Received: 27 March 2023 | Revised: 4 July 2023 | Accepted: 24 July 2023
DOI: 10.1111/cid.13258

ORIGINAL ARTICLE

WILEY

Rehabilitation with implant-supported overdentures in preteens patients with ectodermal dysplasia: A cohort study

Marco Montanari DDS, MS, PhD [1] | Francesco Grande DDS, MS [1] | Luca Lepidi DDS, MS [1] | Gabriela Piana DMD, DDS [2] | Santo Catapano DMD, DDS [1]

[1]Department of Prosthodontics, University of Ferrara, Ferrara, Italy

[2]Department of Biomedical and Neuromotor Science, University of Bologna, Bologna, Italy

Correspondence
Francesco Grande, DDS, MS, Via Luigi Borsari 46, 44121 Ferrara, Italy.
Email: francesco.grande@unife.it; francesco.grande90@gmail.com; s301918@studenti.polito.it

Abstract

Introduction: Hypohidrotic ectodermal dysplasia (HED) patients suffering of oligo-anodontia require early dental treatment to improve oral functions and reduce social impairment. The aim of this study was to evaluate the skeletal growth, implant and prosthetic survival rate, success, and complications after the rehabilitation with a maxillary denture and an implant-supported overdenture provided by a sliding bar in case of severe hypodontia/anodontia related to HED.

Materials and Methods: This retrospective cohort study began in 2009. Nine patients over 7 years old with HED and associated oligo-anodontia who presented at the University of Bologna for dental treatment were included in the study. They were first treated with conventional dentures and then with a maxillary denture and an implant-supported overdenture with a sliding bar connected to two implants placed in the anterior mandible. The subjects treated were followed for 3–12 years. In each case, orthopanoramic and lateral cephalometric radiographic exam were taken before implant placement and annually after prosthetic load. Vertical and transverse dimensions of the mandible in the symphysis area at implant sites were taken on the lateral cephalometric radiography at the time of implant placement and after 5 years from the prosthetic loading to assess the presence or absence of an anterior mandibular growth. Biologic and mechanical complications were also recorded at every visit.

Results: A mandibular vertical growth under the implant apex, at the implant neck, and a sagittal growth of the symphysis after 5 years from the prosthetic loading were observed and measured. Implant and prosthetic success and survival rates were 100% after 8.1 years (mean) follow-up period. No complications were reported except in one patient, where the repositioning of a retentive cap on the counter bar in the superstructure was necessary after 3 years from the prosthetic loading.

Conclusions: The present study suggests that the growth of the mandible near implant sites continues even after their positioning. Implants can be successfully placed and provide support for prosthetic rehabilitation in preteens patients with HED.

KEYWORDS
ectodermal dysplasia, hypodontia, implants, implant-supported overdenture, sliding bar

Summary Box

What is known

- Implant placement in HED patients is documented in some case reports and can be considered an available procedure.
- The implant fixed rehabilitation improves the oral function, esthetics and reduce the social impairment of young patients.
- No studies describe how the edentulous mandible of HED patients grows and if implants can compromise this growth.

What this study adds

- The oral rehabilitation of nine HED patients with a sliding bar able to connect implants and the removable prosthesis did not stop the mandibular growth.
- The present study shows relevant results of implant and prosthetic success in a period of 3-12 years of follow-up.

