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GROWING FACES, RENEWED CONFIDENCE: EARLY COMPLETE DENTURE REHABILITATION IN PEDIATRIC ECTODERMAL DYSPLASIA – A CASE REPORT

Paediatrics

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ABSTRACT

Ectodermal dysplasia (ED) is a rare hereditary condition affecting ectoderm-derived structures and commonly presents with hypodontia or anodontia, sparse hair, and reduced salivary function. Early prosthetic rehabilitation is vital to restore mastication, speech, esthetics, and psychosocial well-being. This case report describes complete denture rehabilitation in two children aged 3 and 5 years with severe oligodontia and anodontia managed in a pediatric dental setup. Clinical features included frontal bossing, depressed nasal bridge, protuberant lips, and reduced lower facial height. Following clinical and radiographic evaluation, complete dentures were fabricated using conventional impression techniques and careful jaw relation records, considering growth. Post-delivery follow-up showed marked improvement in mastication, speech clarity, facial profile, and social confidence. Early prosthetic intervention in ED significantly enhances functional and psychological outcomes, with regular follow-up essential to accommodate craniofacial growth.

KEYWORDS

Ectodermal dysplasia, complete denture, pediatric rehabilitation, oligodontia, anodontia.

INTRODUCTION

Ectodermal dysplasia (ED) is an inherited congenital condition characterised by defective development and function of two or more tissues originating from the embryonic ectoderm. The disorder commonly involves abnormalities of hair (sparse growth or alopecia), nails (dystrophic or malformed), teeth (hypodontia or anodontia), and sweat glands (reduced or absent function).1 The condition is thought to occur in approximately 1:10,000 to 1:1,00,000 live births and is more frequent in males. The majority of cases follow an autosomal recessive mode of inheritance, but it can also be autosomal dominant or X-linked.[2]

In ectodermal dysplasia, genetic mutations frequently disrupt critical signalling pathways responsible for odontogenesis, resulting in dental abnormalities. Variants in genes such as IKBKG, NFKBIA, and LTBP3 affect the EDA/EDAR/EDARADD/NF-κB signalling cascade, which plays a crucial role in enamel knot formation and tooth morphogenesis. Similarly, genes including TP63, CDH3, KDF1, PORCN, KREMEN1, LRP6, and TBX3 are associated with the Wnt/β-catenin pathway, essential for tooth initiation and developmental patterning. Mutations in TRPS1 may also contribute to dental defects through its interaction with the RUNX2 and OSX pathways involved in tooth mineralisation. Additionally, TSPEAR gene variants have been implicated in ED-related dental anomalies, although their precise biological role remains unclear.[3]

Ectodermal dysplasia is classified into hidrotic and hypohidrotic types. Both forms commonly present with dental and hair abnormalities but differ in nail involvement, sweat gland function, and inheritance patterns.4 The hidrotic variant (Clouston type) is characterised by hypotrichosis, nail dystrophy, and palmoplantar hyperkeratosis, with normal sweat gland function and autosomal dominant inheritance. In contrast, the hypohidrotic or anhidrotic form (Christ-Siemens-Touraine syndrome) presents with hypotrichosis, hypodontia or anodontia, and reduced or absent sweat glands, often accompanied by frontal bossing, saddle nose, and everted lips. This type follows an X-linked recessive pattern and is the most common, predominantly affecting males with greater severity.[2]

Orofacial manifestations of ectodermal dysplasia include hypodontia or anodontia with poorly developed alveolar ridges, leading to reduced lower facial height.5 Existing teeth are often conical, especially in the anterior region, with greater tooth absence in the mandible. The oral mucosa may be dry, and characteristic facial features include protuberant lips, frontal bossing, depressed nasal bridge, and hypotrichosis. In the primary dentition, maxillary second molars, canines, central incisors, and mandibular canines are commonly retained. Craniofacial findings may include maxillary retrusion, decreased anterior facial height, and a high-arched palate, occasionally associated with cleft palate.[6]

Early prosthetic rehabilitation is essential for restoring oral function, enhancing facial esthetics, and supporting normal psychological development in children with ectodermal dysplasia. Removable prostheses provide a practical and adaptable treatment option during growth, allowing periodic modification to accommodate craniofacial changes. This report emphasises the importance of early complete denture rehabilitation in young children with ectodermal dysplasia managed in a pediatric dental setting.

