



## Complications of Implant Rehabilitation in Ectodermal Dysplasia Patients: A Review Study

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### ABSTRACT

**Background and aim:** Ectodermal dysplasia (ED) is a hereditary disease that affects ectodermal tissues. Its oral manifestations include hypodontia or oligodontia, which cause the reduction of the height and width of the alveolar ridge. Considering numerous difficulties that these patients encounter with regard to facial appearance, talking, and chewing, their rehabilitation has a major influence on their quality of life. Implantation therapy is considered as a successful therapeutic protocol for substitution of such teeth. This article is a review of implant rehabilitation of ED patients and the related management procedures.

**Materials and methods:** In the present review article, the English articles in PubMed, ScienceDirect, MEDLINE, and Google Scholar databases from January 2000 to December 2017 have been investigated using the following keywords: anodontia, dental implants, ectodermal dysplasia, and hypodontia.

**Conclusion:** There is a multilateral approach to the treatment of ED patients according to their age, the status of oral soft and hard tissues and the remaining teeth. Implantation therapy for such patients is similar to that of non-ED patients.

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## Introduction:

Ectodermal dysplasia (ED) is a congenital disorder defined as an abnormality in at least two tissues originated from the ectodermal layer.<sup>(1,2)</sup> Over 150 ED subtypes have been identified up to the present.<sup>(3)</sup> The two main types of ED are Hidrotic ED (Clouston Syndrome) and Hypohidrotic ED (HED; Christ-Siemens Toussaint Syndrome).<sup>(4-6)</sup> HED is the most common form of ED with X-linked recessive inheritance. Its incidence is one to seven babies per 10,000 live births.<sup>(5,7)</sup> Hidrotic ED is inherited with an autosomal dominant pattern.<sup>(8)</sup>

Typical appearance characteristics of HED patients comprise sparse hair, wide and prominent forehead and chin, saddle nose, dry skin, skin pigmentation around the eyes and the mouth, vision and hearing problems, periorbital wrinkles, lip thickening, mandibular protrusion, and dished-in midface.<sup>(3,5,9,10)</sup> Moreover, HED patients suffer from sensitivity to heat and frequent high fevers due to the absence or the significant decrease of sweat glands.<sup>(5,10,11)</sup> It is worth mentioning that sweat glands are normal in Hidrotic ED patients.<sup>(5)</sup>

Oral findings include hypodontia or anodontia, delayed teeth eruption, root and crown dysmorphism, namely microdontia, anterior conical or peg-shaped teeth, posterior pyramidal and fused roots, and taurodontism, abnormal tooth germination, and dry mouth due to hypoplasia of the salivary glands.<sup>(3,9,11,12)</sup> The lack of teeth eruption occurs mostly in the lower jaw; it results in inadequate bone volume and knife-edge alveolar ridges, leading to decreased facial height and the subsequent senile appearance.<sup>(1,5,12,13)</sup> Thus, these patients have major problems with mastication and utterances. Moreover, their facial appearance can affect their physiological and psychological aspects of life. Therefore, early diagnosis and restoration of oral function can promote their self-esteem and quality of life.<sup>(2,14)</sup>

On the basis of parameters such as the developmental stage, soft tissue anatomy, and dentoalveolar status of ED patients, it is necessary to use a comprehensive multidisciplinary treatment protocol from an early age.<sup>(4,13,15)</sup> Their prosthodontic

treatment includes tissue-supported removable prostheses and implant-supported removable or fixed prostheses.<sup>(1)</sup>

The hypoplastic maxilla poses an undesirable effect on the patient's appearance, which requires a maxillary advancement procedure.<sup>(3,9)</sup> Moreover, dental implants should be placed only when the jaw relationship is corrected.<sup>(10)</sup> Therefore, in some cases, orthognathic surgery is necessary to correct the jaw relationship and skeletal deformity in order to achieve the best possible profile.<sup>(3)</sup>

Insufficient bone support and poor retention due to xerostomia lead to patient discomfort with removable dentures.<sup>(16)</sup> Furthermore, malformed crown shape and irregular distribution of the existing teeth limit crown and bridge integrations.<sup>(11,14)</sup>

Despite the inadequate bone volume, implant-supported prostheses can greatly satisfy ED patients due to the development of their masticatory efficacy, aesthetics, and improvement of their life quality.<sup>(14)</sup> Several studies have shown that implant success rate in ED patients is similar to that in healthy individuals.<sup>(1)</sup> Nevertheless, bone augmentation should be considered as a prerequisite for adequate bone volume and appropriate facial contours.<sup>(4)</sup>

This review study aimed to evaluate the complications of implant rehabilitation of ED patients and its management to achieve a successful treatment outcome.