1 | INTRODUCTION

Ectodermal dysplasia syndrome is a rare group of inherited disorders caused by different chromosomal alterations, with more than 170 different clinical conditions, characterized by agenesis or dysplasia of tissues of ectodermal origin.[1] Clinically, ectodermal dysplasia is classified into two broad categories: the hypohidrotic form, in which sweat glands are absent or significantly decreased, and the hidrotic form, in which sweat glands are normal.[2] Hypohidrotic ectodermal dysplasia (HED) X-linked is an autosomic recessive form characterized by the triad oligo-anodontia, hypotrichosis, hyperhidrosis (Christ-Siemens-Touraine syndrome) and by characteristic dysmorphic facial features.[3] Second, the hydrotic form (Clouston's syndrome) usually spares the sweat glands but affects teeth, hair, and nails and is inherited as an autosomal trait.[4] The prevalence of HED is about 1–9/100 000 worldwide but can also be higher depending on the population considered.[5,6] Mutations in both Ectodysplasin-A and Ectodysplasin-A receptor genes cause X-linked and autosomal HED.[2] The main symptoms include heat intolerance and hyperthermia related to the absence or reduction of the sweat glands. The clinical features include sparse, fine hair, thin skin, reduced sweat and mucous glands, and missing or cone-shaped teeth.[3,6] The diagnosis of HED early in life may be difficult since sparse hair and absent teeth are normal findings at that age. Later in childhood, the diagnosis is easier, based on clinical examination with an accurate family and medical anamnesis.[6] The most frequent dental findings in these subjects are anodontia or oligodontia and tooth shape abnormalities: incisors and canines are usually cone-shaped, while second molars are affected by taurodontism.[7]

The most frequent chief complaint of patients affected with HED is missing and abnormally shaped teeth.[7] A multidisciplinary team approach, including the pediatric dentist, orthodontist, prosthodontist, and an oral-maxillofacial surgeon, is necessary for a successful outcome.[8,9] Early prosthetic oral rehabilitation of these children is recommended as an essential part of HED management to improve function, esthetics, and psychological well-being. This treatment approach presents several challenges, including anatomical abnormalities of teeth and reduced alveolar bone ridges, which can both lead to poor retention and stability of conventional dentures.

Furthermore, these patients are highly demanded for oral prosthesis to restore speech, facial appearance, and comfort, and to achieve partial function of mastication. In young patients, denture retention and stability are major factors to obtain their full cooperation in wearing the prosthesis; however, craniofacial growth shall be considered as well. Implant-supported overdentures have been used over the past 25 years in HED patients to help restore some of these complex oral functions.[10] Implant-supported overdentures provide several advantages, including increased retention and stability of the prosthesis, which lead to an improvement in function and esthetics.[11–13] Fixed restorations also allow these patients to avoid social problems that are associated with partial or full dentures, particularly in young people.[8] However, the patient collaboration is also fundamental, especially in these clinical cases, so a reduced invasiveness should be considered.[14]

Implants are usually contraindicated in pediatric patients because they do not follow the regular growth process of the craniofacial skeleton and act as ankylosed teeth.[15] Additionally, implants can interfere with the position and the eruption of adjacent teeth.[16] However, an exception to this restriction could be performed, following specific guidelines,[15] in children suffering of extended oligodontia or anodontia with the objective of improving retention and stability of removable denture. Several questions about the use of implants in this young patient population remain unanswered. Specifically, the relationship between facial growth and the placement of implant-supported overdentures in children has not been addressed. The aim of this study was to evaluate the skeletal growth, implant and prosthetic survival rate, success, and complications after the rehabilitation with a maxillary denture and an implant-supported overdenture provided of a sliding bar in case of severe hypodontia/anodontia related to HEDs with a follow-up time of 3–12 years.

2 | MATERIALS AND METHODS

For this retrospective cohort study, nine patients with no contraindications to oral surgery were addressed to rehabilitate their oral apparatus to the Dentistry Unit of Special Needs Patients, Department of Biomedical and Neuromotor Sciences, University of Bologna. The study was approved by the Ethics Committee of S. Orsola-Malpighi Hospital, Bologna, Italy (no. 84/2008/U/Disp/AOUBo Sper 16/12/2008).

The inclusion criteria were a diagnosis of HED with associated oligo-anodontia of the maxilla and the mandible, age between 9 and 12 years and a demonstrated ability to maintain adequate oral hygiene. The exclusion criteria were the presence of teeth mesial to the first molar, age before 7 years old, pathologies that contraindicated dental implant surgery like immunodeficiency, diabetes, and cardiac chronic diseases. All patients underwent clinical examination.

Initial radiographic records consisted of an orthopanoramic, a lateral cephalometric exam, and a preoperative CBCT.

