



RESEARCH ARTICLE

PROSTHODONTIC MANAGEMENT OF A CASE OF HYPOHYDROTIC ECTODERMAL DYSPLASIA: A CASE REPORT AND REVIEW

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ARTICLE INFO

Article History:

Received 20th March, 2017

Received in revised form

21st April, 2017

Accepted 29th May, 2017

Published online 30th June, 2017

Keywords:

Removable partial dentures,
Complete denture.

ABSTRACT

Ectodermal dysplasia (ED) is a hereditary disorder characterized by abnormal development of certain tissues and structures of ectodermal origin, like the sweat glands, scalp hair, nails, skin pigmentation, and craniofacial structures. Orofacial characteristics of this syndrome include anodontia or hypodontia, hypoplastic conical teeth, underdevelopment of the alveolar ridges, frontal bossing, a depressed nasal bridge, protuberant lips, and hypotrichosis. A 12-year-old male patient exhibiting the classical features of HED (hypohidrosis, hypotrichosis and hypodontia), visited our Department of Prosthodontics at Chatrapati Shahu Maharaj Shikshan Sanstha Dental College and Hospital. In the clinical and radiographic evaluation, it was evident that the only erupted teeth were three conical teeth in the anterior region and one posterior tooth in the left side of the maxillary arch. Resin composite restoration of occlusal morphology of anterior conical teeth followed by provisional removable partial dentures in the maxillary arch and complete denture in the mandibular arch was fabricated for prosthodontic rehabilitation of this patient after considering his growth and the number and condition of his present teeth. At the 6-month follow-up no major complications occurred and definitive treatment can be planned (if necessary) after the completion of growth.

INTRODUCTION

Ectodermal dysplasia (ED) is a hereditary disorder characterized by abnormal development of certain tissues and structures of ectodermal origin (Wynbrandt, 1990). Freire-Maia (Freire-Maia, 1971) defined the nosologic group of ED as any syndrome that exhibits at least two of the following features: (1) trichodysplasia (abnormal hair), (2) abnormal dentition, (3) onchodysplasia (abnormal nails), and (4) dyshidrosis (abnormal or missing sweat glands) (Wynbrandt, 1990). Certain oral and facial characteristics may also be associated with the syndrome. Because there are more than 100 different ED syndromes (Wynbrandt, 1990 and Freire-Maia, 1994), clinical manifestations depend on the specific syndrome afflicting an individual (Wynbrandt, 1990; Freire-Maia, 1971; Freire-Maia, 1994; Lowry, 1966; Witkop, 1975; Freire-Maia, 1988; Buyse, 1990 and Jones, 1988). The most frequently reported ED syndrome is X-linked hypohidrotic dysplasia (Levin, 1988), also known as Christ-Siemens-Touraine syndrome (Buyse, 1990), which affects one to seven individuals per 10,000 live births. Orofacial characteristics of this syndrome include anodontia or hypodontia, hypoplastic conical teeth, underdevelopment of the alveolar ridges, frontal bossing, a depressed nasal bridge, protuberant lips, and hypotrichosis (Levin, 1988; Bixler, 1988; Champlin, 1987; Clarke, 1987 and Mark, 1996).

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Case Report

A 12 yr old male patient reported to the Dept. of Prosthodontics in C.S.M.S.S. dental college and hospital with the complaint of missing teeth, difficulty in eating and esthetics concern. His father gave history that the child was intolerant to warm weather, suffered from constant thirst, frequent fever of unknown origin at childhood, dry skin, reduced amount of sweating, and scanty hair. He had got hair on his head after the age of 1.5 – 2 yrs. He is also currently consulting a dermatologist. The boy is shy, does not talk much or interact easily, and avoids social gatherings, because of his different appearance. At the clinic, if not on the dental chair, he stays close to his father. The patient exhibited the classical features of HED, characterized by hypohidrosis, hypotrichosis and hypodontia (Nowak, 1988) (Figure 1, 2) In addition to these signs and symptoms, the patient presented with dry wrinkled skin, sparse eyebrows, eyelashes and hair which were light brown in colour. The chin and supraorbital ridges were prominent. Frontal bossing, saddle nose, periorbital pigmentation, hyperkeratosis of the palms and soles of the feet were present (Figure 3). The vertical dimension of the lower face was reduced, mandible seemed prognathic and the lips were thick and protuberant, leading to the characteristic senile appearance. (Figure 2). The patient's history, clinical and radiographic examination revealed that the only erupted teeth were three conical teeth in the anterior region and one posterior tooth in the left side of the maxillary arch, which had erupted at

the age of about 4 years. There was spacing seen between the anterior teeth. (Figure 4) The mandibular arch was edentulous, and the ridge was of the knife edge variety. (Figure 5) No teeth buds or impacted teeth were seen on radiographic examination. (Figure 6) Reduced alveolar bone height and hence shallow vestibular sulcus in was seen in both arches, which may be because of absence of deciduous as well as permanent teeth.



