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Case Report

Multidisciplinary Rehabilitation of Ectodermal Dysplasia Using 3D-Printed Crowns and Conventional Removable Dentures: A Case Report

Ektodermal Displazinin 3D Baskı Kronlar ve Geleneksel Hareketli Protezler Kullanılarak Multidisipliner Rehabilitasyonu: Bir Olgu Sunumu

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ABSTRACT

Introduction: Ectodermal dysplasia is a rare hereditary condition characterized by developmental defects in ectodermal-derived tissues and organs. In cases of ectodermal dysplasia, missing teeth (anodontia, hypodontia), conically shaped teeth, inadequate development of the alveolar crests, and loss of vertical dimension are observed. Due to the lack of most primary and permanent teeth during childhood, the esthetic and chewing functions of these patients need to be restored with a multidisciplinary treatment approach in the early period. This case report describes the rehabilitation of a child patient with ectodermal dysplasia using three-dimensional (3D) printed crown prostheses for existing upper central teeth and removable prostheses for additional missing teeth.

Case Report: The treatment of a 10-year-old child patient who applied to Gazi University, Faculty of Dentistry, due to missing teeth was carried out and completed jointly by the Departments of Pediatric Dentistry and Prosthodontics. The extraoral examination of the patient, who was diagnosed with ectodermal dysplasia, revealed sparse eyebrows, hair, and eyelashes, dry and thin skin, and sagging and swollen lips. Intraoral examination identified only two conical-shaped maxillary central teeth. All procedures to be performed were explained to the patient and their parents, and their consent was obtained. As a result of the prosthetic analysis, it was decided to make a crown restoration for the existing teeth and an upper and lower removable prosthesis for the missing teeth. After tooth preparations, permanent resin crowns were fabricated using 3D printing technology and cemented onto the prepared teeth. The removable prostheses for the upper and lower jaws were completed using traditional methods and delivered to the patient, who was scheduled for follow-up appointments.

Conclusion: Early multidisciplinary intervention is crucial for patients with ectodermal dysplasia starting from childhood due to significant tooth loss, aiming to restore both esthetics and functionality. The goal in designing prostheses is to preserve existing teeth, esthetically restore conical-shaped teeth, and prevent resorption of the alveolar ridges. The application of 3D printing in pediatric dentistry holds substantial potential, offering an aesthetically pleasing, durable, and cost-effective alternative for prosthetic restorations in child patients with ectodermal dysplasia.

Keywords: 3D printing; Additive manufacturing; Ectodermal dysplasia; Pediatric dentistry; Prosthetic rehabilitation

ÖZET

Giriş: Ektodermal displazi, ektodermal kaynaklı doku ve organların gelişimsel kusurları ile karakterize nadir bir kalıtsal durumdur. Ektodermal displazi vakalarında eksik dişler (anodonti, hipodonti), konik biçimli dişler, alveolar kretlerin yetersiz gelişimi ve dikey boyut kaybı gözlenir. Çocukluk döneminde birincil ve kalıcı dişlerin çoğunun eksik olması nedeniyle, bu hastaların estetik ve çiğneme fonksiyonlarının erken dönemde multidisipliner bir tedavi yaklaşımı ile restore edilmesi gerekmektedir. Bu olgu sunumu, ektodermal displazili bir çocuk hastanın mevcut üst santral dişleri için üç boyutlu (3B) baskı kronlar ve eksik dişler için hareketli protezler kullanılarak rehabilitasyonunu anlatmaktadır.