2.1 | Preoperative procedures

Clinical and radiographic examination revealed severe 3-dimensional bone atrophy of the edentulous maxilla and mandible in all patients. CBCT images (SkyView, Myray) were obtained and DICOM data were imported into an open-source image processing software (Osirix Imaging Software). In this way, the 3-dimensional image of the anatomic area was reconstructed, radiographic artifacts removed, and then it was exported in an STL file format for the realization of rapid prototyping models by 3-dimensional printing (ZPrinter 310 Plus, Zcorporation).

Then, the placement of two tapered implants in the symphysis region was simulated for each 3-dimensional printed model and a second surgical template used as a pilot drill to guide implant positioning was realized.

2.2 | Surgical procedure

The surgical procedure was performed under antibiotic and antiseptic prophylaxis by administering 1 g Augmentin (amoxicillin and clavulanic acid) 1 h before the surgery and 0.2% chlorhexidine mouthwash started 3 days before.

After performing local anesthesia, a buccal-lingual full-thickness flap was raised, and the first surgical template was placed over the alveolar crest to guide mandibular ridge flattening with a surgical bur. After the osteoplasty, the second surgical template was positioned in the mandible as a pilot drill. Then, implant insertion was performed by freehand. Two tapered implants (3.8 × 10 mm^2 Keystone Dental or 3.5 × 10 mm^2 AnyOne Megagen) were placed in the corresponding positions of teeth 33 and 43 (Figure 1). The flap was sutured using a 4-0 absorbable synthetic suture material (Ethicon FS-2). A panoramic radiograph was taken after the surgery to verify the correct implant placement. Post-operative instructions about diet and oral hygiene were given to the patient and the implants were left healing to osseointegrate for a period of at least 4 months.

2.3 | Prosthetic treatment

The patients used conventional complete maxillary and mandibular dentures to allow a submerged healing period. After 4 months, the implants were exposed and low-profile attachments were used (OT Equator, Rhein83) to stabilize the pre-existing prosthesis (Figure 2). After 2 weeks, a master impression with medium-body polyvinylsiloxane impression material (Elite HD, Zhermack) was taken using a custom resin tray. Two occlusion rims were fabricated and

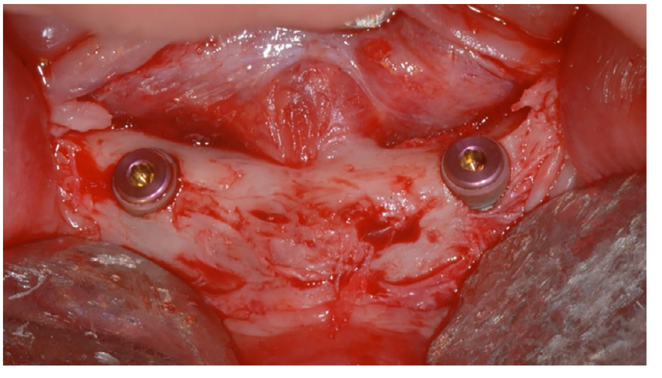

FIGURE 1 Implants placed in anterior mandible in position 33 43.

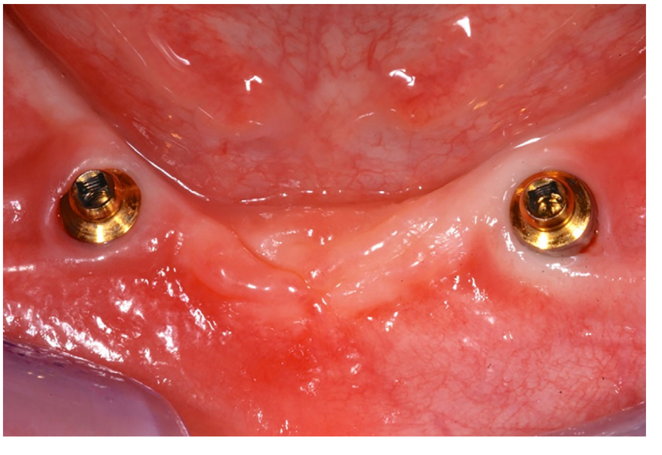

FIGURE 2 OT Equator attachment placed on the two implants in order to stabilize the pre-existing prosthesis and then anchor the bar of the final prosthesis.

