Cu sil Denture for Rehabilitation of Ectodermal Dysplasia

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ABSTRACT

Ectodermal dysplasia (ED) is an inherited disorder of ectoderm that results in aberrant development of tissues of ectodermal origin, namely skin, hair, nail, eccrine glands, and teeth. They present with missing teeth, sparse scalp hair, hyperpyrexia related to intolerance to heat.

Patients suffering from ED can lead normal life as it does not affect intelligence. But being bald and edentulous at any stage of life lowers the confidence of a person. Young children and adolescents go through the harsher scenario.

As this disorder manifests in infancy, prosthetic rehabilitation for missing and crooked teeth not only helps in, mastication, speech articulation, social acceptance and self-confidence.

This case report includes prosthetic rehabilitation of a 16-years-old male with ectodermal dysplasia with a complete denture on the lower arch and cu sil denture on the upper arch.

Key words: Cu sil denture, Ectodermal dysplasia, Partial anodontia

INTRODUCTION

E ctodermal dysplasia (ED) is a rare genetic disorder affecting ectodermal derivatives such as skin, hair, nail, eccrine glands, and teeth. Person suffering from this disorder have distinct features of sparse scalp and body hair, intolerance to heat, anodontia or oligodontia, abnormal or missing sweat gland. Other features of ectodermal dysplasia are prominent forehead, depressed nasal bridge, prominent lips, periorbital wrinkling and pigmentation and subcutaneous fat is often diminished or absent.¹.

Dental defects are rare in hidrotic ED but not in hypohidrotic ED. Patients suffering from ED have anodontia or oligodontia of permanent and deciduous teeth and if present, the teeth have

Conflict of Interest: None

*Corresponding Author Dr. Samjhana Maharjan; PG Resident, Department of Prosthodontics and Crown-Bridge, College of Dental Surgery, B.P. Koirala Institute of Health sciences E-mail: isam_mhj@yahoo.com a conical or truncated shape. Anodontia affects alveolar bone growth, so they have reduced lower facial height, class III profile due to returded maxilla and prominent mandible.

The disorder is usually diagnosed by the second year of life mainly due to lack of teeth eruption, hyperpyrexia related to intolerance to heat, sparse scalp and body hair. Prosthetic rehabilitation can enhance the quality of life.

In this case report, the patient with ED was provided with the complete denture in the mandibular arch and the cu sil denture in the maxillary arch. The cu sil denture is the acrylic denture with holes lined by soft relining material which allows natural teeth to emerge through it while maintaining peripheral seal², ³. It also had additional advantages of sealing food and fluids entering the denture, cushioning effect, and splints each natural tooth to hard denture base⁴.

CASE PRESENTATION

The 16-year-old patient, accompanied by his grandmother, visited the Department of

Prosthodontics and Crown Bridge of BPKIHS with a complaint of missing upper and lower teeth. Examination revealed a completely edentulous mandibular arch and a partially edentulous maxillary arch (only 3 teeth were present). Patient had distinctive facial feature of sparse scalp and facial hair, prominent lips, decreased lower facial height and protruded chin (Figure 1). Patient revealed that his deciduous as well as permanent never completely erupted (Figure 2). These features were classical characteristics of ectodermal dysplasia. Additionally, the patient had asymmetric facial features with the left side of the face slightly larger than the right side of the face.

He had a history of wearing a set of dentures for a few years, but it fractured during the COVID pandemic which crippled him in food selection, phonetics, and shyness to participate in normal activities for teenagers.

Intraoral radiograph of teeth revealed hypodense bone around the teeth and the roots of the teeth were conical with an apparently calcified pulp canal. Diagnostic cast revealed severe undercut on lingual area with knife ridge on mandibular arch and maxillary arch had low well-rounded residual alveolar ridge. So, after discussing with patient, complete denture was planned in lower arch and cu sil denture in upper arch.

After making primary impressions, custom trays were fabricated and border molding was performed. As the maxillary arch still had three remaining teeth, pick-up impression with alginate was done after border moulding (Figure 3). Jaw records were obtained, teeth arrangement was done and try-in was completed. After dewaxing, the undercut of the mandibular arch was blocked with putty (Figure 4). Then the remaining acrylization process was completed.

At the time of insertion, the holes around the retained teeth were enlarged by 4- 5 mm for easy removal and insertion of the denture. Border extension and occlusion were checked. Chairside soft relining material (mollosil[®]) was mixed and applied on space created earlier on both upper and lower dentures and the dentures were held in position until material sets. After material sets, dentures were removed and excess was trimmed (Figure 5)(Figure 6).

The patient was highly satisfied with the finished denture (Figure 7) (Figure 8). Oral and denture hygiene instructions were given and patient was recalled after 1, 3 and 6month for follow up.





Figure 1: Frontal and Lateral view



Figure 2: Intraoral View a) maxilla b) mandible



maxilla

Figure 6: Mandibular complete

denture with soft liner, intaglio



Figure 3: Pick up impression of Figure 4: Undercut blockout with putty in mandibular master cast



Figure 5: Maxillary Cu sil denture, intaglio surface



Figure 7: Post treatment intraoral view

Figure 8: Post treatment view of denture in situ

DISCUSSION

surface

Treatment for ectodermal dysplasia is still investigation. Administration under of recombinant protein therapy to pregnant mice has successfully treated the tabby pheonotype in offspring(Gaide et al 2003). Casal et al. reported that postnatal administration of recombinant EDA in dogs has successfully achieved normal tooth development, restored lacrimation, and improved sweating ability. ⁶ .These therapies have shown success in mice and dogs, but they have yet to be applied in humans.

In ectodermal dysplasia with partial anodontia, remaining teeth should be saved in any possible way because it is known that the presence of teeth reduces the amount of alveolar bone resorption and improves proprioception. Teeth supported and/or implant supported overdenture can be next option patient which significantly reduce amount of alveolar bone resorption over time and improve retention and stability(Crum et al 1978)⁸.

In this case, due to calcified pulp canals and presence of hypodense bone around teeth, overdenture was excluded⁹. Patient had financial restrictions for grafting and impant placement.

Cu sil dentures have been extensively used in cases with remaining natural teeth and when extraction or endodontic treatment is not a viable option (Jain et al. 2015). In this case, the cu sil denture was chosen as the patient had few remaining teeth that were retained to maintain proprioception and reduce the resorption of the alveolar bone.

Cu-sil denture has many advantages that include maintenance of peripheral seal, prevention of food slippage into the denture, cushioning effect and splinting of natural teeth to denture base. It also saves patient from tedious handling of clasps as in case of removable partial denture. The proprioception can be maintained by retaining the healthy teeth. Therefore, it can be viable alternative to extensive implant therapy when there are limited resources.

With the help of chairside soft liner and meticulous technique of fabrication the patient was satisfied and the denture were satisfactory in subsequent follow-ups.

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