Early implant placement for a patient with ectodermal dysplasia: Thirteen years of clinical care

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Ectodermal dysplasia is a genetic disorder that is usually transmitted as an X-linked recessive trait.1-3 Ninety-five percent of patients with ectodermal dysplasia have the X-linked form.5 It is a rare disorder that can affect the development of the teeth, hair, nails, and sweat glands.1-3 The diagnosis of ectodermal dysplasia is often difficult to make in the first 2 years because the manifestations are difficult to detect.3 Missing teeth are often an important part of the diagnostic process. Dental manifestations of hypohidrotic ectodermal dysplasia include hypodontia, anodontia, and malformed teeth.1-3 In the absence of teeth, the alveolar ridge is compromised, making a removable prosthesis difficult to wear.4 In addition, patients with hypohidrotic ectodermal dysplasia often begin wearing a removable prosthesis as young as 3 years of age.5 These patients can experience difficulties with stability and retention of the removable prostheses because of the compromised alveolar ridge. Endosseous implants can improve the retention of the removable prostheses; however, the accepted standard regarding the timing of implant placement is that placement should be delayed until the cessation of growth.6-13 Severe situations of patients with ectodermal dysplasia have been reported as exceptions to this treatment guideline.5,7,9,14-17 In addition, the placement of implants helps maintain alveolar bone.18-22 Early treatments with dental implants can be considered when positive outcomes can improve the quality of life. If implant placement is planned, the potential complications must be discussed with the parents, who should also know that the advantages outweigh the risks.

Treatment of the dental manifestations of a patient with ectodermal dysplasia often begins at a young age. Not only is treatment often initiated at a young age but also managing care throughout the various transitions of dental development can be challenging. The objective of treatment is to improve function and esthetics; however, compromised alveolar development, the number of missing teeth, and the shape and position of the existing teeth are complicating factors that make the transitions of growth difficult to manage. Treatment requires a multidisciplinary team approach. A multidisciplinary team of

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ABSTRACT

Patients with ectodermal dysplasia have abnormalities of 2 or more structures that originate from the ectoderm. The oral manifestations often include the congenital absence of teeth and malformed teeth. This clinical report describes the interdisciplinary care from childhood through the definitive dental rehabilitation completed at skeletal maturation to replace the missing teeth in a patient with ectodermal dysplasia. Treatment began at 9 years of age with an implant-assisted mandibular overdenture to improve function and replace the missing mandibular teeth. Orthodontic treatment for the consolidation of space, composite resin restorations, and interim removable dental prostheses were provided to improve esthetics and replace the missing maxillary teeth. Skeletal growth was monitored, and orthognathic surgery was performed at the cessation of growth. The definitive rehabilitation consisted of a mandibular fixed dental prosthesis supported by dental implants and a maxillary removable dental prosthesis to restore the patient to esthetics and function. (J Prosthet Dent 2018;119:702-9)
specialists who work together to provide care throughout the phases of growth should be formed.23–25 The team of specialists should develop a long-term plan of care. The patient’s short-term needs should be addressed in conjunction with the long-term plan throughout the transitions of dental and skeletal development.

CLINICAL REPORT

A 9-year-old boy was referred for a consultation from his pediatric dentist in 2002. He had been previously diagnosed with the hypohidrotic form of ectodermal dysplasia. Clinical and radiographic examination revealed the absence of the majority of permanent dentition. The permanent teeth present included the maxillary central and lateral incisors and both the maxillary and mandibular first molars. In addition, the left maxillary primary molar was also present. The retained primary molar was ankylosed and the left posterior maxillary segment was impacted. The anterior mandibular alveolar ridge was compromised by the absence of primary and permanent dentition. The patient presented for a consultation regarding removable prostheses to replace the missing teeth. The patient’s mother stated that he was beginning to display concern regarding his smile and had recently asked about replacing his missing teeth. A multidisciplinary team of specialists comprising a prosthodontist, an orthodontist, and an oral and maxillofacial surgeon was formed in a university setting.

In 2002, composite resin (Herculite; Kerr Corp) was added to the maxillary central and lateral incisors to improve the shape and minimize the diastemas (Fig. 1). At that time, it was determined that the patient would benefit from the placement of 2 mandibular anterior implants to assist in the support of a mandibular overdenture. A diagnostic tooth arrangement was completed to facilitate the fabrication of a surgical guide. In 2003, 2 implants (3.25 mm Spline; Zimmer Biomet) were placed in the anterior mandible in the lateral incisor locations. The patient tolerated the procedure well. After 4 months of healing, the implants were uncovered and healing abutments (3.25×4.5 mm) were placed (Fig. 2). Two weeks later, O-Ring attachments (3.25×3 mm; Zimmer Biomet) that extended approximately 1 mm supragingivally were delivered using the O-Ring attachment seating tool (Fig. 3). A definitive impression was made in a border-molded custom tray with modeling plastic impression compound (Green Impression Compound; Kerr Corp) and polyvinyl siloxane impression material (Reprosil VPS Light Body and Medium Body; Dentsply Sirona). A maxillary alginate impression was made using a stock tray. A facebow transfer procedure and centric relation record was made using wax rims at the planned vertical dimension of occlusion.