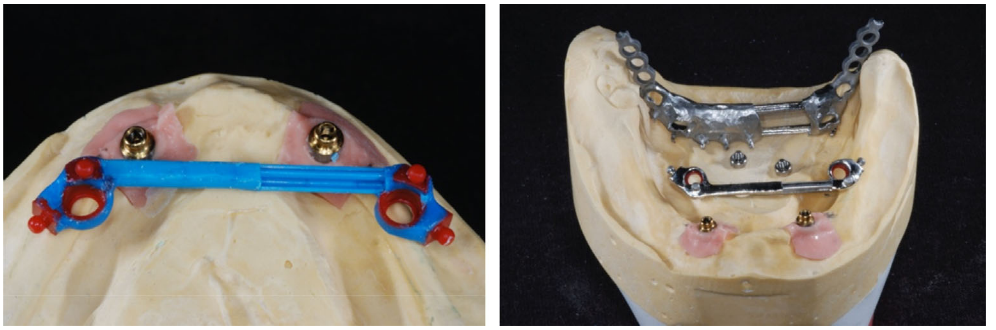

FIGURE 3 A,B. A prosthetic bar was made with the characteristic to be divided into two halves able to slide and follow the mandible growth. Each bar showed four ball attachments on its surface to increase the prosthesis retention. (A). Wax-up of the sliding bar. (C) Disassembled components of the implant-supported denture.

FIGURE 4 The bar was connected to OT Equator, which acted as a multi-unit abutment able to anchor the prosthesis bar and no more as direct implant attachments. The Seeger ring ensures stability and passivity of the abutment-bar complex.

used to register maxillo-mandibular relationship together with a small Fox's "plan" according to the patient's age and following conventional phonetic and esthetic criteria. A mandibular try-in denture with anterior and posterior teeth simulating tooth eruption was then clinically tested for function and esthetic using two different types of teeth for deciduous (Bambino tooth, Major Dental) and permanent teeth (Acrismart, Ruthinium). A prosthetic bar with a rounded section of 1.95 mm diameter, divided into two halves capable of sliding on each other without friction was made. The sliding bar was delivered with the two halves closed and no space between them; if a mandibular transversal growth occurs, the two halves of the bar would slide, and a space at the midline would be shown. Each bar had four small ball-shaped attachments on its surface to increase the prosthesis retention (Figure 3). Then, this sliding bar was connected to OT Equator attachments both with screws and with two elastic Seeger rings (Elastic Seeger Rhein83), that provide the system of a snap-on retention in addition to the screw and a passivation of the framework (Figure 4).[9] Thanks to this peculiar connection, the bar was also allowed to rotate around its vertical axis on the OT Equator attachments. During the lab procedures, the implant-supported overdenture was sectioned into two parts connected by a sliding transversal plane that allowed bar and prosthesis expansion (Figure 5).

2.4 | Measurements of mandible dimensions

Vertical and transverse dimensions of the mandible symphysis area were measured on the lateral cephalometric radiography before the rehabilitation and during the follow-up after 5 years from the implant placement (Figure 6). Two different dentists (FG and SC), who were not directly involved in the surgical or prosthetic procedures, performed all the measurements directly on the analogic supports through the use of a millimeter scale. They did not know the patient and when the radiograph analyzed was taken. The mean of the two different measurements was taken as the real value).

The following measurements were taken into consideration immediately after prosthetic loading and after 5 years to assess vertical and horizontal bone growth in the anterior mandible (Table 1):

- Implant apex-Menton: distance between the apex of the implant and the lower edge of the symphysis to evaluate the amount of bone apposition in vertical dimension (red line in fig. 8);
- Implant neck-bone crest: distance between the implant neck and the bone crest at the midline to evaluate the vertical growth of the alveolar crest after the surgical bone flattening (blue line in fig. 8);
- Thickness of the symphysis: this measure was taken by drawing a line from the Pogonion to the lingual corticalized point of the symphysis perpendicularly to the implant axis and passing at the apex of the implant (black line in fig. 8). It could also be indicative of the mandibular sagittal growth at the symphysis area.

To measure the transversal growth of the mandible at the symphysis, we directly took the implants as a reference; if the distance between them changed, the sliding bar would slice.