Vaka Raporu: Gazi Üniversitesi Diş Hekimliği Fakültesi'ne eksik dişler nedeniyle başvuran 10 yaşındaki bir çocuk hastanın tedavisi, Çocuk Diş Hekimliği ve Protetik Diş Tedavisi Anabilim Dalları tarafından birlikte gerçekleştirilmiş ve tamamlanmıştır. Ektodermal displazi tanısı konan hastanın ekstraoral muayenesinde seyrek kaşlar, saç ve kirpikler, kuru ve ince cilt, sarkık ve şiş dudaklar gözlenmiştir. İntraoral muayenede yalnızca iki konik biçimli maksiller santral diş tespit edilmiştir. Yapılacak tüm işlemler hasta ve ailesine açıklanmış ve onayları alınmıştır. Protetik analiz sonucunda mevcut dişler için kron restorasyonu ve eksik dişler için üst ve alt hareketli protez yapılmasına karar verilmiştir. Diş preparasyonunun ardından, 3D baskı teknolojisi kullanılarak daimi rezin kronlar üretilmiş ve prepare edilmiş dişlere simante edilmiştir. Üst ve alt çeneler için hareketli protezler geleneksel yöntemler kullanılarak tamamlanmış ve hastaya teslim edilmiştir; hastaya takip randevuları planlanmıştır.

Sonuç: Çocukluk döneminden itibaren ciddi diş kaybı olan Ektodermal displazi hastalarında, estetik ve fonksiyonelliği geri kazandırmayı amaçlayan erken multidisipliner müdahale çok önemlidir. Protezlerin tasarımındaki amaç, mevcut dişleri korumak, konik biçimli dişleri estetik olarak restore etmek ve alveolar kretlerin rezorpsiyonunu önlemektir. Çocuk diş hekimliği alanında 3D baskı uygulaması, Ektodermal displazili çocuk hastalar için estetik açıdan tatmin edici, dayanıklı ve maliyet etkin bir alternatif sunan büyük bir potansiyele sahiptir.

Anahtar Kelimeler: 3D baskı; Çocuk diş hekimliği; Eklemeli imalat; Ektodermal Displazi; Protetik rehabilitasyon

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INTRODUCTION

Ectodermal Dysplasias (EDs) are a varied group of congenital disorders characterized by anomalies in at least two ectodermal appendages, such as hair, teeth, nails, or sweat glands. Typical manifestations include abnormal hair growth (hypotrichosis), dental anomalies, dystrophic nails (onychodysplasia), and functional defects in the sweat glands (hypohidrosis or anhidrosis). The estimated incidence is 1 in 100,000 births, but this rate may vary and may be higher in certain populations. EDs present a wide array of inheritance patterns, which can include autosomal dominant, autosomal recessive, and X-linked dominant or recessive forms. These disorders can also occur sporadically due to mutations during intrauterine development, resulting in a wide range of clinical presentations.[1-3]

Ectodermal dysplasia is mainly classified into two types: Hypohidrotic/Anhidrotic ED and Hidrotic ED. The dentition and hair are similarly affected in both types of ED.[1,3] Dental anomalies are observed in 79% of individuals with ED, affecting both primary and permanent dentition.[4,5] The most common dental findings in individuals with ED include anodontia, oligodontia or hypodontia, tooth size and shape abnormalities, mineralized tissue defects, and tooth eruption issues.[6,7] The absence of primary and/or permanent teeth is the most frequently reported oral manifestation amongst these individuals. Hypodontia is prevalent, with tooth agenesis generally observed more frequently in lower jaw.[6] Shape deviations such as small size are also common. Incisors and canines often exhibit a conical shape, commonly described as “peg-shaped”. Second molars typically exhibit taurodontism due to the Hertwig’s epithelial sheath, which is of ectodermal origin, determining the root bifurcation site.[6,7]

In addition to oral manifestations, individuals with ED exhibit a generalized symmetric reduction of the craniofacial complex.[8] Dental agenesis and its impact on the growth and development of the jaws are significant clinical manifestations of ED. Maxillofacial growth is affected due to the absence of teeth, leading to craniofacial skeletal discrepancies. In sections without teeth, the alveolar edges display underdevelopment. Consequently, alveolar bone is thinner and less in volume compared to the alveoli surrounding the existing teeth. Insufficient height of the alveolar edges leads to a decrease in lower face height, thereby reducing the vertical dimension of occlusion.[7,9]

