple algorithm that guided the treatment decision making process (Fig. 3). In this algorithm the factors taken into consideration include: presence of posterior natural teeth, facial aesthetics and lip support, number and size of existing natural teeth and occlusal vertical dimension.

A RPD would be the best option if posterior natural teeth are present such as in case 1 (Rockman et al. 2007). If the existing teeth are characterised by anatomical and morphological abnormalities, proper contours can be achieved with the use of composite resin restorations and a silicon matrix as described in case 1 and other reports (Sakai et al. 2006). This is an easy clinical procedure and preparation of the teeth prior to bonding may not be necessary (Khazaie et al. 2010).

Pretreatment extraoral aesthetic evaluation of the patient is very critical. These patients are frequently characterised by inadequate facial aesthetics due to improper lip support and the use of an OD may be a more appropriate treatment option because the presence of the anterior denture flange can provide an improved aesthetic result (Bidra et al. 2010).

The presence of substantially hypoplastic or partially erupted teeth does not favour bonded composite restorations. The utilization of an OD instead could be a more feasible option (Aydinbelge et al. 2013). In addition, inadequate occlusal vertical dimension combined with the absence of posterior teeth should guide towards the use of an OD, such as in case 2 (Pae et al. 2011). Any treatment approach involving an OD will often require endodontic treatment of the existing teeth and crown reduction as described in case 2, or placement of an extra-coronal coping (Shigli et al. 2005; Bidra et al. 2010; Pae et al. 2011). If the proposed occlusal vertical dimension or the size of the existing teeth provides adequate space for restorative materials, the teeth could be left unmodified.

It should be generally understood that this algorithm is a simplified tool and should be used with caution, since it will cover most but not all the ED patients. The final decision should be made on a case-by-case basis and based on each patient's individualised needs.

The careful follow-up of ED patients after removable prosthodontic rehabilitation is extremely important to ensure a successful treatment outcome and to avoid complications. Problems associated with anatomic and morphological abnormalities of existing teeth and atrophic alveolar ridges may result in poor retention and stability of the prostheses. Progressive alveolar bone resorption is another issue, since the edentulous ridge is loaded at an early age. Furthermore, satisfactory oral hygiene could be much more difficult. As a result, periodontal/soft tissue complications and increased caries rates may further compromise the prosthetic outcome (Pigno et al. 1996; Hickey and Vergo 2001; Bidra et al. 2010).

Education of the patient and the patient's parents regarding possible problems and maintenance of any prosthesis is mandatory. Periodic recalls should be employed. Both presented cases were followed-up for more than two years for application of the preventive programme and adjustments of their prostheses to address changes in occlusion and fit, related to each patient's growth. A broad guideline recommends relining/rebasing intraoral prostheses in a growing patient every 2-4 years and remaking a new prosthesis after 4-6 years (Vergo 2001).

Alternative treatment approaches not covered by the proposed algorithm may include the use of a combined RPD/OD prosthesis (Della Valle et al. 2004), magnets

(Rockman et al. 2007), ODs with windows to accommodate erupting posterior teeth (Bonilla et al. 1997; Dall'Oca et al. 2008) and dental implants. A heightened interest for the use of osseointegrated dental implants in the growing ED population is noted in the literature (Guckes et al. 1991; Vergo 2001; Kramer et al. 2007; Rockman et al. 2007; Klineberg et al. 2013). However, they cannot follow the alveolar bone growth and can become ankylosed and potentially buried (Guckes et al. 1991; Kearns et al. 1999). Therefore, implants are indicated only in the anterior mandible of ED patients that are older than 12 years and exhibit anodontia (NFED 2003). Provisional implants provide an alternative option, because they do not osseointegrate, they are retrievable, and do not interfere with the growing bone (Artopoulou et al. 2009).

CONCLUSION

The main factors guiding the decision process towards the choice of a RPD or an OD in ED patients are the presence of posterior natural teeth, facial aesthetics, lip support, number and size of existing natural teeth, and the occlusal vertical dimension.

Acknowledgement

Informed consent was obtained from all individuals and their parents for whom identifying information is included in this article. The authors would like to thank Dr. T. Gerard Bradley, Associate Dean of Research & Graduate Studies at Marquette University School of Dentistry for reviewing and editing the manuscript.

Figure Captions

Fig. 1 Photographs of Case 1. **a** Initial extraoral profile view. Note the fine sparse hair, flattened facial appearance, increased nasolabial fold and pseudo-class III jaw relationship. **b** Initial clinical frontal view at the age of 2.5 years showing conical shape of maxillary central incisors and congenitally missing teeth. Microbial plaque is present. Also note the bilateral telescopic posterior occlusal relationship. **c** Panoramic radiograph at the age of 3 years revealing absence of eight primary teeth and seventeen permanent teeth germs. All four mandibular primary incisors, two maxillary primary lateral incisors, and two maxillary primary molars were congenitally missing. Also, all four mandibular permanent incisors, two maxillary permanent incisors, all premolars, two mandibular permanent molars, and one maxillary permanent molar were congenitally missing. **d** Clinical photograph displaying build-ups of the maxillary central incisors and prosthetic rehabilitation with maxillary and mandibular RPDs. Note the enamel defect on the right maxillary primary canine. Wrought wire was used for the retentive clasp arms

Fig. 2 Photographs of Case 2. **a** Initial clinical frontal view of Case 2 at the age of four. All primary teeth were absent with the exception of four primary canines. Note the conical shape of the teeth and the significantly underdeveloped alveolar ridges. **b** Abutment teeth for mandibular OD after root canal treatment, occlusal reduction and filling of coronal part of the root canals with amalgam. Gingival inflammation is noted around the abutments, **c** Clinical photograph displaying prosthetic rehabilitation with maxillary and mandibular ODs

Fig. 3 Algorithm to facilitate the treatment decision process in ED patients, different parameters determine the choice between OD and RPD as the most appropriate treatment

REFERENCES

Artopoulou I, Martin JW, Suchko GD. Prosthodontic rehabilitation of a 10-year-old ectodermal dysplasia patient using provisional implants. Pediatr Dent. 2009;31:52-57.