A clinical evaluation of the mandibular overdenture was completed where the vertical dimension of occlusion, centric relation record, and esthetics were verified. An ideal occlusal plane was established with the mandibular overdenture; therefore, no occlusion was possible on the posterior left side. Orthodontic treatment was planned to address the impacted left posterior segment and the midline diastema. The mandibular overdenture was delivered and evaluated for tissue contact and retention of the O-Ring attachments (Fig. 4). Instructions were provided to the patient regarding placement and removal of the prosthesis. He was instructed to remove the prosthesis at night. At this point in treatment, the primary goal of a mandibular removable prosthesis was achieved, namely replacing the mandibular missing teeth and establishing a mandibular occlusal plane (Fig. 5). The patient was referred for orthodontic treatment to close the midline diastema, consolidate space, and evaluate the maxillary occlusal plane (Fig. 6).

In 2004, orthodontic treatment was initiated. A consultation with the orthodontist was completed to discuss the long-term prosthetic goals in conjunction with the orthodontic plan. The long-term prosthetic plan included complete coverage crowns on the maxillary right molar and the central and lateral incisors. Unfortunately, the maxillary left segment was severely compromised.
impacted and could not be repositioned with orthodontic treatment alone. In addition, the position and occlusal plane of the mandibular permanent molars were not favorable. Because the permanent molars were the only 2 teeth in the mandible, the long-term plan was to extract them.

To summarize, the maxillary left primary and permanent first molar and the mandibular molars were planned in the long-term for extraction. These 5 teeth were retained during the transitional years for the purpose of maintaining alveolar bone levels, orthodontic anchorage, and function. Orthodontic treatment successfully consolidated the anterior space, and the composite resin restorations were modified to reduce the mesial-distal width and minimize the midline diastema. The patient received a Hawley appliance for retention. The Hawley appliance also replaced the missing maxillary premolars and canines in this transitional phase of treatment (Fig. 7).

Between 2008 and 2010, the patient exhibited mandibular growth. This growth resulted in the wear of the teeth on the mandibular overdenture and an eventual Class III relationship (Fig. 8). In 2010, at 17 years of age, the patient experienced a sports-related injury where an orthopedic surgeon determined that his growth plates were closed and the chance of further growth was minimal. An assessment of skeletal maturation was initiated using lateral cephalometric radiographs. In 2011, a lateral cephalometric radiograph was exposed with the patient in an ideal vertical relationship. This revealed a true maxillary anterior-posterior hypoplasia that would require correction in order to facilitate definitive prosthetic restoration.

The patient’s maxilla was advanced using a Lefort 1 osteotomy, and stability was enhanced with grafts collected from the iliac crest (Fig. 9). Ideal positioning of the maxilla at the time of surgery was determined with interocclusal splints developed from mounted casts and a waxing of the expected position of the teeth when fully restored. Orthodontic appliances remained in place during surgery and for a period after surgery to control tooth position and aid the surgeon in maintaining proper maxillary stability. The definitive prosthodontics treatment plan to replace the missing teeth was a mandibular fixed, detachable, implant-supported prosthesis and a maxillary implant-assisted removable over-partial dental

Figure 3. Intraoral occlusal view of ball abutments in mandible.

Figure 4. Mandibular overdenture with O-Ring attachments.

Figure 5. Intraoral frontal view of first mandibular overdenture.

Figure 6. Frontal extraoral view at delivery of mandibular overdenture.

Figure 7.
prosthesis (RDP) using extracoronal precision attachments (Strategy-DE; Implant Direct).

In 2012, after the first surgical phase, a new mandibular overdenture was fabricated at the treatment vertical dimension of occlusion. In addition, a maxillary interim removable over-partial prosthesis was also fabricated that replaced the missing maxillary premolars and canines (Fig. 10). After a period of healing and postoperative orthodontics to finalize the tooth position, the maxillary left retained primary molar and impacted first permanent molar were removed; they had been maintained to assist with orthodontic anchorage and orthognathic surgery but were not restorable. Implants (3.5 mm Tapered Screwvent; Zimmer Biomet) were placed in the maxillary right premolar and maxillary left molar regions. Additionally, 3.5 mm diameter implants (Zimmer Biomet) were placed in the mandibular canine and central incisor sites, coordinating the position with implants in the mandibular lateral incisor position placed when the patient was 10 years old (Fig. 11). Adequate space between the mandibular lateral incisor implants was planned for the initial implant surgery while taking into account the space posterior to these implants and the mental nerve position. The spacing was relatively predictable, as no further significant growth was expected in the anterior mandible.