Recent advancements in three-dimensional (3D) printing technology have significantly expanded its applications in pediatric dentistry. In addition to its use in crown restorations, 3D printing has been employed in the fabrication of space maintainers, removable and fixed prostheses, and surgical guides, offering a more precise and patient-friendly approach. Compared to traditional methods, 3D-printed prostheses provide enhanced esthetics, better adaptation, and reduced treatment time, which is particularly beneficial in pediatric patients who may have limited tolerance for lengthy procedures. The ability to digitally design and rapidly manufacture customized prosthetic solutions makes 3D printing an increasingly viable option in modern pediatric dental care.[10]

Early prosthetic oral rehabilitation is recommended for these individuals to enhance orofacial development, restore functions such as chewing, swallowing, and phonetics, promote esthetics and psychological well-being, and minimize the impact on their quality of life. The rehabilitation treatment options for individuals with ED can vary widely and a multidisciplinary team approach is essential to the successful management of ED. This case report aimed to describe the rehabilitation of a child patient with ectodermal dysplasia using 3D printed crown prostheses for existing upper central teeth and removable prostheses for the additional missing teeth.

CASE REPORT

A 10-year-old male with Hidrotic Ectodermal Dysplasia was referred to the Gazi University, Faculty of Dentistry, Department of Pediatric Dentistry, for an examination due to the non-eruption of his permanent teeth. Extraoral examination revealed the characteristic features of ED, including brittle and fine blonde hair, narrow eyebrows and eyelashes, a depressed nasal bridge, a prominent forehead, periorbital and perioral pigmentation, protuberant lips, and an aged appearance (Figure 1).

An intraoral examination revealed the presence of only cone-shaped maxillary central incisors, with an-

Figure 1. Extra-oral view of the patient.

odontia in the mandible, low alveolar ridges, a loss of vertical dimension, and a reduced sulcus depth in the posterior region of the maxilla and mandible (Figure 2A, 2B). The radiographic examination confirmed the mandibular anodontia and the absence of all maxillary teeth except for two permanent maxillary teeth (Figure 2C).

The treatment plan included maintaining oral hygiene, crown restoration of the existing teeth, and fabricating maxillary and mandibular removable prostheses for the missing teeth. A consent form was signed by the parent of the patient.

After the permanent maxillary central teeth were prepared, impressions were made using an irreversible hydrocolloid (Tropicalgin; Zhermack, Badia Polesine, Italy) (Figure 3A). Following the fabrication of custom trays, border molding was established, and final impressions were made using a zinc oxide-eugenol impression paste (Cavex Outline; Cavex Holland BV, Haarlem, The Netherlands) material. Gypsum models were subsequently produced, and custom bases of acrylic resin were prepared. Finally, both the occlusal vertical dimension and the occlusal relationship were recorded.

Digital impressions were obtained from the plaster models of the maxilla and mandible using an intraoral optical scanner (Trios5, 3Shape, Denmark).