Fig. 1. Pre-treatment profile photograph (Front view)



Fig. 2. Pre-treatment profile photograph (Side view)



Fig. 3. Hyperkeratosis of the palms



Fig. 4. Maxillary arch



Fig. 5. Mandibular arch



Fig. 6. Orthopantomogram radiograph of the patient

The oral mucosa was slightly dry and patient had lowered salivary secretions. Oral rehabilitation with provisional removable partial dentures in the maxillary arch and complete dentures in the mandibular arch was planned for this patient to improve both the maxilla-mandibular relationship, as well as to provide improvements in aesthetics, speech and masticatory efficiency. The conical teeth were to be reconstructed with light cured composite resin for re-establishing normal occlusal morphology. Although routine procedures for construction of removable dentures were used, case specific modifications were made and described below.

Treatment Procedure

- The maxillary primary impression was made using irreversible hydrocolloid impression material (MecAlgin Chromatic Alginate, Mectron Dental India Pvt. Ltd.) supported on stock perforated metal tray. The patient was instructed to rinse his mouth with water just before the impression, to prevent the alginate from sticking to his teeth and tearing off due to his lowered salivary secretions.
- The mandibular primary impression was made with low fusing impression compound (Y-Dent’s Impression Composition, MDM Corporation) supported on stock non-perforated metal tray.
- Maxillary and mandibular casts were poured in Plaster of Paris. (Figure 7)
- On the maxillary primary cast, mock up of the composite restoration for the anterior conical teeth was performed in inlay wax, to make them look like central and lateral incisors. (Figure 8)
- A silicone index (Aquasil Soft Putty Regular Set, Dentsply Ltd.) was made of this mock up, and preserved until next appointment. (Figure 8)
- Acrylic custom tray (Asian Special Instant Tray Material, Asian Acrylates) with modelling wax spacer

(Modelling Wax, Deepti Dental Products of India Pvt. Ltd.) was fabricated for partially edentulous maxillary arch, keeping 3 mm of modelling wax spacer in the region of the teeth and 2mm in the edentulous region. Acrylic custom tray (Asian Special Instant Tray Material, Asian Acrylates) with wax spacer (Spacer Wax, Deepti Dental Products of India Pvt. Ltd.) was fabricated for edentulous mandibular arch in accordance to the selective pressure impression technique.

- At the next appointment, the direct composite restoration (Solare Asia – A2 Shade, GC Corporation, Tokyo, Japan) was placed, according to manufacturer’s instruction, with the silicone index placed palatally for guidance. After achieving the proper shape, finishing and polishing was performed. (Figure 9)
- Final impression was taken as follows: Border molding was done using high fusing impression compound (Aslate Impression compound Soft Green Tracing Sticks, Asian Acrylates). Final impressions were made using irreversible hydrocolloid (Chromatic Alginate, Mectron Dental India Pvt. Ltd.) for the maxillary arch (Figure 10), and light body-injection type polyvinyl siloxane impression material (Aquasil Ultra LV, Denstply Ltd.) for the mandibular arch (Figure 11), and master casts were poured.
- Temporary record bases were fabricated on the master casts and wax rims (Modelling Wax, Deepti Dental Products of India Pvt. Ltd.) were fabricated. The maxillo-mandibular relationship was recorded conventionally, after assessing the phonetics and esthetics. The patient had difficulty in giving a proper centric relation as he had never had teeth as centric stops, and hence had abnormal chewing patterns.
- A teeth mould was selected after calculating the inter-canine distance for the anterior teeth, and semi-anatomic posterior teeth were selected and arranged using the monoplane occlusion scheme.
- The try-in appointment was carried out conventionally (Figure 12) and after the approval by the parents and patient, the waxed-up dentures were processed in heat-polymerizing acrylic resin (Acralyn – ‘H’ Denture Material, Asian Acrylates). Clasps were made for upper removable partial dentures to ensure retention (Figure 13, 14, 15).
- On the subsequent appointment, the dentures were inserted placed and proper fit was verified. Instructions were given for use of dentures. Recall appointments were scheduled for 24 hours, 1 week and 3 weeks. Pressure points were relieved and clasps adjusted at the second recall.
- At the 6-month follow-up no major complications occurred.