2.5 | Follow-up

After prosthetic rehabilitation on implants, the patients could have a regular diet without restrictions on food consistency, which was difficult while wearing conventional dentures.

The patients were followed with regular visits every month for the first year and every 6 months after 1 year from the prosthetic

FIGURE 5 Sectioning of the lower overdenture in two parts connected by a sliding transversal plane that allowed bar and prosthesis expansion. (A) Lower prosthesis before sectioning. (B) Lower prosthesis sectioned: lower view. (C) Lower prosthesis sectioned: frontal view.

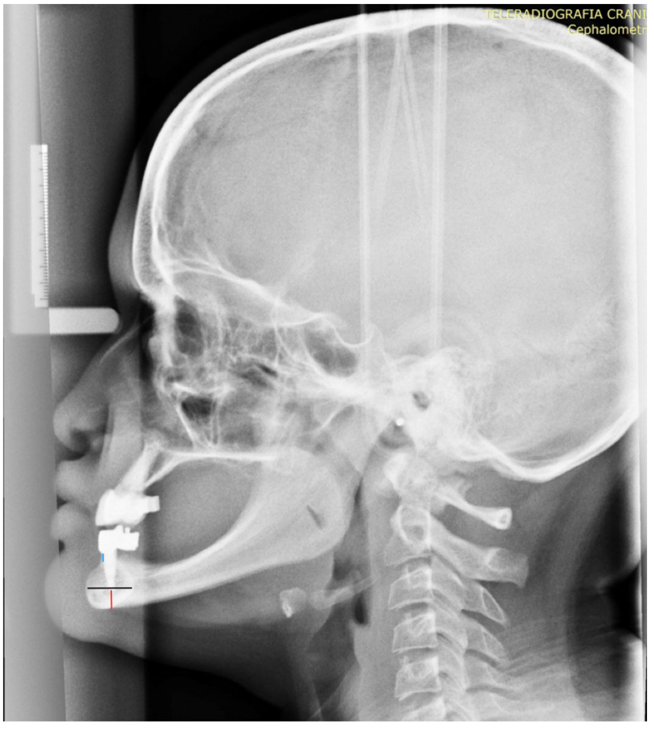

FIGURE 6 Measurements of the mandible in the symphysis area on the latero-lateral cephalometric radiography taken before the rehabilitation and after 5 years of follow-up. The red line indicates the measure implant apex-lower mandibular edge, the black line indicates the thickness of the symphysis, and the blue line indicates the distance between the implant neck and bone crest.

load. The resilient caps of the OT Equator attachments, positioned in the overdenture counter bar, were changed approximately once a year for every patient, when a dimensional change was observed because of wear. A panoramic and a cephalometric radiograph were taken annually after prosthetic load with the same machine (Hyperion X5, Myray Cefla) with a 1:1 standard ratio. Implant survival was considered as the absence of failure and the maintenance of implant in situ. Implant success was considered as the implant remains in function without pain, rotation, bleeding/pus, soft tissue inflammation, pocket probing depth of more than 5 mm in combination with a bleeding index of 3 and absence of radiographic peri-implant bone resorption higher than 2 mm after

TABLE 1 Measurements taken immediately and after 5 years from prosthetic loading regarding the distances from implant apex to the lower mandibular edge, from the implant neck to the bone crest and the sagittal dimension (thickness) of the symphysis.

	Implant apex-Menton (mm)		Implant neck-bone crest (mm)		Thickness of the symphysis (mm)	
	Initial	*After 5 years*	*Initial*	*After 5 years*	*Initial*	*After 5 years*
1	6	8	0	2.5	12	14
2	6	8	0	2.5	13	15.5
3	5	-	0	-	12	-
4	7	9	0	2.5	14	15
5	5	6.5	0	4	12.5	14.5
6	7	10	0	1	12	15
7	5	9	0	2	11	14
Range (mm)	5-7	6.5-10	0	1-4	11-14	14-15.5
Median	6	9	0	2	12	14.5
Variance	0.81	0.7	0	2.33	1.07	0.33

2 years.[17] Prosthetic survival was defined as the maintenance of the prosthesis function. Prosthetic success was considered when it remained in situ providing an adequate function, and esthetics with the support of all the implants and no technical prosthetic complications occurred during a 5-year period.[18] Biologic and mechanical complications regarding implants and prosthesis were recorded at every visit.