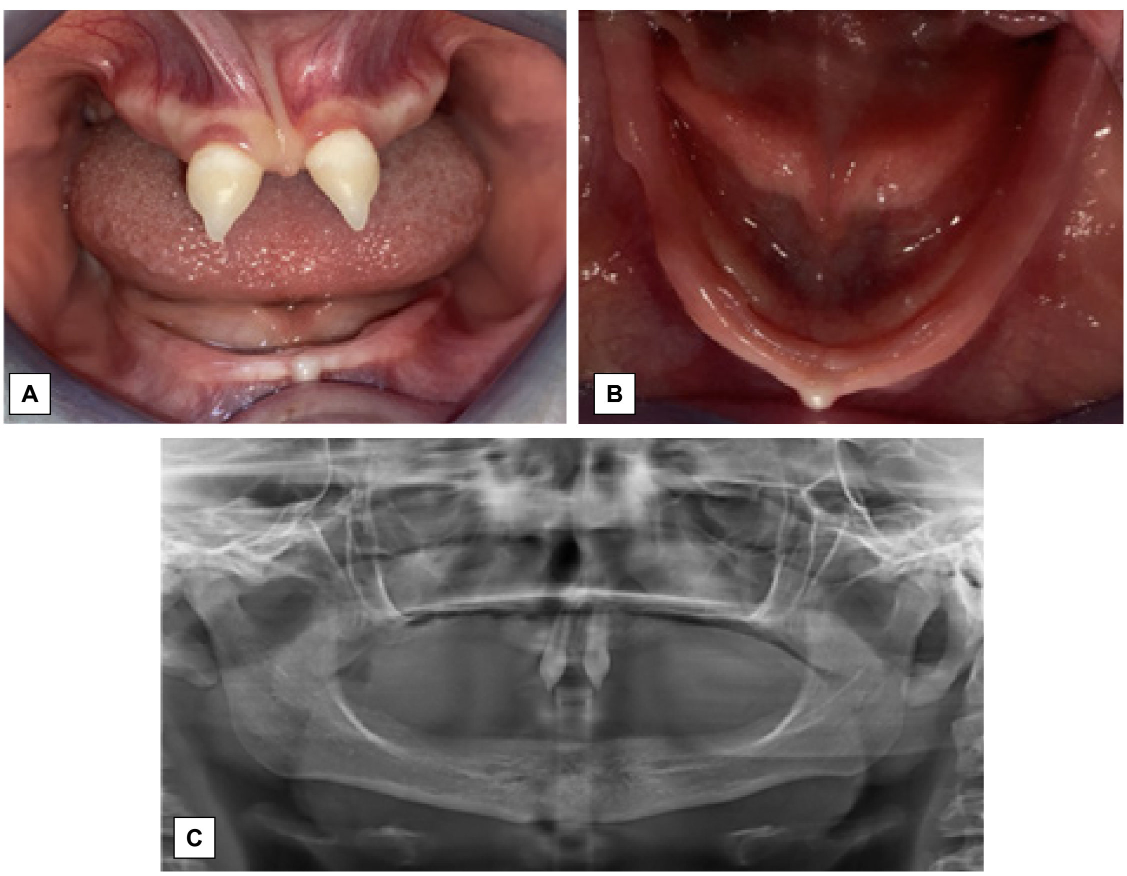

Figure 2. **A)** Intraoral preoperative view of the maxilla, **B)** Intraoral preoperative view of the mandible, **C)** Panoramic radiograph