## Materials and Methods:

The English language papers in PubMed, ScienceDirect, MEDLINE, and Google Scholar electronic databases, from January 2000 to December 2017, were surveyed using the following keywords: anodontia, dental implants, ectodermal dysplasia, and hypodontia. Several articles were initially found and screened in detail. Afterwards, thirty articles, which were more relevant to the aim of the present study, were chosen to be deeply read and taken into consideration (Table 1).

**Table 1: Summary of studies on dental implant placement in ectodermal dysplasia (ED) patients**

No.	First author	Age (year)	Gender	Extraoral findings	Intraoral findings	Radiographic findings	Type of ED	Used techniques
1	Grecchi (10)	38	F	Typical ED form of face and skin annex	Two lateral incisors, two canines, one right premolar, and one molar (all in the mandible)	NM	Hidrotic	Implant insertion (12 fixtures) + immediate loading
2	Kilic (17)	6	M	The characteristic features of ED syndrome, including hypotrichosis, prominent forehead, flat nose, thick lips, prominent chin, and sparse hair and eyelashes	An edentulous mandible and a maxilla with two first molars with severe alveolar ridge atrophy	Two unerupted second maxillary molars and advanced alveolar bone resorption	Hypohidrotic	A maxillary removable partial prosthesis and a mini-implant-retained mandibular overdenture
3	Knobloch (15)	9	M	NM	The absence of the majority of the permanent dentition	Absence of the majority of the permanent dentition	Hypohidrotic	A mandibular fixed dental prosthesis supported by dental implants and a maxillary removable dental prosthesis
4	Cezaria Triches (12)	8	F	Light-colored, thin, and dry hair, thin and sparse eyebrows and lashes, dry skin, prominent lips, senile appearance	Congenital absence of several primary and permanent teeth, microdontia, phonetic alteration, chewing difficulties, and upper labial hypertrophic frenulum	Congenital absence of several primary and permanent teeth and tooth buds, deficient root development, and large pulp chambers	NM	Maxillary denture and mandibular implant-supported fixed prosthesis
5	Bayat (18)	18	M	NM	Severe hypodontia, dry mucosa, loss of vertical dimension, underdeveloped alveolar ridges, and Class III jaw relationship	Severe maxillary and mandibular hypoplasia	Hypohidrotic	Seven implants in the maxilla and seven implants in the mandible + a screw-retained metal-ceramic fixed partial denture
6	Wu (2)	21	F	Sparse hair, enlarged forehead and chin, "saddle" nose, periocular hyperpigmentation, wrinkling, and protuberant lips	Edentulous mandible and maxilla	Severely atrophied alveolar bone in the maxilla and the mandible	NM	Use of zygomatic implants and conventional implants as bases for bone augmentation
		19	M					
7	Rajan (1)	26	F	NM	Extensive bone loss around the supporting abutment teeth	Extensive bone loss around the supporting abutment teeth; both the maxillary and mandibular ridges were severely atrophic	NM	A fixed prosthesis immediately loaded on four zygomatic implants

8	Stern (4)	31	F	Small lips, depressed nasal bridge, a concave facial profile, hypotrichosis, periocular hyperpigmentation, and fingernails that were thick, abnormally shaped, discolored, rigid, and brittle	Small maxillary and mandibular arches, oligodontia with retained deciduous teeth, mild generalized gingivitis, caries, and failing maxillary and mandibular crown and bridge restorations	Alveolar atrophy with sinus pneumatization and congenitally missing teeth	Hypohidrotic	Fully implant-supported fixed prosthesis using immediate implants and narrow-diameter implants
9	Koyuncuoglu (11)	22	M	Fine, sparse, and thin hair, scant eyelashes and eyebrows, frontal bossing, a depressed nasal bridge, protuberant lips, a prominent chin, and a resultant concave facial profile	All permanent teeth were missing, except for the maxillary central incisors and mandibular right canine, increased vertical dimension of occlusion	NM	Hypohidrotic	An implant tooth-supported telescopic partial denture at the mandible and a tooth-supported telescopic partial denture at the maxilla
10	Dhima (9)	17	M	Sparse hair, intolerance to light, hypodontia, and maxillary hypoplasia	Severe maxillary hypoplasia, hypodontia, velopharyngeal insufficiency, non-restorable caries, and generalized moderate periodontitis	The need for significant anterior and vertical advancement	NM	Mandibular implant-retained fixed prosthesis and a maxillary implant-retained detachable prosthesis
11	Priya (16)	22	F	The patient's facial skin was dry and soft, increased thickness of fingernails	Missing teeth with generalized microdontia	Presence of retained primary teeth in both arches	Hypoanhydrotic	Implant-retained fixed partial denture
12	Ghovei i (8)	19	F	Frontal prominence, mild Angle Class III jaw relationship, and prominent lips	Absence of the majority of the permanent dentition, loss of vertical dimension of occlusion	Enlarged pulp chambers in all permanent molars, narrow and short roots in all first premolars	Hypohidrotic	Eleven implants placed in the maxilla and the mandible with metal-ceramic implant-supported fixed prostheses