Figure 1. Preoperative clinical and radiographic findings in a child with Hypohidrotic Ectodermal Dysplasia: (A) Maxillary arch; (B) Mandibular arch; (C) Orthopantomogram showing complete anodontia.

CASE DESCRIPTION

Case 1

A 5-year-old male child presented with an inability to chew food due to the absence of teeth and reduced social interaction. He had been

diagnosed with hypohidrotic ectodermal dysplasia at 1 year of age based on clinical and genetic evaluation. The child was born to non-consanguineous parents with no significant family history. Clinical exome sequencing revealed a hemizygous likely pathogenic missense variant in exon 7 of the EDA gene (c.895G>A; p.Gly299Ser), consistent with X-linked hypohidrotic ectodermal dysplasia, along with a heterozygous variant of uncertain significance in the EDARADD gene.

General examination showed hypotrichosis with sparse scalp hair, eyebrows, and eyelashes; dry anhidrotic skin; periorbital hyperpigmentation; depressed nasal bridge; thick everted lips; and reduced lower facial height. Intraoral examination revealed complete anodontia in both arches with atrophic ridges and a shallow palate. The oral mucosa appeared dry, and the tongue was relatively enlarged. Orthopantomogram confirmed the complete absence of tooth buds with atrophic maxillary and mandibular alveolar ridges.

Prosthodontic management

Considering the patient's age and complete anodontia, removable complete dentures were planned for both arches to restore mastication, esthetics, speech, and psychosocial well-being. Preliminary impressions were made using irreversible hydrocolloid (Algitex, DPI, India), followed by custom tray fabrication and border moulding with low-fusing impression compound (Maarc Dental Pvt. Ltd., India). Final impressions were recorded using elastomeric material (FlexiGum A-Silicone, Waldent Innovations Pvt. Ltd., India) to accurately capture the atrophic ridges.

Jaw relations were recorded with careful evaluation of the vertical dimension in view of reduced lower facial height and ongoing growth. Trial dentures were assessed for esthetics, phonetics, lip support, and occlusion. Heat-cured acrylic complete dentures were fabricated, inserted, and adjusted to eliminate pressure areas. Post-insertion instructions were provided to the parents.

At 24-hour and one-week reviews, minor adjustments were made with satisfactory tissue response. Subsequent monthly follow-ups showed improved mastication, speech clarity, facial profile, and social confidence. Three-month recall visits were scheduled to monitor growth-related changes and the need for relining or prosthesis replacement. The child adapted well, with no significant complications.

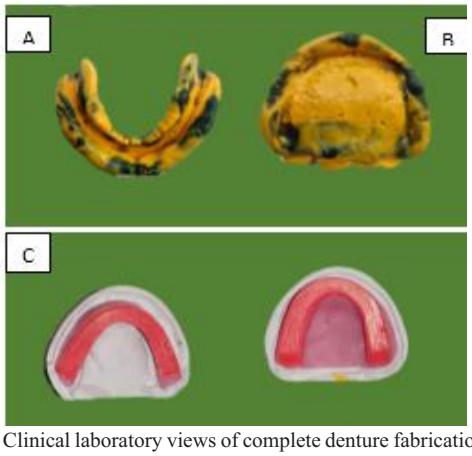
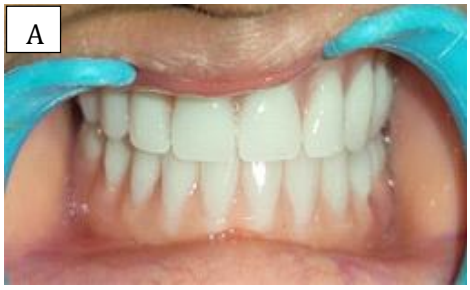


Figure 2. Clinical laboratory views of complete denture fabrication in a child with Hypohidrotic Ectodermal Dysplasia: (A) Mandibular impression demonstrating reduced ridge height; (B) Maxillary impression showing atrophic ridge morphology; (C) Trial denture bases adapted over master casts prior to processing.