In 2014, crown preparations were completed on the maxillary right permanent molar and central and lateral incisors. The existing composite resin restorations were removed, and caries was detected on the maxillary incisors. Both maxillary lateral incisors and the right central incisor required endodontic treatment because of the extent of the caries. Definitive impressions for metal-ceramic crowns were made with polyvinyl siloxane impression material (Reprosil VPS Light Body and Heavy Body; Dentsply Sirona). Interim restorations were fabricated (Integrity Temporary Material; Dentsply Sirona) and cemented with interim cement (TempBond; Kerr Corp). The metal-ceramic maxillary crowns were fabricated at the treatment vertical dimension of occlusion and luted with resin-modified glass ionomer cement (RelyX Luting Plus Resin-Modified Glass Ionomer Cement; 3M ESPE). Locator abutments (Zest Anchors LLC) were seated and tightened to 30 Ncm for the maxillary implants (Fig. 12).

The maxillary RDP was designed using an extracoronal precision attachment system was used to assist
the cobalt-chromium removable partial denture prosthesis (Strategy-DE; Implant Direct) to provide maximum esthetics and avoid the use of conventional clasps, especially on the maxillary lateral incisor abutment teeth. The definitive impression for the RDP framework was made with irreversible hydrocolloid (Jeltrate Alginate; Dentsply Sirona). The framework fit was verified, and the steel housing and plastic female, yellow-coded inserts were seated intraorally to the cast high noble metal male configuration in the mesial aspect of the maxillary right first molar and in the distal aspect of the maxillary lateral incisors. Adequate prosthetic space was created between the acrylic resin intaglio surface of the RDP and the attachment housing on the abutment teeth to allow for a uniform pickup material thickness, thus ensuring a satisfactory pickup of the attachments (Fig. 13).

In conjunction with the maxillary RDP, the mandibular fixed detachable prosthesis was fabricated. As determined in the definitive treatment plan, the mandibular first permanent molars were extracted before making the definitive impression (Fig. 14). The original ball abutments were removed from the mandibular anterior implants, and tapered abutments were placed on all 5 mandibular anterior implants that extended no more than 1 mm supragingivally. Direct impression posts were placed on the 3 tapered screw vent implants, and indirect impression posts were placed on the original spline implants. The definitive impression was made in a custom tray with polyvinyl siloxane impression material (Reprosil VPS Light Body and Heavy Body; Dentsply Sirona). A titanium-milled framework was fabricated (Bella Tek Hybrid Bar; Zimmer Biomet 3i). Passive fit was verified clinically using the 1-screw test. The tooth arrangement was clinically evaluated with the mandibular fixed detachable prosthesis and the maxillary RDP.

The maxillary and mandibular prostheses were processed with heat-polymerized acrylic resin (Lucitone 199; Dentsply Sirona). The maxillary attachments were picked up intraorally using an autopolymerizing bis-acryl composite resin (EZ Pick Up; Sterngold). Once polymerization was complete, the RDP was removed and the housing’s attachment to the acrylic resin intaglio surface verified. Evident voids were filled with the same pickup material, and a smooth and convex surface was obtained surrounding the attachments. The framework fit and the occlusion were verified again after pickup to ensure the absence of discrepancies. The mandibular prosthesis was seated. The screw access holes in the mandibular fixed detachable prosthesis were sealed with cotton pellets and an interim resin material (Fermit; Ivoclar Vivadent AG). The definitive rehabilitation was complete at age 22 (Figs. 15, 16).
DISCUSSION

The 13-year dental treatment of a patient with ectodermal dysplasia is described in this clinical report. The patient initially presented for treatment at 9 years of age. An important aspect of the prosthodontic plan was to establish a team of practitioners who could work together to determine both a short- and long-term treatment plan. The treatment required a multidisciplinary team approach. Short-term care should be delivered with the long-term treatment plan in mind. In this particular patient, the long-term prosthodontic plan established initially was a mandibular fixed implant-supported prosthesis, complete coverage crowns for the maxillary first molar and the central and lateral incisors, and maxillary posterior implants. The initial plan included the placement of 2 mandibular anterior implants to assist a mandibular overdenture and orthodontic treatment to consolidate space in the maxillary anterior. Placing implants in the anterior mandible before skeletal maturation is indicated for the patient with ectodermal dysplasia because of the unique clinical presentation. Specifically, transverse growth of the anterior mandible is complete when the lateral incisors typically erupt, which is around age 6. Additionally, in the absence of teeth, vertical growth will not occur. The early cessation of transverse growth and the absence of vertical growth minimize the potential for complications resulting from early implant placement in the anterior mandible.