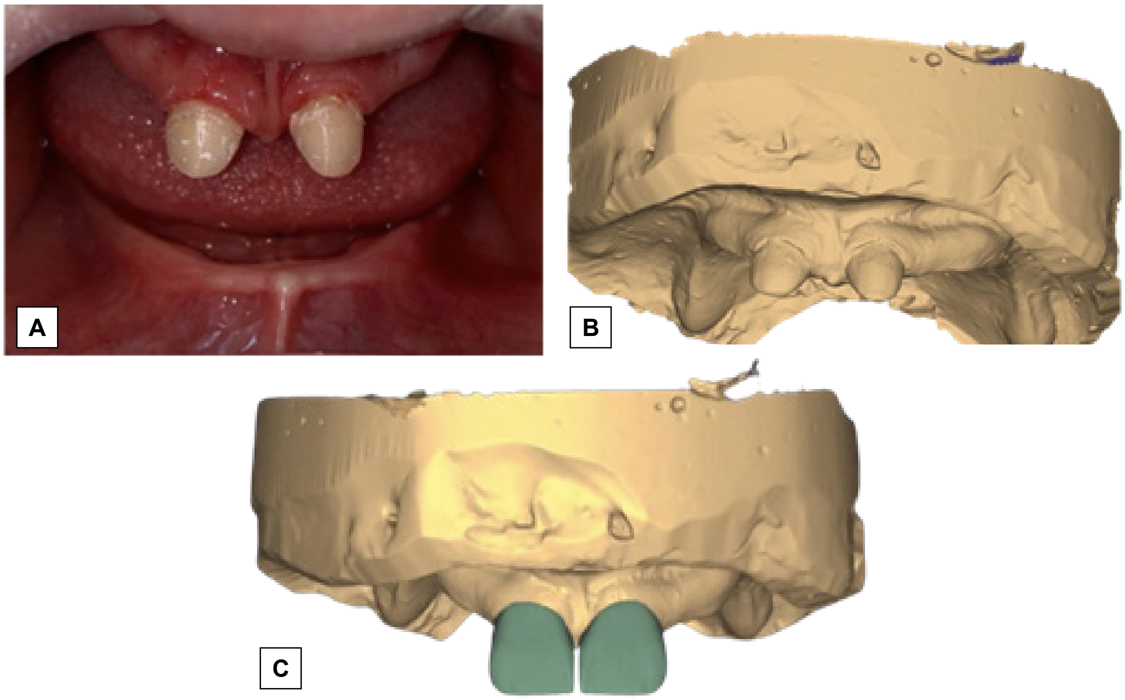

Figure 3. A) Preparation of cone-shaped maxillary central incisors, **B)** The digital impression of the prepared tooth, **C)** Crowns designed using software program

Afterward, these impressions were digitized to create virtual models (Figure 3B). Then, CAD software (exocad GmbH, Darmstadt, Germany) was used to design the crowns on the virtual model (Figure 3C). Following this, the crown designs were converted to the Standard Triangulation Language (STL) file format to allow for 3D printing, and the file was consequently sent to a laboratory to construct restorations via a 3D printer. Moreover, the STL file was incorporated into the PreForm software (Formlabs Inc., Somerville, MA, ABD) to automate the construction of the support structures. Ultimately, the crowns were printed using Permanent Crown Resin (Formlabs Inc) on a 3D printer (Formlabs Form 3B, Formlabs Inc).

The crowns were cleaned in the printer's washing unit (FormWash; Formlabs Inc) using isopropyl alcohol for 3 minutes after fabrication. Subsequently, they were subjected to a polymerization phase in the printer's curing unit (FormCure; Formlabs Inc). The structural supports were removed post-polymerization. An additional post-polymerization stage was carried out, involving a 20-min exposure at 60 °C within the same curing device. Surface finishing was performed according to the manufacturer's recommendations.

The tooth surface was prepared for cementation by applying a bonding agent (Scotchbond Universal Plus, 3M ESPE) and then air-drying. The inner surface of the restoration was prepared in accordance with the manufacturer's guidelines. Before cementation, the crowns were sandblasted with 50 µm Al_2O_3 particles for 20 s and then cleaned using an ultrasonic cleaner. The finished crowns were then silanized (G-Multi Primer; GC, Tokyo, Japan) and cemented using adhesive resin cement (G-CEM ONE; GC) (Figure 4).

Figure 4. Cementation of 3D-printed crowns

Figure 5. A) Wax try-in, **B)** Fabricated maxillary and mandibular removable prostheses

The arranged teeth were verified in the mouth during the wax try-in appointment (Figure 5A). After assessing occlusion, esthetics, and phonetics, the dentures were fabricated from acrylic resin (Figure 5B). After laboratory processing, the prostheses were placed in the patient's mouth, and necessary adjustments were made. Then, a direct polysiloxane soft liner (Mollosil Plus, Detax, Ettlingen, Germany) was used to improve the fit and comfort of the prosthetics. The removable prostheses were then delivered to the patient (Figure 6). Follow-up visits were scheduled at 6-month intervals to adjust the dentures, evaluate growth and development, and monitor oral hygiene.