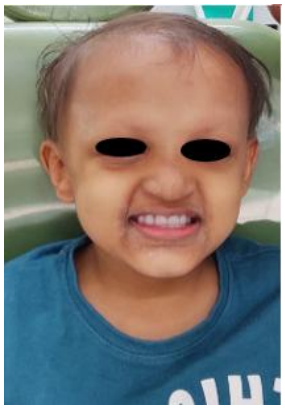


Figure 3. (A) Intra-oral photographs showing maxillary and mandibular arches (B)Photographs showing aesthetics of the denture.

Case 2

A 3-year-old male child presented with complaints of difficulty in mastication and reduced social interaction. The parents expressed concern regarding missing teeth and requested prosthetic rehabilitation. The child was born to non-consanguineous parents and had been diagnosed with hypohidrotic ectodermal dysplasia at the age of one year. Genetic analysis by clinical exome sequencing identified a hemizygous likely pathogenic missense variant in exon 8 of the EDA gene (c.1073A>T; p.Gln358Leu), confirming X-linked hypohidrotic ectodermal dysplasia.

On general examination, the child was moderately built and exhibited characteristic features of ectodermal dysplasia, including hypotrichosis and heat intolerance. The patient appeared shy and was initially uncooperative in the dental setting.

Intraoral examination revealed completely edentulous maxillary and mandibular arches, with an atrophic mandibular ridge and relatively adequate maxillary ridge height. The vertical dimension was reduced, accompanied by angular cheilitis and perioral/periorbital pigmentation. OPG demonstrated developing maxillary permanent first molars at Nolla's stage 6, with the absence of other tooth buds.

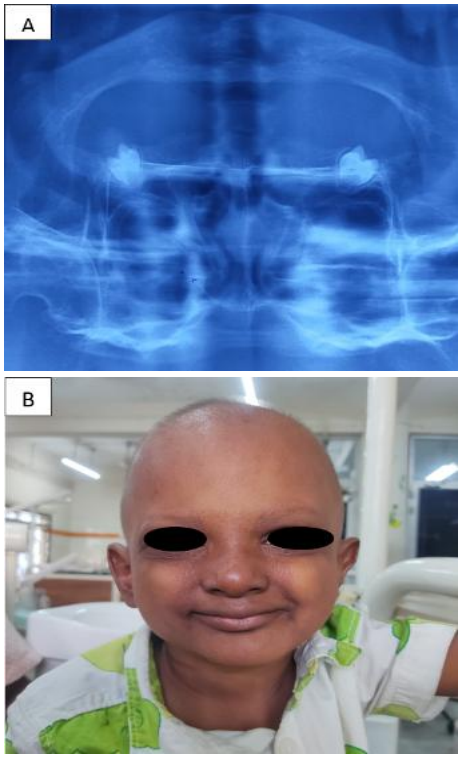


Figure 4. (A) Orthopantomogram demonstrating developing maxillary first molars (Nolla's stage 6) with absence of other tooth buds. (B) Extraoral view of a 3-year-old child with hypohidrotic ectodermal dysplasia showing characteristic facial features.

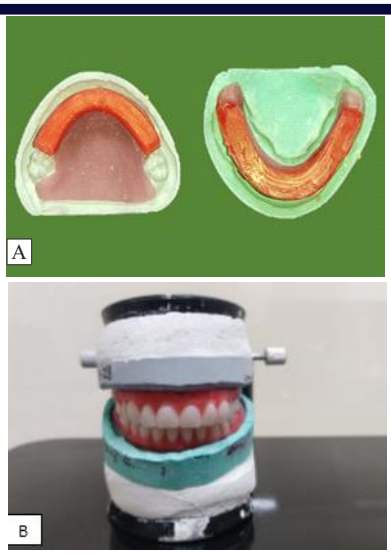


Figure 5. (A) Trial denture bases adapted over master casts prior to processing (B) Wax trial of maxillary and mandibular complete dentures mounted on an articulator