3 | RESULTS

Six months after implants placement, one patient (KD) left the study and the final rehabilitation was not delivered. The patient was then excluded from the descriptive analysis of the data.

The remaining eight patients (mean age: 10.4 years old at the time of the implant surgery) were followed with a mean follow-up period of 8.1 years (Table 2).

From the analysis of the cephalometric radiographs initially and after 5 years from the prosthetic loading, the mandible dimensions near the site of implant placement changed as follows:

- The distance between the apex of the implant and the lower mandibular edge, namely Menton, ranged from 5 to 7 mm (median 6, variance 0.81) initially and from 6.5 to 10 mm (median 9, variance 0.7) after 5 years of follow-up;
- Implant neck-bone crest measured 0 mm initially and ranged from 1 to 4 mm (median 2, variance 2.33) after 5 years;
- Sagittal symphysis thickness ranged from 11 to 14 mm (median 12, variance 1.07) initially and from 14 to 15.5 mm (median 14.5, variance 0.33) after 5 years.

Implant and prosthesis success and survival rates during the follow-up period were 100%. No biological complications were recorded during the period of follow-up. One minor mechanical complication occurred in one patient (MA) at 3 years from the prosthetic load; it foresaw the repositioning of one resilient cap on the counter bar in the overdenture superstructure.

4 | DISCUSSION

Treatment of the nine preteens HED patients was performed using a multidisciplinary approach involving pediatric dentistry, prosthodontics, and oral surgery taking into consideration several factors such as age, type of prosthesis needed, cranio-facial growth pattern, tooth development, and eruption.

In the present study, the implant and prosthesis survival and success rate were 100% after a mean period of 8.1 years of follow-up. Only in one case, a minor prosthetic complication occurred: the overdenture could not be reinserted on the bar after a period of non-wearing. This was probably due to a different mandibular vertical growth at the two implants because they were positioned at different distances from the mandible midline. The complication was simply solved by changing the position of one resilient cap on the counter bar in the overdenture.

Prosthetic rehabilitation of HED patients was performed as soon as possible, to improve masicatory function, maxillo-facial growth, esthetics, and speech-reducing social impairment. Our protocol consists of early rehabilitation with complete removable dentures when the patient is about 2.5–3 years old and implant placement, when possible, after 4–5 years.

In literature, implant treatment in young patients has been accepted but with conflicting opinions regarding the timing. To avoid implant displacement caused by craniofacial growth, some authors suggest implant placement after the age of 13–14 years,[9] while others encourage it even before puberty, for optimum functional and psychological development.[16,19,20] Furthermore, implant placement is more indicated in fully edentulous growing patients compared to partially edentulous because the presence of neighboring teeth could create infraocclusion problem.[21–23]

TABLE 2 Table describing the type of patients treated, complications recorded and amount of anterior mandibular expansion.

Patient	Age at time of surgery (years)	Years from implant placement (years)	Type of alveolar ridge (according to Cawood & Howell)	Need to adjust sliding bar	Biological/mechanical complications	Amount of anterior mandibular expansion
1	12	12	Class IV	No need	No complications	No expansion
2	9	12	Class IV	No need	No complications	No expansion
3	11	11	Class IV	No need	No complications	No expansion
4	10	11	Class IV	No need	Repositioning of a resilient cap into the counter bar (2 years)	No expansion
5	12	9	Class V	No need	No complications	No expansion
6	10	6	Class IV	No need	No complications	No expansion
7	11	5	Class IV	No need	No complications	No expansion
8	9	4	Class IV	No need	No complications	No expansion
9	10	3	Class IV	No need	No complications	No expansion
Mean	10.4	8.1				

Differently from our study, the literature shows a high implant failure rate in young EDs children that in Bergendal et al. reaches 65%.[24] They reported that it could be probably related to the small jaw associated with the high bone density which, in turn, is much more exposed to overheating during implant path preparation.[8,24] Lesot et al.[25] found the increased bone density of the mandible in individuals with HED and concluded that this skeletal phenotype is associated with the EDA mutation and confirms the involvement of the EDA-NF-kB signaling pathway in bone metabolism.[25,26] The small crestal thickness may be responsible for the implant threads exposure increasing the failure probability.