Figure 6. Postoperative extra-oral view of the patient.

DISCUSSION

In this case report, the rehabilitation of a child with ED using 3D-printed crowns for the existing upper central teeth and removable prostheses for the additional missing teeth was presented.

The management of EDs often necessitates a multidisciplinary approach to address the various symptoms and complications, ensuring the maintenance of dental health. Restoring oral function and esthetics is critical for these patients, as it significantly impacts their psychological well-being. Comprehensive oral rehabilitation for these patients is essential from functional, physiological, and psychosocial perspectives.[1] The treatment should be planned by a team of specialists, including particularly pediatric dentists and prosthodontists.[2] Treating these patients involves several key points as growth and development (understanding the growth patterns and developmental milestones of pediatric patients), behavioral management (employing strategies to manage and support the behavior of young patients in a dental visit), prosthesis fabrication techniques (skills in creating prosthetic devices suitable for children), restorative techniques (modifying existing teeth using various restorative methods), motivating patients and parents (encouraging both the child and their caregivers to use and care for the prosthesis effectively), and long-term follow-up (providing ongoing care, including adjustments or replacements of the prosthesis as the child grows).[11]

Prosthodontic rehabilitation at an early age for pediatric patients with ED offers several advantages, including improved oral functions (chewing and speech), physiological benefits (self-esteem and social interactions), facilitated facial development (jaw growth and facial symmetry), prevention of oral issues (avoiding malocclusions and maintaining space), better adaptation to prostheses (ease of adjustment and habit formation), and improved quality of life (daily activities and overall well-being).[1,11-20] It is recommended that children with EDs use a dental prosthesis before they start school, typically around ages 3-4. Similarly, Schnabl *et al*.[12] stated that the median age for prosthetic treatment of ED patients was 4 years. In the presented case report, the child was 10 years old and had not previously worn prostheses, indicating a relatively late treatment age.

The prosthodontic rehabilitation of a patient with ED can involve the use of fixed or removable prostheses; however, each option requires specific considerations. Fixed partial dentures are less commonly used in the ED treatment due to the limited number of teeth. Fixed prostheses with rigid connectors can also hinder natural jaw development and cause further complications.[13,14] The most preferred method of treatment is using removable prostheses.[1] These prostheses can be adjusted and replaced according to growth.[11] However, clinicians may face some challenges. ED affects both primary and permanent teeth, usually resulting in conical or peg-shaped teeth.[14] Due to the absence of teeth, the alveolar ridges often remain underdeveloped, complicating denture fitting, retention, and stability. Ensuring dentures remain stable and function properly with less bone support is more challenging.[16]

Furthermore, children are continuously growing, necessitating frequent adjustments and replacements of dentures to accommodate changes in the alveolar bone and facial structure.[16] The knife-edge morphology of the alveolar ridges, coupled with decreased alveolar bone height, is a common observation.[1] Underdeveloped alveolar ridges and dry oral mucosa can compromise the retention and stability of prostheses. Frequent denture relining or replacement is necessary due to changes in the vertical dimension of occlusion and mandibular posture.[17] Bone augmentation procedures are not suitable for pediatric patients; therefore, soft liners can be used to improve the fit and comfort of the prosthetics.[16,17]

Successful treatments of children with ED were presented as case reports.[1,2,18-20] Abdulla *et al*.[1] presented the prosthetic treatment of a young boy with ED. In the treatment plan, the patient's conical maxillary anterior teeth were restored using composite strip crowns, and then removable partial dentures were delivered, and satisfactory results were obtained. Bolaca *et al*.[2] presented two cases with ED. In the first one, the child had two cone-shaped central incisors, and a maxillary overdenture and a mandibular complete denture were fabricated for the patients. The second patient was treated with maxillary and mandibular removable partial dentures. Both prosthetic treatments were reported as successful, resulting in significant enhancements in aesthetic outcome, speech, and mastication. These advancements were critical in dental prosthetic treatments, significantly improving the children's quality of life.

