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Case Report

HED: prosthesis simplified-A case report

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Abstract

Ectodermal dysplasia is a genetic disorder of ectodermal origin causing deformities in derma tissues, odontoblast, hair follicles etc. Hypohidrotic ectodermal dysplasia (Christ-Siemens-Touraine syndrome) is the most prevalent type among 150 different variants of ectodermal dysplasia. A young girl reported with complete anodontia in maxillary arch, single molar tooth in mandibular arch, underdeveloped alveolar ridges and decreased salivation. This clinical report describes the restoration of the patient with reservoir upper denture and lower removable partial denture to alleviate dry mouth and improve esthetics and function.

Keywords: Anodontia, Hypodontia, Dry mouth, Reservoir, Artificial saliva

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1. Introduction

Ectodermal dysplasia is a one of the major hereditary disorder with developmental failure of two or more ectodermal structures. Skin, nails, hair, endocrine glands and teeth are primarily affected. General systemic findings include thin, scaly & dry skin with light pigmentation, Scalp and body hair is sparse whereas sweat and sebaceous glands are reduced in number, absent, or non-functioning and may result in elevated body temperature.

In almost 150 variants, Hypohidrotic ectodermal dysplasia also known as "Christ-Siemens-Touraine syndrome" is a major one that shows oral and facial manifestations predominantly in humans.[1-11] Affected individuals have characteristic facial features such as hyper pigmented skin around the eyes, large & squat ears, thick transposed lips, wrinkled face, forehead bump, hollow cheeks and saddle nose. The oral manifestations include hypodontia or anodontia with or without cleft lip and palate. Teeth present usually appear tapered, conical or pointed associated with poorly formed alveolar ridges and reduced lower facial height. These patients have senile appearance, slight mental retardation and are socially isolated.[12-20]

Early detection and management of oral & systemic manifestations of these patients will bring a tremendous change in their quality of life by improving their differences in speech, mastication, appearance and aids in the social and psychological wellbeing.[3,4,9,12-14] The effected individuals should seek a multidisciplinary approach of a general physician, speech therapist, psychiatrist , Dentist etc in formulating the treatment outcome. In general, in dentistry, A specialised prosthodontic approach is needed for the management of oral manifestations.

Prosthodontic treatments include removable partial dentures, complete dentures, fixed partial dentures and implant supported dentures.[6,7,14] Removable dentures are the most frequently reported treatment modality for dental management of ectodermal dysplasia patients.[3-6] Some ectodermal dysplasia patients suffer from decreased salivation due to aplasia or hypoplasia of salivary glands. Decreased salivation leads to difficulty in normal oral and

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orophyaryngeal functions including speaking, eating swallowing and extreme discomfort in wearing dentures.[21-23]

This clinical report describes the prosthodontic rehabilitation of a patient with hypohidrotic ectodemal dysplasia with maxillary reservoir denture to ease dry mouth.

2. Case Report

An 11-year-old female patient was reported to Department of Prosthodontics, Narayana dental college, India, because of unacceptable appearance, difficulty in eating food and dry mouth. Patient was not going to school because of unesthetic appearance. The patient exhibited classical features of hypohidrotic ectodermal dysplasia including sparse hair, eyelashes and eyebrows, pigmentation around the eyes, severe hypohidrosis, sunken nasal bridge with prominent forehead and everted lips (**Figure 1**).

Figure 1: Facial photograph of the patient showing the patient's characteristics of hypohidrotic ectodermal dysplasia

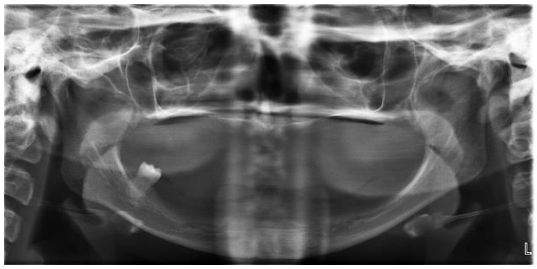

Figure 2: Panoramic radiographic image showing completely edentulous maxilla and a molar in right side of the mandible.

Figure 3: Master casts.

Clinical examination revealed completely edentulous maxilla and only one molar was present in right mandibular arch. The alveolar process of both the maxilla and mandible was poorly developed (**Figure 2**). The oral mucosa was normal in appearance and was dry due to hypoplasia of salivary glands. The palate was shallow and tongue was relatively large. Considering the age and clinical situation of the patient, a maxillary complete denture and mandibular removable partial denture were determined to be the treatment of choice. A reservoir for artificial saliva in maxillary denture was planned to improve comfort and deglutition of food.

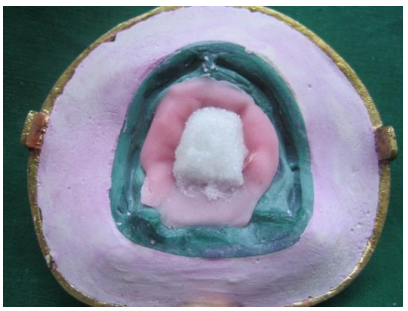

Figure 4: Putty adapted on the palatal side of the waxed denture to determine reservoir space.