In our study, the patients' age was higher compared to Bergendal[24] and even if some implant threads were exposed (Figure 1), they were covered by the adjacent soft tissues. The crestal bone was flattened before implant surgery providing a sufficient width at the implant neck. In addition, a partial thickness flap was raised favoring the maintenance of the soft tissue volumes in the healing phase. Moreover, before the implant surgery, an evaluation of the alveolar crests and a surgery simulation on 3-dimensional printed models were performed.

Another possible explanation for the absence of implant or prosthetic complications/failures may be due to the connection system used. The Seeger rings positioned between the bar walls and the subequatorial portion of the OT Equators allows to center the bar on the attachments before screwing, passivating the framework and compensating at the same time the small misfit originating from the prosthetic fabrication processes.[27] Moreover, the Seeger rings reduce the stress on the screws during the masticatory cycles, reducing the possibility of screw loosening/fracture.[28–30] In our study, we used a low-profile attachment (OT Equator) that has a smaller volume than a conventional MUA and is coated with titanium nitride for an improved biocompatibility with soft tissues.[31,32]

Another factor that is worth of consideration in terms of implant prosthetic success was the sliding bar, able to anchor the overdenture and to follow the mandibular transverse growth or bone deformation during chewing or mouth opening and closure.[33–35] Our decision to proceed with a sliding bar was based on the fact that the mandible was extremely thin in these patients and possibly subjected to deformation during functioning.[34] In addition, this bar, by sliding instead of rigidly blocking the implants, avoids any horizontal interferences with the mandible growth. Furthermore, the possibility of each half of the bar to have a rotation on the OT Equator attachment gives more degree of freedom to the prosthesis itself that could be important in young patients. However, since the fact that implant positioning could affect the mandibular growth, we decided to monitor it in the area near the implants. Radiographic examinations showed that implants followed the mandible growth while keeping their original relationship with the jawbone. At the same time, the mandible growth did not stop: after 5 years of follow-up, a vertical and sagittal growth in the symphysis region at implant sites was observed and measured. The bone growth was also present under the sliding bar between the implants as demonstrated by the increased distance between the implant neck and the bone crest seen in our sample. This could be probably due to an improvement in the masticatory function and to a reduced occlusal loading that is exerted with a mobile denture (Wolff's Law).[36]

Maxillary and mandibular growth follow a downward and forward direction vector in the sagittal plane, in addition to bone remodeling.[37] During childhood, the jaws are changing structures: in absence of teeth, such as in HED, an anti-clockwise rotation of the mandible results in a Class III skeletal relation.[38]

Implants in the maxillary arch are contraindicated for the risk of implant dislocations due to vertical maxillary growth and bone resorption in the nasal floor. In addition, maxillary transversal growth occurs mainly in the midpalatal suture: so, a cross-arch fixed restoration could affect the bone growth.[39]

However, in the anterior mandible, alveolar remodeling has a smaller influence on implant rehabilitation. In the follow-up period, the sliding bars did not show any expansion, highlighting the lack of growth of the mandibular symphysis in a horizontal direction in the frontal plane. This is in accordance with the theory that the transversal growth of the anterior mandible occurs mainly in early childhood (6 months to 2 years) by apposition and resorption of bone in the buccal and lingual surfaces.[37,38] After that age, mandibular growth occurs posteriorly at condyle and ramus, possibly causing a rotation of the mandible and changes in occlusal plane angulations.[37,38] In the present report, this was not observed, but we recognize the importance of putting particular attention to a vertical dimension increase over time due to patient growth.

Studies reported 1–2 mm bone apposition and resorption, which are encouraging results in terms of implants survival in growing patients.[40] Some reports showed that normal craniofacial morphology did not differ significantly between implant-treated and non-implant-treated patients with HED, suggesting that implant treatment did not necessarily affect normal craniofacial growth.[41,42] However, in those studies, an evaluation of the entire craniofacial growth was performed, but the authors did not focus on the areas near implant placement. In addition, in the study of Johnson,[41] the patient sample was predominantly constituted of patients treated in the late adolescence, when growth was almost concluded. These reasons made a comparison between our studies quite difficult.