13	Al-Ibrahimi (3)	15	F	Dry and pale skin, no body hair on arms or legs, hyperpigmentation and linear wrinkles around the eyes, protuberant lips, and depressed nasal bridge	Underdeveloped maxillary ridge with minimal height and width, a prognathic mandible, macroglossia, oligodontia, malformation of the permanent teeth, and hypoplastic enamel	NM	Hypohidrotic	Maxillary tooth-supported fixed detachable telescopic prosthesis and implant-supported mandibular fixed partial denture
14	Mosha verinia (26)	24	M	NM	Missing of most of the permanent dentition	NM	NM	Maxillary and mandibular implants to support fixed metal-ceramic prostheses
15	Jain (27)	11	M	Sparse hair, frontal bossing, depressed nasal bridge, prominent supra-orbital ridges, sunken cheeks, hyperpigmented skin around the eyes, protuberant lips, and decreased lower facial height	Absence of saliva and dry oral mucosa, cone-shaped teeth, and underdeveloped edentulous mandibular alveolar ridge	No evidence of any impacted tooth	Hypohidrotic	Maxillary flexible removable partial denture and mandibular conventional complete denture
16	Kramer (37)	8	M	Thin, sparse, and blond hair, and sparse eyelashes and eyebrows	Severe hypodontia	NM	Hypohidrotic	Early insertion of dental implants at the age of 8 years

M=Male, F=Female, NM=Not Mentioned

## Discussion:

Replacing the lost soft and hard tissues and restoring the missing teeth are the main goals in the treatment of ED patients to achieve a proper vertical dimension and facial soft tissue appearance.<sup>(17)</sup> Despite severe bone deficiencies, implant rehabilitation can be a good solution for ED patients. Several methods have been introduced for alveolar bone augmentation, including autogenous, allogenic, and xenogeneic bone grafts, vertical distraction osteogenesis (DO), sinus floor augmentation, and the combination of the mentioned modalities.<sup>(2,17,18)</sup>

Kilic et al reported that dental implant insertions in association with guided bone regeneration would be a successful treatment in ED patients.<sup>(17)</sup> However, unpredictable resorption of the bone graft and delayed placement of implants are among the disadvantages of the above-mentioned procedure.<sup>(1)</sup> Autogenous bone grafts, harvested from extraoral (iliac, fibula or scapula) or intraoral (chin, external oblique ridge, and mandibular ramus) sources, are the gold standard for augmentation of severe alveolar bone deficiencies.<sup>(2)</sup> Several reports indicated the significant resorption of iliac grafts.<sup>(19)</sup> Fresh-Frozen Bone (FFB) allografts without donor site morbidity and hospitalization are a proper source for alveolar ridge reconstructions.<sup>(7)</sup> Implant failures often occur in the anterior zone of the maxilla due to bone graft resorption; this could be due to the presence of more fibrous bone in ED patients and the lower vascularization of severely atrophic ridges. Similarly, gingival grafting is occasionally necessary for increasing the vestibular depth and keratinized gingival width at the implant placement site.<sup>(11)</sup>

Distraction osteogenesis (DO) is another way for vertical correction without causing donor site morbidity.<sup>(9)</sup> The existence of multiple medullary bones inside the mandible allows the proper use of the DO technique.<sup>(20)</sup> Onlay grafting after vertical compensation can provide sufficient alveolar width and prepare the alveolar ridge for implant placement.<sup>(20)</sup> Successful treatment depends on the patient's cooperation with regard to activating the device four times a day; however, the device may cause scarring and skin infection around the fixation pins.<sup>(9)</sup>

Gérard Scortecchi introduced mini-distraction

osteogenesis in 2016. In this method, clot formation causes stem cell stimulation to modify bone matrix tensions before implant placement.<sup>(21)</sup> Odin et al reported the use of a flapless osteotensor in an ED patient, which was activated for 21 (for type I bone) to 45 days (for type IV bone) before implant placement.<sup>(22)</sup> The most increased bone height was observed in the sinus area. Transmission of mandibular bone from type I to active type II was observed after 15 days, and ultimately, a successful immediate implant-supported complete denture was implemented without using bone grafts.<sup>(22)</sup>