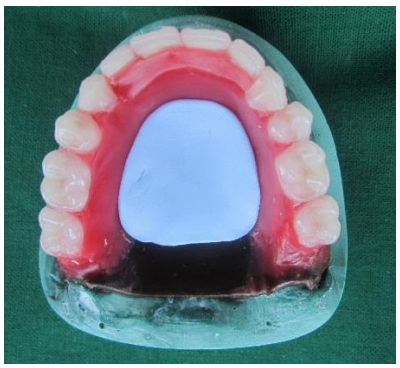

Figure 5: Salt placed in the palate region to create reservoir space during packing of the maxillary denture

Figure 6: Maxillary denture showing two pores in anterior palate for passage of artificial salivary substitute.

Figure 7: Facial photograph after insertion of the dentures.

Preliminary impressions were made with irreversible hydrocolloid (Zelgan; Dentsply, India). Custom trays were fabricated with autopolymerising resin (DPI-Cold cure:

Dental Products of India Ltd, Mumbai, India) and master impressions were made with light body addition silicone impression material (Elite HD+; Zhermack, Italy). Temporary denture bases were fabricated on the master cast (**Figure 3**) for recording maxillomandibular relations. The physiological rest position was determined by facial measurements and verified by phonetics. The casts were transferred to the articulator and teeth setting was done. Maxillary and mandibular trial dentures were tried in, vertical dimension of occlusion and centric relation was verified. Putty was adapted in the palate and functionally moulded by asking the patient to pronounce linguo palatal, linguoalveolar, sibilants and linguodental sounds for determining the reservoir space. Phonetics of the patient was verified in the trial denture (**Figure 4**). Approval of the esthetic appearance & exposure of teeth during smiling, fullness of cheeks were taken by patient and her parents .

Finishing and polishing of waxed dentures were flasked and subjected to compression molding technique.After wax elimination, Lost salt technique was employed in the fabrication of the reservoir (**Figure 5**). The waxed dentures were processed in heat polymerising acrylic resin (DPI-Heat cure: Dental Products of India Ltd, Mumbai, India). The dentures were retrieved after polymerisation, finished and polished. Two holes were placed in the anterior maxilla of maxillary denture and salt was flushed out with hot water (**Figure 6**). The resulting reservoir was filled with artificial salivary substitute (Wet Mouth; ICPA Health Products, Mumbai, India). The reservoir denture and mandibular removable partial denture were delivered and patient was trained to suck the artificial saliva from the reservoir in the denture. The patient and her parents were instructed about filling of the reservoir and appropriate home care. The patient was put on for 3 days follow up for any adjustments and then scheduled for regular recall appointments (**Figure 7**).

3. Discussion

Ectodermal dysplasia is a rare inborn disorder illustrated by malformations in tissues that are derived from ectoderm. Its prevalence in the population is about 1:100000. Hypohidrotic ectodermal dysplasia (Christ – Siemens-Touraine syndrome) is the most common form which involves hypodontia, hypohidrosis, hypotrichosis. Patients suffering from ectodermal dysplasia suffer from poor psychological and physiologic development due to unacceptable estehtics and abnormal oral function. Early and extensive prosthodontic treatment is required throughout childhood because of lack of most of deciduous and permanent dentition.[3-18]

Prosthodontic rehabilitation of the ectodermal dysplasia patient involves a series of removable partial dentures/complete dentures initially during growing years and a definitive prosthesis following completion of jaw growth.[12-16] Complete denture was planned in maxillary arch and a removable partial denture in mandibular arch was planned for this patient because she was still young to undergo rehabilitation with implants. Patient had difficulty in oral functions including eating, speaking and swallowing due to decreased salivation, hence a reservoir was planned in maxillary denture. The reservoir would hold approximately 3.2 cc of artificial salivary substitute. The flow rate of the saliva substitute was acceptable to the patient, and refill of the reservoir was required every 4 hours. The reservoir denture improved her symptoms of dry mouth and made her feel more comfortable. The patient's speech was unaltered as the reservoir space was determined during the try in stage of the waxed dentures. Patient gained confidence and rejoined the school. Patient was put on periodic recall every three months for further evaluation and any necessary prosthesis modification or replacement as a result of continuous growth of jaws

4. Conclusion

This clinical report describes the characteristics of hypohidrotic ectodermal dysplasia. Prosthetic rehabilitation of ectodermal dysplasia patient is recommended at an early age to encourage normal psychologic and physiologic development. Complete maxillary denture with reservoir and mandibular removable partial denture relieved dry mouth and improved esthetics, psychological, and masticatory function.

5. Source of Funding

None.

6. Conflict of Interest

None.

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