

## Six-Year Survival of a Mini Dental Implant-Retained Overdenture in a Child with Ectodermal Dysplasia

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### Keywords

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### Abstract

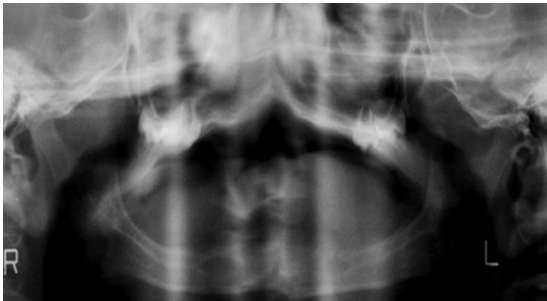
Patients with ectodermal dysplasia (ED) experience several problems caused by abnormal development and functioning of the head and neck region. In addition to developmental nasal cartilage abnormalities and absence of sweat glands, hair, and eyebrows, edentulism or developmental disorders of teeth (cone-shaped teeth) are commonly observed in these types of patients. ED is also characterized by underdeveloped alveolar ridges, a decreased occlusal vertical dimension, reduced salivary secretion, and dry oral mucosa, which make prosthetic rehabilitation difficult. Few studies of intraosseous dental implant-retained prostheses have described adverse effects on craniofacial growth and esthetic and functional disadvantages, while some researchers have described the advantages of this treatment option as an alternative option in these cases. Due to the associated alveolar bone deficiency, dental mini-implant therapy may be a treatment option for these patients; however, there are isolated cases in the literature regarding the rehabilitation of ED patients with mini-implant-supported overdentures. This clinical report describes the rehabilitation of a 6-year-old child with ED using a maxillary removable partial prosthesis and a mini-implant-retained mandibular overdenture. The clinical and radiographic findings of this prosthetic rehabilitation during the 6-year follow-up are also presented.

Ectodermal dysplasia (ED) is a hereditary disease first described by Thurman and characterized by the abnormal development of tissues of ectodermal origin, such as skin, nail, hair, and teeth.<sup>1</sup> Hypohidrotic ectodermal dysplasia (HED), the most common type of ED, is also known as Christ-Siemens-Touraine syndrome or anhidrotic dysplasia.<sup>2</sup> Nail dystrophy, alopecia, or hypotrichosis (sparse, shiny hair or eyelashes), lack of sweat glands, and palmar-plantar hyperkeratosis are commonly seen abnormalities in patients with HED, which is seen in 1 in 5000 to 1 in 10,000 births.<sup>3</sup> The disease can be inherited in an autosomal dominant, recessive, or X-chromosomal manner and is more common and severe in males than in females.<sup>4</sup> These patients, with typical features such as a prominent forehead and chin, flat nose, and thick lips, may face psychosocial problems due to their appearance.

ED is characterized by anodontia or oligodontia (cone-shaped teeth), underdeveloped alveolar ridges, a decreased occlusal vertical dimension (OVD), reduced salivary secretion, and dry oral mucosa, making prosthetic rehabilitation difficult.<sup>5,6</sup> For many years, early application of total or partial removable prostheses was used to ensure esthetic appearance, phonation, and function.<sup>7,8</sup> However, increases in dental caries, periodontal problems, and complications such as alveolar ridge resorption have prompted researchers to consider alternative treatment options. A few studies of intraosseous dental implant-retained prostheses have described adverse effects on craniofacial growth and esthetic and functional disadvantages,<sup>9,10</sup> while some researchers have described the advantages of this treatment option as an alternative option in these cases.<sup>11-13</sup>



**Figure 1** Extraoral view of the patient.



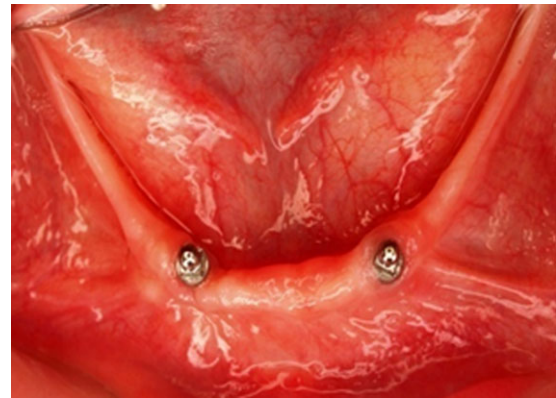
**Figure 2** Preoperative radiographic view of the patient.