In term of the healing potential, it has been shown that there is no significant difference in osseointegration and survival rate between ED and non-ED patients. Silthampitag et al evaluated bone density via cone-beam computed tomography (CBCT).<sup>(23)</sup> They found that ED patients showed lower bone formation than the control group at the extraction site. In addition, it has been shown that female ED patients have a more compact bone with greater trabecular connectedness compared to male patients.<sup>(23)</sup>

Alveolar resorption decreases implant success rate in the maxilla more than the mandible. Guckes et al showed that implant placement in the maxilla is 2.8 times more prone to failure than in the mandible.<sup>(24)</sup> In 1998, Brånemark introduced zygomatic implant (ZI), which is a good alternative for implant rehabilitation of severely atrophic maxilla without using grafting materials.<sup>(25)</sup> In this technique, two ZIs on either side were fixed in the lateral orbital rims and in the zygomatic arches. Firstly, the implant was placed in the canine regions and then near the second premolar and first molar regions.<sup>(1,5)</sup> The survival rate of ZIs has been reported to be 96.7% during a 12-year follow-up.<sup>(2)</sup> However, some clinical problems have been reported in the literature such as hematoma and cheek biting.<sup>(2)</sup> Labial positioning and the lack of keratinized mucosa result in soft tissue recession and calculus accumulation in the exposed threads of the implants and the subsequent gingival hyperplasia. The use of soft tissue grafts and smooth surfaces are recommended to prevent recessions.<sup>(1)</sup>

Congenital orofacial defects in ED patients increase the importance of oral rehabilitation

from childhood in order to improve physical and psychological well-being.<sup>(26)</sup> The most common treatment for young ED patients is the use of removable dentures, which are not sufficiently effective due to their poor retention.<sup>(16)</sup> It is notable that denture wearing from an early age can cause progressive resorption of the basal bone and trouble for later prosthetic treatments.<sup>(11,17)</sup>

Many articles have stated that implant placement should be postponed until the cessation of the dynamic growth of the jaws;<sup>(27-30)</sup> the reason is that the implant cannot participate in the growing process due to the absence of the periodontal ligament (PDL). It may even impede the growth of the jaw bone;<sup>(12,17)</sup> therefore, it behaves like an ankylosed tooth, becoming embedded, displaced or infraocclusive.<sup>(31,32)</sup> Hence, horizontal loading may be increased due to insufficient crown/implant length ratios.<sup>(33)</sup> The implant can also affect the morphology as well as the eruption of adjacent tooth buds.<sup>(17,34)</sup> Furthermore, the remodeling pattern of the jaws can cause implant exposure during growth.<sup>(32)</sup> Conversely, there is evidence representing the success of early implant therapy in children, especially in case of severely atrophic ridges.<sup>(12)</sup> It seems that the edentulous span and the growth pattern of the jaws can affect the outcome of this treatment protocol.

The choice of treatment in children with severe hypodontia may be two intraosseous implants in the interforaminal regions of the mandible since most of the transversal growth occurs till the age of six years.<sup>(12,15,32)</sup> Growth changes can be negligible after the eruption of the permanent mandibular incisors.<sup>(31,35)</sup> Early implant placement can bring some benefits to patients. It can slow down alveolar ridge resorption and preserve the residual bone height and width. Furthermore, it can stimulate bone formation until the patient's maturity, when a definitive implant-retained prosthesis can be delivered.<sup>(15,32,36)</sup>

Moreover, the implants can supply significant stability and retention for implant-retained prostheses.<sup>(15)</sup> Guckes et al reported a 91% survival rate of early implant placement in the anterior zone of the mandible.<sup>(24)</sup> Excellent local blood supply and healing potential are some other desirable factors for implant placement at an early age.<sup>(12)</sup> It is noteworthy that the vertical

growth of the mandible requires prosthetic replacement to adjust the occlusal plane during the growth period.<sup>(24)</sup>

The remodeling pattern of the posterior mandible, the nasal floor, the antral floor, and the maxillary sutural surfaces can lead to implant exposure in the posterior mandible and maxilla, respectively. Moreover, the transversal growth of the maxilla occurs mostly at the mid-palatal suture; implant placement may restrict this growth, causing maxillary constriction.<sup>(37)</sup> Hence, implant insertion should be delayed in these sites until the completion of the growth.<sup>(15,32,34,37)</sup>

### Conclusion:

A comprehensive multidisciplinary approach should be considered for the treatment of ED patients. The reconstruction strategy depends on the patients' age, developmental stage, the anatomy of oral soft and hard tissues, and the number of missing teeth. Implant-supported restorations have a high success rate similar to the rate in non-ED patients.

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