Aydinbelge M, Gumus HO, Sekerci AE, Demetoglu U, Etoz OA. Implants in children with hypohidrotic ectodermal dysplasia: an alternative approach to esthetic management: case report and review of the literature. Pediatr Dent. 2013;35:441-446.

Bergendal B. Orodental manifestations in ectodermal dysplasia-A review. Am J Med Genet A. 2014.

Bidra AS, Martin JW, Feldman E. Complete denture prosthodontics in children with ectodermal dysplasia: review of principles and techniques. Compend Contin Educ Dent. 2010;31:426-433; quiz 434, 444.

Bonilla ED, Guerra L, Luna O. Overdenture prosthesis for oral rehabilitation of hypohidrotic ectodermal dysplasia: a case report. Quintessence Int. 1997;28:657-665.

Dall'Oca S, Ceppi E, Pompa G, Polimeni A. X-linked hypohidrotic ectodermal dysplasia: a ten-year case report and clinical considerations. Eur J Paediatr Dent. 2008;9:14-18.

Della Valle D, Chevitarese AB, Maia LC, Farinhas JA. Alternative rehabilitation treatment for a patient with ectodermal dysplasia. J Clin Pediatr Dent. 2004;28:103-106.

Guckes AD, Brahim JS, McCarthy GR, Rudy SF, Cooper LF. Using endosseous dental implants for patients with ectodermal dysplasia. J Am Dent Assoc. 1991;122:59-62.

Hickey AJ, Salter M. Prosthodontic and psychological factors in treating patients with congenital and craniofacial defects. J Prosthet Dent. 2006;95:392-396.

Hickey AJ, Vergo TJ, Jr. Prosthetic treatments for patients with ectodermal dysplasia. J Prosthet Dent. 2001;86:364-368.

Ioannidou-Marathiotou I, Kotsiomiti E, Gioka C. The contribution of orthodontics to the prosthodontic treatment of ectodermal dysplasia: a long-term clinical report. J Am Dent Assoc. 2010;141:1340-1345.

Itin PH. Ectodermal dysplasia: thoughts and practical concepts concerning disease classification - the role of functional pathways in the molecular genetic diagnosis. Dermatology. 2013;226:111-114.

Kearns G, Sharma A, Perrott D, et al. Placement of endosseous implants in children and adolescents with hereditary ectodermal dysplasia. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1999;88:5-10.

Khazaie R, Berroeta EM, Borrero C, Torbati A, Chee W. Five-year follow-up treatment of an ectodermal dysplasia patient with maxillary anterior composites and mandibular denture: a clinical report. J Prosthodont. 2010;19:294-298.

Klineberg I, Cameron A, Hobkirk J, et al. Rehabilitation of children with ectodermal dysplasia. Part 2: an international consensus meeting. Int J Oral Maxillofac Implants. 2013;28:1101-1109.

Kramer FJ, Baethge C, Tschernitschek H. Implants in children with ectodermal dysplasia: a case report and literature review. Clin Oral Implants Res. 2007;18:140-146.

NFED. Scientific Advisory Board: Parameters of Oral Health Care for Individuals affected by Ectodermal Dysplasia Syndromes, Mascoutah, IL. National Foundation for Ectodermal Dysplasias. 2003:1-28.

Nowak AJ. Dental treatment for patients with ectodermal dysplasias. Birth Defects Orig Artic Ser. 1988;24:243-252.

Pae A, Kim K, Kim HS, Kwon KR. Overdenture restoration in a growing patient with hypohidrotic ectodermal dysplasia: a clinical report. Quintessence Int. 2011;42:235-238.

Pigno MA, Blackman RB, Cronin RJ, Jr., Cavazos E. Prosthodontic management of ectodermal dysplasia: a review of the literature. J Prosthet Dent. 1996;76:541-545.

Pinheiro M, Freire-Maia N. Ectodermal dysplasias: a clinical classification and a causal review. Am J Med Genet. 1994;53:153-162.

Rockman RA, Hall KB, Fiebiger M. Magnetic retention of dental prostheses in a child with ectodermal dysplasia. J Am Dent Assoc. 2007;138:610-615.

Sakai VT, Oliveira TM, Pessan JP, Santos CF, Machado MA. Alternative oral rehabilitation of children with hypodontia and conical tooth shape: a clinical report. Quintessence Int. 2006;37:725-730.

Shigli A, Reddy RP, Hugar SM, Deshpande D. Hypohidrotic ectodermal dysplasia: a unique approach to esthetic and prosthetic management: a case report. J Indian Soc Pedod Prev Dent. 2005;23:31-34.

Vergo TJ, Jr. Prosthodontics for pediatric patients with congenital/developmental orofacial anomalies: a long-term follow-up. J Prosthet Dent. 2001;86:342-347.