The purpose of this report was to evaluate the 6-year survival of mini dental implants after rehabilitation with a ball-attachment-retained mandibular overdenture in a patient with ED.

## Clinical report

A 6-year-old boy with ED was referred to the Karadeniz Technical University (KTU) Faculty of Dentistry, Department of Prosthodontics, due to esthetic, functional, and phonetic dysfunctions. Extraoral examination revealed the characteristic features of ED syndrome, including hypotrichosis, prominent forehead, flat nose, thick lips, prominent chin, and sparse hair and eyelashes (Fig 1). Intraoral findings revealed an edentulous mandible and a maxilla with two first molars with severe alveolar ridge atrophy. Radiological examination supported the clinical findings and revealed two unerupted second maxillary molars and advanced alveolar bone resorption (Fig 2). Cephalometric evaluation revealed a reduced vertical dimension.

Treatment options considered for this child after clinical consultations with the KTU Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, included constructing a mandibular overdenture prosthesis supported by two mini-implants and a clasp-retained removable maxillary partial



**Figure 3** Postoperative positive healing, 4-month early follow-up.

denture. A written informed consent form was signed by the patient. Two mini-implants (Endure MDI/ImtecCorp<sup>®</sup>; 3M ESPE, St. Paul, MN) 2.9 mm in diameter and 13 mm in length were inserted without complication in the canine region of the anterior mandible under general anesthesia in the KTU Faculty of Medicine Farabi Hospital. Following a 4-month uncomplicated healing period, clinical and radiographic examination established that the two mini-implants were successfully osseointegrated (Fig 3).

Preliminary impressions were made for a custom-made tray using irreversible hydrocolloid, as soon as the soft tissues adjacent to the abutment cylinder healed. Definitive impressions of both arches were made using polyether impression material (Impregum Duo Soft; 3M ESPE) with custom trays. Maxillomandibular relationship records were obtained, and the casts were mounted in an articulator. The artificial teeth were arranged in wax for trial evaluation. Primary tooth shaped prosthetic teeth were selected to provide an age-appropriate appearance. Centric relation position and OVD were verified. The positions of the prosthetic teeth were evaluated intraorally, and the necessary corrections were made. The waxed trial dentures were processed in a heat-polymerized denture base resin, and matrix attachments were cured into the overdenture. A ball-attachment mandibular overdenture and a traditional maxillary removable partial prosthesis were applied for prosthodontic rehabilitation of lost tissues, adjustment of OVD, and elimination of functional and esthetic problems (Fig 4).

The patient and parents were instructed to maintain a soft diet for the first few days to facilitate conformity; the necessity of regular cleaning and maintenance was also explained. The patient was trained to place and remove the prosthesis properly and was instructed to take it out at night. One-day, 3-day, 1-week, 1-month, 3-month, and 6-month follow-up appointments were then scheduled. Following prosthetic complication-free follow-up periods of 15 months, 5 years, and 6 years, with cephalometric evaluation of the growth pattern of the jaw, dentures were renewed three times using the same treatment methods (Fig 5). This procedure allowed seamless normal growth of unerupted maxillary second molars, and the eruption of these molars was thus completed uneventfully.

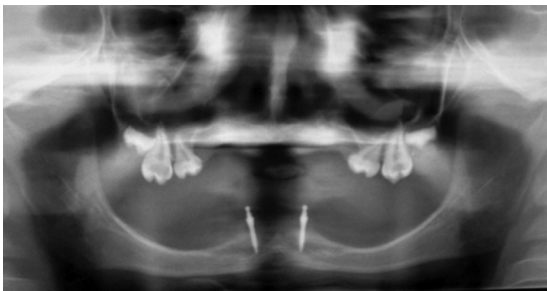
During the 6-year follow up period, no evidence was observed of intensive resorption in the peri-implant bone, and no



**Figure 4** Intraoral views of final dentures.



**Figure 5** Patient's growth pattern using cephalometric evaluation at 5 years.



**Figure 6** Radiographic view of patient at 6-year follow-up.

prosthesis-related functional and esthetic problems occurred (Figs 6 and 7). Prosthetic rehabilitations motivate child patients during the growing period, since they satisfy functional and esthetic requirements.