Török *et al*.[18] reported a prosthetic treatment of a 16-year-old boy with ectrodactyly-ectodermal dysplasia-cleft lip/palate syndrome (EEC). In this case, a telescopic retained overdenture was fabricated for the mandible. Impression was taken with an intra-oral scanner, and the telescopes were digitally designed. Selective laser sintering was then used to fabricate the primary and secondary telescopes. The combination of digital and traditional prosthetic methods resulted in an exceptionally effective overdenture.

Seremidi *et al*.[19] reported three clinical cases with a 2-year follow-up. The children were very young (3.5 years old), and exhibited severe oligodontia due to ED. The cases were treated with interim removable dentures to replace missing teeth, restore vertical dimension, and enhance function and esthetics. Two years post-treatment, patients and parents reported excellent adaptation to the prostheses and high satisfaction with esthetics. Montanari *et al*.[3] conducted a retrospective cohort study to assess skeletal growth, implant, and prosthetic survival rates, as well as success rates, and possible complications following a rehabilitation procedure involving a maxillary denture and an implant-supported overdenture attached by a sliding bar in ED patients. Over 7 years, nine patients received conventional dentures, followed by a maxillary denture and an implant-supported overdenture with a sliding bar connected to two implants in the anterior mandible. These patients were monitored for 3 to 12 years. The study concluded that mandibular growth near implant sites continues following their placement. Moreover, the implants provided effective support for prostheses in preteen patients with ED, demonstrating high success and survival rates with minimal complications. In patients with ED, the use of implant-supported prostheses is relatively rare, but they are still utilized. Sun *et al*.[20] documented the treatment of a 16-year-old patient with ED who had severe hypodontia, maxillary retrusion, and a thin knife-edge alveolar crest. The treatment involved distraction osteogenesis, a bone graft from the iliac crest, implant placement, and fabricating implant-supported overdentures. Despite requiring an additional implant during the 10-year follow-up, the treatment plan achieved a satisfactory outcome for the patient.

In the present case report, the remaining central teeth were restored with separated 3D-printed crowns. The crowns were not splinted to allow for natural jaw development. The use of 3D-printed resin crowns was selected for several reasons, including their precise customization for a perfect fit and improved comfort, quick production for time-efficient treatment, reduced cost, minimally invasive preparation procedure, and natural aesthetics to boost the child's smile and confidence.

In this case, the other missing teeth were restored with removable prostheses. Given the patient's age, a conservative treatment plan was preferred. To avoid possible trauma on the alveolar ridges, the mucosal sides of the prostheses were relined by using a soft denture liner. The patient was satisfied with the prostheses, and he expressed that he could comfortably use them. This treatment approach notably responded to the distinct needs of pediatric patients with ED, promoting both oral health and overall well-being. Due to the growth of alveolar ridges, prostheses may need modifications every 2 to 3 years, requiring relining or fabrication of new prostheses as necessary. Regular relining and rebasing of prosthetics should not discourage clinicians from intervening early in children's dental care. These procedures enable the child to develop normal speech, chewing, and swallowing functions, maintain proper facial support, and enhance temporomandibular joint function.[16] Thus, the patient has been scheduled for a follow-up every six months.

CONCLUSION

In managing prosthodontic rehabilitation for pediatric patients with ED, the choice of prosthesis depends on the individual's needs and anatomical considerations of the patient. Typically, removable prosthodontics are the most suitable option for growing children, while 3D-printed crowns can be viable alternatives to conventional treatments. Treating pediatric ED patients with fixed or removable prostheses not only improves their oral function and esthetics but also significantly enhances their psychological and social well-being. Regular clinical follow-ups and a personalized treatment approach are crucial to ensuring the best possible outcomes for these pediatric patients.

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