Early implant placement had 2 primary benefits for this patient. First, placement of implants can slow the resorptive process by changing the load mechanism on bone. In the long term, alveolar height and width will be necessary for implant placement and success of the fixed implant-supported prosthesis. Second, in the absence of teeth, the alveolar ridge is compromised, affecting the stability of a mandibular denture. The implants provide significant assistance with retention and stability of the resulting prosthesis. One important consideration when determining the position of the 2 anterior implants before skeletal maturation is that the long-term plan includes 5 implants in the anterior mandible. The addition of 3 anterior implants for the definitive rehabilitation will require space both between the implants and posterior to the implants to allow for the placement of 3 additional implants at the cessation of growth. The position of the initial 2 implants must be determined with the long-term plan to place 3 additional implants to support the definitive implant-supported prosthesis.

In the absence of permanent teeth, the primary teeth are often retained. Practitioners often need to make the decision regarding the timing of extractions of primary and nonfunctional permanent teeth. In this patient, the mandibular first molars and the maxillary left permanent molar and retained primary molar were maintained throughout growth until the definitive rehabilitation. Early on, the maxillary left segment was determined to be ankylosed, resulting in an unfavorable occlusal plane for the impacted first molar that was too significant to correct with surgical repositioning or conventional fixed restorations. Maintaining the left permanent and primary molar during growth and the transitional years, even though the long-term plan was altered and did not include these teeth, provided the benefit of anchorage for orthodontics by maintaining bone height and width as well as additional support for the removable prostheses.
During growth, the mandible grows downward and forward, and the maxilla should follow in a similar pattern. In this patient, a Class III malocclusion was noted at the cessation of growth. First and foremost, the treatment vertical dimension of occlusion should be developed to determine if there is a relative or absolute skeletal deficiency. Once the vertical dimension of occlusion is determined, a lateral cephalometric radiograph is exposed to assess the skeletal relationship. In this situation, a lateral cephalometric radiograph exposed with a maxillary and mandibular diagnostic tooth arrangement at the treatment vertical dimension revealed that the patient did present a maxillary deficiency; therefore, a Lefort I osteotomy was performed. The appropriate treatment method of addressing the class III malocclusion can only be determined by assessing the skeletal relationship at the treatment vertical dimension.

Several reports have discussed the implications of restoring implants before skeletal maturation. Because the implant does not respond to vertical growth, it is at risk of being submerged, resulting in soft tissue complications and a restoration in infraocclusion. Determining the completion of growth is important in treatment planning for implant-supported restorations in growing patients. Although several methods have been discussed in the literature for determining the cessation of growth, serial lateral cephalometric radiographs have provided valuable information regarding skeletal maturation. Cephalometric tracings superimposed from radiographs made at 6-month intervals that do not show change over a 1-year period are used to determine if growth is complete. An assessment of skeletal maturity is indicated for all growing patients with missing teeth; however, when implants are planned in proximity to permanent natural teeth, an assessment is critical. While it is accepted treatment to place implants in an edentulous mandible before the cessation of growth, implants should not be placed in a growing maxilla. Implant placement should be delayed in the maxilla until growth is complete. In the situation described, the maxillary implants were delayed until skeletal maturation was determined.

Treatment of these complex situations requires a multidisciplinary team approach. A multidisciplinary team of specialists should be formed to work together to provide care throughout the growth phases. A timeline of the treatment and development is presented in Figure 17 where the various treatment procedures for this patient are presented in line with skeletal development. Together, the team of providers should consider all treatment options. In this particular patient, the 2 alternative options considered for the maxilla included extracting the maxillary teeth and restoration with a complete-arch fixed-implant supported prosthesis. The patient chose to maintain his remaining teeth. For the mandible, maintaining the 2-implant overdenture and existing molars was considered an option. The patient preferred a fixed option; therefore, the fixed implant-supported restoration was selected. The mesial inclination of the mandibular molars limited the posterior extent of the implant-supported restoration, and the molars extracted. Once rehabilitation is complete, a follow-up recall plan to monitor the outcome of treatment should be established.

Figure 17. Timeline of development and treatment.
SUMMARY

The dental rehabilitation of a patient with ectodermal dysplasia presents several challenges to the practitioner. The team of dental specialists must establish both short- and long-term goals and work together to achieve a favorable result. Communication between the various practitioners at all stages is critical to accomplish a positive outcome. This report presents early implant placement in a 9-year-old boy with ectodermal dysplasia and the transitional treatment completed throughout skeletal maturation over the course of 13 years to achieve definitive rehabilitation. Early implant placement provided the benefit of a functional mandibular prosthesis assisted by 2 anterior implants throughout growth. The location of the early mandibular anterior implants was planned and carried out to allow for the addition of 3 additional implants at the cessation of growth to support the definitive mandibular fixed implant-supported prosthesis. Care is administered at each stage of treatment that is guided by the long-term goals established by the team of providers.

REFERENCES


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