**Figure 7** Extraoral view of patient at 6-year follow-up.

## Discussion

Patients with ED are exposed to several problems caused by abnormal development and functioning of the head and neck region. In addition to developmental nasal cartilage abnormalities and absence of sweat glands, hair, and eyebrows, edentulism or developmental disorders of teeth are commonly observed in these patients. In addition, due to xerostomia in the oral cavity and oropharyngeal area, difficulty in moving food particles and swallowing are the main functional complaints reported.<sup>14</sup> These patients should therefore be specifically evaluated for oral function. This situation should be considered during the treatment planning process, and continuity of personal oral care should be established.



The most important objectives of dental treatments are to replace soft and hard tissue loss and restore missing teeth, to adjust the vertical dimension, and to support the facial soft tissues. In cases of ED, oligodontia is more common (80%) than anodontia, and complete dentures, removable partial prostheses, fixed partial prostheses, or implant-retained dentures can be selected to eliminate functional and esthetic problems.<sup>15,16</sup>

In many cases of alveolar ridge atrophy or knife-edge alveolar crest caused by the congenital absence of teeth, retention and stability of complete or removable partial dentures can be lost due to inadequate bone support. Furthermore, some studies have reported that implant-retained prostheses with their high success rates may represent an alternative to traditional removable dentures.<sup>17-19</sup> Considering the age and growth pattern of our patient, a dental implant prosthesis was an appropriate option for long-term prosthetic prognosis. The most important advantage of this modality is better esthetic and functional outcomes due to improved retention and stability.<sup>20</sup> However, it should be kept in mind that the lack of adequate bone mass/volume may prevent the implant placement process in cases of ED, which frequently present with alveolar ridge resorption as a result of congenitally missing teeth. In this case, a relatively narrower diameter intraosseous mini dental implant procedure was performed due to the insufficient vestibulo-lingual bone thickness of the mandible.

Many researchers have reported potential problems related to intraosseous implant procedures in growing children.<sup>11,21-24</sup> In contrast to natural teeth, there is no capacity for physiological movements and compensatory eruptions of intraosseous implants due to the absence of periodontal ligaments. Therefore, osseointegrated implants are considered to behave similarly to ankylosed teeth and become submerged due to the growth process associated with continued eruption of adjacent natural teeth.<sup>25</sup> Kearns *et al* suggested that partially edentulous patients who are still growing should be evaluated carefully before placement of dental implants, and that intraosseous implant applications are preferable in patients with anodontia.<sup>20</sup> Since lateral growth of the anterior mandible is usually completed at 3 years of age, and due to the lower vertical growth rate, the implant-retained prosthesis can be considered in patients who are edentulous in this region.<sup>15,16,20</sup>

For preserving residual alveolar bone, dental implants could be installed in the posterior mandibular area; however, ongoing growth and development should be considered. Several problems, such as infraocclusion and multidimensional dislocation when compared with the developing teeth, may occur due to installation of dental implants in a growing patient.<sup>26</sup> In the posterior mandible, growth changes occur in late childhood with large amounts of anteroposterior, transverse, and vertical growth. When compared with the posterior mandible, alveolar growth seems relatively small when teeth are missing in the anterior part of the mandible. The anterior mandible may be the most suitable site for implant placement in children with severe hypodontia.<sup>26,27</sup> Some researchers have described the intraosseous implant as one of the most important treatment options in growing patients because of physiological conservation of bone tissue.<sup>28</sup> The transverse and vertical growth pattern did not differ significantly between implant-treated and

non-treated children with ED, and successful results can be expected with implant-supported prostheses.<sup>26</sup> Our objective in the present case was to alter the loading mechanism and to minimize bone loss by placing intraosseous dental implants in the mandible, in which congenital anodontia and severe alveolar ridge resorption were present.