Fig. 7. Maxillary and mandibular diagnostic casts



Fig. 8. A silicone index of the wax mock up



Fig. 9. Completed direct composite restorations of the anterior teeth



Fig. 10. Final impression of the maxillary arch in alginate impression material



Fig. 11. Final impression of the mandibular arch in light bodied addition silicone impression material



Fig. 12. The try-in appointment



Fig.13. Prosthodontic rehabilitation by provisional removable partial dentures in the maxillary arch and complete denture in the mandibular arch



Fig. 14. Post-treatment profile photograph (Front view) Dentures have instilled self confidence in the patient.



Fig. 15. Post-treatment profile photograph (Side view) Vertical dimension restored

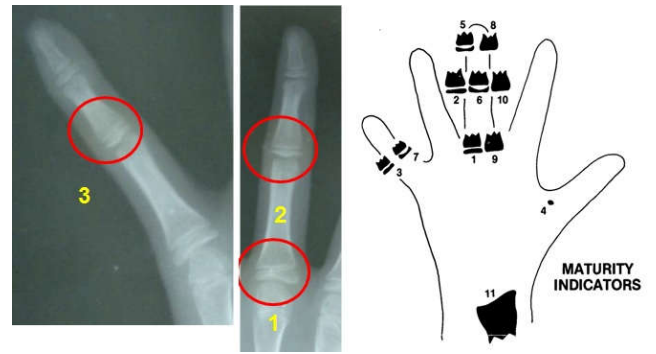


Fig. 16. Skeletal age estimation by hand-wrist radiograph according to Fishman's skeletal maturity indicators system

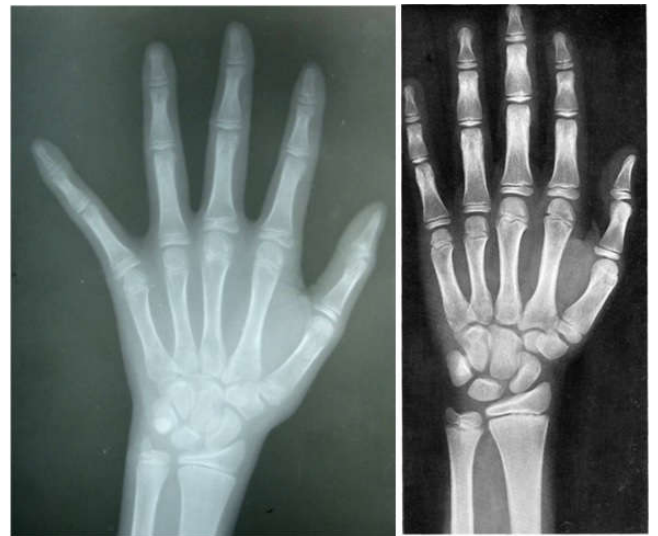


Fig. 17. Skeletal age estimation by hand-wrist radiograph comparing with Greulich & Pyle's atlas



Fig. 18. Skeletal age estimation by lateral cephalometric radiograph according to Hassel & Farman's system

Table 1. A Tentative treatment plan for a patient with Ectodermal Dysplasia (Bergandal)

Age (yrs)	Treatment
6 - 12	Prophylaxis Supervised occlusal development
12 - 14	Orthodontic treatment (if necessary) Maintenance care
14 - 15	Semipermanent prothetic treatment: composite retained onlay FPD and/or RPD with cast metal framework
15 - 20	Maintenance care Tomographic examination: Quantity and Quality of bone in edentulous areas Genetic counseling Dermatologist consultation when needed
20 - 25	Conventional and / or Implant supported Fixed Prosthodontics at patient's demand. Follow up & Maintenance care.