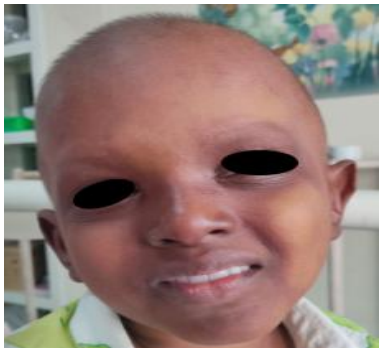


Figure 6. Post insertion Extraoral view.

Prosthodontic management

Complete denture rehabilitation was initially planned, anticipating the eruption of the maxillary permanent first molars. Owing to the child's shy and uncooperative behaviour, behavior management strategies including the Tell–Show–Do technique, positive reinforcement, and audiovisual distraction were employed to reduce anxiety and enhance cooperation during clinical procedures.

During jaw relation recording, an eruption bulge was noted in the maxillary tuberosity region, and the patient was kept under periodic review to monitor the eruption. After approximately 2.5 months, upon complete eruption of the maxillary first molars, a modified maxillary prosthesis was fabricated incorporating clasps on the erupted molars to improve retention. Preliminary and secondary impressions were made, followed by jaw relation recording, trial evaluation, and insertion of the prosthesis. The patient was placed on regular follow-up to assess adaptation, prosthesis stability, and growth-related changes.

DISCUSSION

Children with hereditary conditions such as ectodermal dysplasia often experience compromised psychological and physical development due to impaired esthetics and dysfunctional orofacial structures. Early intervention is therefore essential. Effective management requires a multidisciplinary approach involving pediatric dentists, psychologists, ENT specialists, and speech therapists. The dental treatment plan depends on the severity of the condition and must be tailored according to the patient's age, growth, and development of the stomatognathic system. The pediatric dentist plays a central role in addressing these special needs, with the primary objective of restoring function and esthetics to support normal physical, emotional, and social development.[6]

In young patients, complete dentures or removable partial dentures represent reversible treatment options that effectively improve function and esthetics without compromising developing oral structures.7 Complete denture therapy helps restore alveolar height, enhance facial profile, and improve mastication, phonetics, and psychological well-being. Management of ectodermal dysplasia typically involves interim removable prostheses during active craniofacial growth, followed by definitive rehabilitation after skeletal maturity.4 In the present cases, restoration of vertical dimension improved lip support and facial profile, consistent with previously reported outcomes. Endosseous implants may serve as an alternative treatment; however, their placement in growing patients remains debatable. Ongoing craniofacial growth can affect implant positioning and long-term stability, and implant-supported prostheses may require repeated modification, potentially increasing biomechanical stress. Current guidelines generally recommend delaying implant placement until skeletal growth is nearly or fully complete. Nonetheless, early implant therapy may be considered in selected cases of severe oligodontia or anodontia, particularly in the mandible.8 In the present cases, implant therapy was deferred due to the patient's young age and anticipated craniofacial growth. Behaviour management is essential in pediatric prosthodontic care. Techniques such as Tell–Show–Do and positive reinforcement enhance cooperation and treatment acceptance in young children.9 In addition, audiovisual distraction methods were employed during clinical procedures to further reduce anxiety and improve compliance.

Long-term follow-up is critical, as prostheses often require relining, adjustment, or replacement to accommodate craniofacial growth and eruption of permanent teeth. In the second case, eruption of the maxillary first molars required modification of the prosthesis, emphasising the need for periodic evaluation. Implant-supported prostheses may be considered once skeletal growth is complete.

These cases highlight the significance of early diagnosis, multidisciplinary management, and timely prosthetic intervention in improving functional efficiency and psychosocial well-being in children with hypohidrotic ectodermal dysplasia.

CONCLUSION

Early complete denture rehabilitation in children with hypohidrotic ectodermal dysplasia significantly enhances functional efficiency, facial esthetics, and psychosocial well-being, underscoring the importance of timely intervention and regular follow-up during growth.

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