Some researchers have emphasized the necessity of renewing the prosthesis to ensure adaptation to the changes in the occlusal plane during the growth period. Prosthetic treatment should be designed to compensate for and adapt to changes in growth pattern.<sup>22</sup> Considering the rapid growth pattern in young individuals, the tooth or implant-retained removable denture option is usually suggested as a therapeutic option. In this report, following insertion of two mini-implants in the anterior mandible, a removable ball-attachment-retained overdenture and removable partial maxillary prosthesis were installed. At 15-month, 5-year, and 6-year follow-ups, renewal of the prostheses was considered with the aid of cephalometric evaluations of the growth pattern and the changes in the occlusal plane. The normal growth pattern was thus maintained with this procedure, and the eruption of the maxillary second molars was completed uneventfully in this case.

Bone resorption in both the maxilla and mandible is a foregone conclusion for patients treated with a removable prosthesis due to the hypodontia or anodontia. Because these patients start using removable prostheses at an early age, they may have severe resorption in alveolar bone. After adolescence, rehabilitation of a patient with dental implants not only supports a prosthesis, but also maintains the alveolar bone in the long term; however, severe bone resorption can be a major challenge for implant insertion in edentulous ED patients.<sup>29</sup> Several methods have been used to augment bone volume in ED patients, including the use of vertical distraction osteogenesis, autogenous bone grafts, allogenic and xenogenic bone grafts, and a combination of these modalities.<sup>29-31</sup> Zygomatic implants can be an alternative for rehabilitation of ED patients.<sup>31</sup> Previous studies demonstrated that dental implants and bone grafts are valuable tools for oral rehabilitation of ED patients for improved clinical outcome.<sup>29-31</sup>

Numerous studies and reports have described the rehabilitation of patients with ED using dental implants.<sup>11,13,15,26,32,33</sup> However, there are only limited reports of mini dental implants in children with ED.<sup>16,34</sup> Our report is therefore important in describing a long-term (6-year) follow-up of a 6-year-old child with ED syndrome treated using two mini dental implant-supported mandibular overdentures.

## Conclusion

The multidisciplinary approach, prosthetic treatment, and 6-year follow-up period described have resulted in patient satisfaction in terms of esthetic appearance and oral function. The placement of dental mini-implants allowed the patient to accept and handle removable dentures easily. The prosthetic rehabilitation with dental mini-implants is an appropriate technique for this case, and it may further psychologically motivate child patients in the social environment during the growing period.