DISCUSSION

The clinical features of ED causes substantial social problems in persons afflicted with the condition (Imirzalioglu, 2002; Hickey, 2001 and Abadi, 2001). Sarwer et al (Sarwer, 1999), reported greater discontent with facial appearance, lesser self-esteem, and anxiety regarding quality of life in individuals born with craniofacial anomalies, as compared with non-facially disfigured adults. The characteristics associated with ED will often result in afflicted individuals having an abnormal appearance. Normal psychologic and social development of young ED patients dictates that they look and feel as normal as possible (Mark, 1996; Nowak, 1988; Tanner, 1988; Jones, 1978 and Nussbaum, 1976). Therefore, dental appearance in these patients is extremely important because it can affect their self-confidence (Mark, 1996). Prosthodontic treatment of ED can include fixed, removable, or implant-supported prostheses. These treatment approaches can be used individually or in combination to provide favourable results (Mark, 1996).

A provisional treatment plan has been given for a patient with Ectodermal Dysplasia by Bergandal (Bergandal, 2001), as in Table 1. Unfortunately, because of the diminished number of teeth, patients often present with issues related to occlusal vertical height and esthetics (Pavarina, 2001 and Itthagarun, 1997). Also, because of the limited tooth structure remaining, these patients may require comprehensive restorative and prosthetic treatment to attain proper function, esthetics, and comfort. Such needs can create a challenging treatment point-of-view (Pavarina, 2001). Two main factors affecting the success of dental treatment for Ectodermal dysplasia are: insufficient supported underlying mucosal tissues, and reduced salivary flow (Jahangiri, 2011). If the child is cooperative, prosthodontic intervention in those as young as age 2 or 3 years can be successful. This affords the child the fighting chance to develop normal forms of speech, chewing, and swallowing; normal facial support; improved temporomandibular joint function; and improved self-esteem (Imirzalioglu, 2002; Hickey, 2001). When a child with ED reaches his or her early teens, orthodontic treatment may be indicated to correct the diastemata and spaces (Imirzalioglu, 2002).

Skeletal age Considerations

To estimate the skeletal age of the patient, a Lateral Cephalogram and a Hand-Wrist Radiograph was used.

Hand-wrist Radiograph

- According to Fishman's skeletal maturity indicators system, the skeletal pattern resembled that of S.M.I. 2, i.e. width of epiphysis equal to that of diaphysis in the middle phalynx of third finger. Hence, skeletal age in years was found to be 11.68 ± 1.06 years, and therefore percentage of growth completed was estimated to be 16.7 % (Figure 16)
- Comparing with Greulich & Pyle's atlas, skeletal age was found to be 12 years 6 months (Figure 17)

Lateral Cephalogram

According to hassel & farman, considering C2, C3, C4 cervical vertebrae the skeletal pattern resembled that of Stage 3, i.e. Transition stage: Distinct concavities in inferior borders of C2 and C3. A concavity was beginning to develop in inferior border of C4. Therefore, accordingly, 25 % to 65 % of adolescent growth is expected. (Figure 18)

Fixed Prosthodontics

Fixed prosthodontics are rarely used exclusively in the treatment of ED, primarily because many afflicted individuals have a minimal number of teeth.¹³ Also, patients are often quite young at their first visit for treatment and fixed partial dentures (FPDs) with rigid connectors should be avoided in actively growing young patients, as they could interfere with growth of jaws, especially if the prosthesis crosses the midline. (Mark, 1996 and Hogeboom, 1961). Individual crown restorations, though not limited with concerns regarding jaw growth, may be restricted by larger pulp sizes and shorter crown heights (Mark, 1996; Smith, 1990). Recently, direct composite restorations have become the more desirable method of restoring normal morphology to hypoplastic teeth frequently found in ED patients (Mark, 1996; Nowak, 1988; Till, 1992; Goepferd, 1981). Crowns and direct composite restorations are often used to provide proper contours on the hypoplastic teeth that will be used as abutments for removable prtial denture (RPDs) (Mark, 1996; Hogeboom, 1961; Till, 1992; Bolender, 1964; Alexander, 1969; Borg, 1977 and Cruz, 1981). In the present case, the patient is still actively growing and the pulp size is large as seen on radiographic examination. Hence FPD was contraindicated, but direct composite restorations were used to restore occlusal morphology.