## References

- Priolo M, Lagana C: Ectodermal dysplasias: a new clinical-genetic classification. *J Med Genet* 2001;38:579-585
- Baskan Z, Yavuz I, Ulku R, et al: Evaluation of ectodermal dysplasia. *Keio J Med* 2006;22:171-176
- National Foundation for Ectodermal Dysplasias: What is hypohidrotic ectodermal dysplasia? [http://nfed.org/index.php/about\\_ed/hypohidrotic-ectodermal-dysplasia](http://nfed.org/index.php/about_ed/hypohidrotic-ectodermal-dysplasia). Accessed April 11, 2015
- Bergendal B, McAllister A, Stecksen-Blicks C: Orofacial dysfunction in ectodermal dysplasias measured using the Nordic Orofacial Test-Screening protocol. *Acta Odontol Scand* 2009;67:377-381.
- Suri S, Carmichael RP, Tompson BD: Simultaneous functional and fixed appliance therapy for growth modification and dental alignment prior to prosthetic habilitation in hypohidrotic ectodermal dysplasia: a clinical report. *J Prosthet Dent* 2004;92:428-433
- Pavarina AC, Machado AL, Vergani CE, et al: Overlay removable partial dentures for a patient with ectodermal dysplasia: a clinical report. *J Prosthet Dent* 2001;86:574-577
- Herer PD: Treatment of anhidrotic ectodermal dysplasia: report of case. *ASDC J Dent Child* 1975;42:133-136
- Itthagarun A, King NM: Oral rehabilitation of a hypohidrotic ectodermal dysplasia patient: a 6-year follow-up. *Quintessence Int* 2000;31:642-648
- OpHeji DG, Opdebeeck H, van Steenberghe D, et al: Age as compromising factor or implant insertion. *Periodontol* 2000 2003;33:172-184
- Rossi E, Andreasen JO: Maxillary bone growth and implant positioning in a young patient: a case report. *Int J Periodontics Restorative Dent* 2003;23:113-119
- Guckes AD, Brahim JS, McCarthy GR, et al: Using endosseous dental implants for patients with ectodermal dysplasia. *J Am Dent Assoc* 1991;122:59-62
- Lekholm U: The use of osseointegrated implants in growing jaws. *Int J Oral Maxillofac Implants* 1993;8:243-244
- Smith RA, Vargervik K, Kearns G, et al: Placement of an endosseous implant in a growing child with ectodermal dysplasia. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1993;75:669-673
- Mehta U, Brunworth J, Fete TJ, et al: Head and neck manifestations and quality of life of patients with ectodermal dysplasia. *Otolaryngol Head Neck Surg* 2007;136:843-847
- Guckes AD, Scurreia MS, King TS, et al: Prospective clinical trial of dental implants in persons with ectodermal dysplasia. *J Prosthet Dent* 2002;88:21-25
- Guler N, Cildir S, Iseri U, et al: Hypohidrotic ectodermal dysplasia with bilateral impacted teeth at the coronoid process: a case rehabilitated with mini dental implants. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2005;99:E34-E38
- Adell R, Lekholm U, Rockler B, et al: A 15-year study of osseointegrated implants in the treatment of the edentulous jaw. *Int J Oral Surg* 1981;10:387-416
- Zarb GA, Symington JM: Osseointegrated dental implants: preliminary report on a replication study. *J Prosthet Dent* 1983;50:271-276
- Albrektsson T: A multicenter report on osseointegrated oral implants. *J Prosthet Dent* 1988;60:75-84
- Kearns G, Sharma A, Perrott D, et al: Placement of endosseous implants in children and adolescents with hereditary ectodermal dysplasia. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999;88:5-10
- Cronin RJ, Jr., Oesterle LJ, Ranly DM: Mandibular implants and the growing patient. *Int J Oral Maxillofac Implants* 1994;9:55-62
- Oesterle LJ, Cronin RJ, Ranly DM: Maxillary implants and the growing patient. *Int J Oral Maxillofac Implants* 1993;8:377-387
- Thilander B, Odman J, Grondahl K, et al: Osseointegrated implants in adolescents—an alternative in replacing missing teeth. *Eur J Orthodont* 1994;16:84-95
- Thilander B, Ödman J, Gröndahl K, et al: Aspects on osseointegrated implants inserted in growing jaws. A biometric and radiographic study in the young pig. *Eur J Orthodont* 1992;14:99-109
- Odman J, Grondahl K, Lekholm U, et al: The effect of osseointegrated implants on the dentoalveolar development—a clinical and radiographic study in growing pigs. *Eur J Orthodont* 1991;13:279-286
- Kramer FJ, Baethge C, Tschernitschek H: Implants in children with ectodermal dysplasia: a case report and literature review. *Clin Oral Implants Res* 2007;18:140-146
- Becktor KB, Becktor JP, Keller EE: Growth analysis of a patient with ectodermal dysplasia treated with endosseous implants: a case report. *Int J Oral Maxillofac Implants* 2001;16:864-874
- Mishra SK, Chowdhary N, Chowdhary R: Dental implants in growing children. *J Indian Soc Pedod Prev Dent* 2013;31:3-9
- Imirzalioglu P, Uckan S, Haydar SG: Surgical and prosthodontic treatment alternatives for children and adolescents with ectodermal dysplasia: a clinical report. *J Prosthet Dent* 2002;88:569-572
- Grecchi F, Pagliani L, Mancini GE, et al: Implant treatment in grafted and native bone in patients affected by ectodermal dysplasia. *J Craniofac Surg* 2010;216:1776-1780
- Wu Y, Wang XD, Wang F, et al: Restoration of oral function for adult edentulous patients with ectodermal dysplasia: a prospective preliminary clinical study. *Clin Implant Dent Relat Res* 2015 Mar 2. doi: 10.1111/cid.12296. [Epub ahead of print]
- Kearns G, Sharma A, Perrott D, et al: Placement of endosseous implants in children and adolescents with hereditary ectodermal dysplasia. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999;88:5-10
- Guckes AD, McCarthy GR, Brahim J: Use of endosseous implants in a 3-year-old child with ectodermal dysplasia: case report and 5-year follow-up. *Pediatr Dent* 1997;19:282-285
- Sfeir E, Nassif N, Moukarzel C: Use of mini dental implants in ectodermal dysplasia children: follow-up of three cases. *Eur J Paediatr Dent* 2014;15:207-212