Implant Prosthodontics

Recently concern has been expressed in the literature regarding placement of osseointegrated implants in a growing jaw.¹³ Studies carried out by Odman et al.³⁵ Thilander et al.³⁶ and Sennerby et al.³⁷ demonstrated that implants placed in the jaws of growing pigs do not behave like normal teeth but instead become ankylosed in the bone. It can be assumed that implants placed in children would behave similarly. Oesterle et al.³⁸ & Cronin et al.³⁴ concluded that implants placed after age 15 years for girls and 18 years for boys provided the most predictable prognosis. They also cautioned against the overgeneralization of clinical reports, because of the variation in growth among individuals, and recommended retrievability in implant-supported prostheses fabricated for children to facilitate adjustments related to growth and development.

According to Cronin et al.³⁴ and Oesterle et al.³⁸, possible outcomes of early implant placement are implant submergence because of jaw growth, implant exposure because of bone resorption associated with jaw growth, implant movement because of jaw growth, limitation of jaw growth if the implants are connected by a rigid prosthesis that crosses the midline. Thilander et al.³⁹ conducted a 3 year longitudinal study on single crown implants, and concluded that there was a positive correlation between craniofacial growth and infraocclusion of the restorations. Even though minor infraocclusion did occur in most of the restorations, they concluded that implants are acceptable for that age group (13 to 19 years) provided that growth and development is complete, with all teeth fully erupted. The dental and skeletal maturity, and NOT the chronological age of the patient is important to avoid infraocclusion of an implant restoration.³⁹ The literature therefore suggests that the timing of treatment is important if implants are placed in young ED patients because of possible complications resulting from jaw growth. In the present case, considering that the patient is still actively growing, the prosthesis will need remakes at regular intervals to keep up with the growth. The residual alveolar ridge is deficient. Also, the economic factors and risk (surgical morbidity) vs benefits are to be considered while planning for implants in such a patient. Hence, implant supported restoration was contraindicated in this case until completion of growth of the patient.

Removable Prosthodontics

The use of removable partial dentures (RPDs) is a reversible treatment that can significantly improve function and esthetics without endangering compromised dentitions (Pavarina, 2001 and Graser, 1990). Because anodontia or hypodontia is typical in individuals with this condition, complete dentures, partial dentures, or overdentures are often part of the treatment provided. Complete denture prosthesis given to patient alters the alveolar height, provides a better musculocutaneous profile and causes a considerable improvement in esthetics, mastication, phonetic function and psychological support. Retention can be enhanced with different impression technique as that for resorbed ridges. They can be remade if necessary, are cost effective, and the patient will get accustomed to dentures early, which will improve his confidence and self esteem. Although complete dentures can provide an acceptable esthetic and functional result, the underdevelopment of the edentulous alveolar ridges and maxillary tuberosities in individuals with ED can compromise denture retention and lateral and anteroposterior stability, they may need periodic adjustment due to growth changes (Akshay Bhargava, 2010). When there are teeth present for support overdentures are a desirable treatment option for these patients (Mark, 1996). When there are no teeth available for complete denture support, vestibuloplasty and ridge augmentation can be used to improve retention (Mark, 1996; Nowak, 1988; Thilander, 1994). Periodic recalls of young ED patients are also important because prosthesis modification or replacement will be needed as a result of continuing growth and development (Mark, 1996; Nowak, 1988; Goepferd, 1981; Sarnat, 1953 and Nomura, 1993). The occlusion of the prosthesis must also be monitored for changes because of jaw growth (Nomura, 1993). Other problems related to removable prostheses are speech difficulties, dietary limitations, and loss of the prosthesis

(Sarnat, 1953). The usual treatment for ED focuses on a series of complete or partial denture during the years when growth of the oro-facial region is taking place and definite rehabilitation after the completion of jaw growth. An interim acrylic upper removable partial denture and lower complete denture was fabricated for the present patient with frequent recalls and relining and rebasing or remake if necessary. Overdentures were not considered because tooth preparation cannot be done or telescopic crown cannot be given due to large pulp size. Instead, the conical teeth were modified with direct composite restorations.

Conclusion

The treatment of patients with ectodermal dysplasia is important from a psychological point of view as patients are shy or displeased with their appearance. The present patient and his parents were satisfied with the overall aesthetics and fit of the dentures. Recalls & Rebasing and Relining or Remake of denture is necessary at regular intervals to accommodate jaw growth. Definitive treatment can be planned (if necessary) after the growth has completed.

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