The absence of complications and of infraocclusion problems, the constant relationship between implants and alveolar crest and the continuous mandibular growth near implant sites confirm the goodness of this type of treatment.

The limits of this study are the limited sample, the retrospectivity of the study, the relative short follow-up considering the patients' age, the absence of 3-dimensional measurements of the patient mandible that are more reliable than 2D and the lack of an OHIP-14 score. In addition, our data cannot be compared with a conventional mandibular growth because of the absence of a control group but demonstrated only that the mandible continued to grow even after implants positioning. We are not also able to assess if our sliding bar that avoids horizontal but not 3-dimensional interferences may have disturbed the growth anyway. Moreover, the absence also of an additional control group with a conventional framework bar that rigidly fixed the implants cannot allow us to sustain the superiority or inferiority of this protocol. So, the impact of this prosthetic rehabilitation on the mandibular and craniofacial growth remains unanswered and the most appropriate type of prosthetic rehabilitation to treat preteens HED patients with predictability has yet to be understood. However, the high implant and prosthetic success rate observed in our sample suggests considering this type of rehabilitation as promising.

5 | CONCLUSION

Implant oral rehabilitation in preteens represents a challenge because of possible interferences with the patient's growth. The surgical/prosthetic protocol applied in this study had 100% implant and prosthetic success. The sliding bar, although no expansion was observed, guaranteed to avoid any interferences due to mandibular deformation during functioning and to transversal growth and may be responsible for the success rate documented in the present study.

AUTHOR CONTRIBUTIONS

All authors contributed substantially to the interpretation of the data for the work, they contributed to drafting and critically revising the manuscript, they gave their final approval of the version to be published, and they agreed to be accountable for all aspects of the work. Additionally, Marco Montanari and Gabriela Piana conceived the ideas for the concept and design of the study; Marco Montanari collected the clinical and radiographic records; Francesco Grande and Santo Catapano analyzed and performed the measurements on the lateral radiographic exam; Francesco Grande led the writing; and Luca Lepidi critically reviewed and edited the manuscript.

CONFLICT OF INTEREST STATEMENT

Marco Montanari is a speaker of Rhein83; nevertheless, this study was self-supported. Data belonged to the authors and by no means did the company interfere with the conduct of the trial or the publication of its results. This work was not previously presented and received no financial support.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

REFERENCES

1. Burn J. Ectodermal dysplasias: a clinical and genetic study. *J Med Genet*. 1985;22(6):503-504.
2. Itin PH, Fistarol SK. Ectodermal dysplasias. *Am J Med Genet C Semin Med Genet*. 2004;131C(1):45-51. doi:10.1002/ajmg.c.30033
3. Wright JT, Grange DK, Fete M. Hypohidrotic ectodermal dysplasia. In: Adam MP, Mirzaa GM, Pagon RA, et al., eds. *GeneReviews®*. University of Washington; 1993. Accessed May 3, 2023. http://www.ncbi.nlm.nih.gov/books/NBK1112/
4. More CB, Bhavsar K, Joshi J, Varma SN, Tailor M. Hereditary ectodermal dysplasia: a retrospective study. *J Nat Sci Biol Med*. 2013;4(2):445-450. doi:10.4103/0976-9668.117012

5. RESERVED IUAR. Orphanet: Hypohidrotic ectodermal dysplasia. Accessed May 3, 2023 https://www.orpha.net/consor/cgi-bin/OC_Exp.php?lng=EN&Expert=238468
6. Nguyen-Nielsen M, Skovbo S, Svaneby D, Pedersen L, Fryzek J. The prevalence of X-linked hypohidrotic ectodermal dysplasia (XLHED) in Denmark, 1995-2010. *Eur J Med Genet*. 2013;56(5):236-242. doi:10.1016/j.ejmg.2013.01.012
7. Yap AKW, Klineberg I. Dental implants in patients with ectodermal dysplasia and tooth agenesis: a critical review of the literature. *Int J Prosthodont*. 2009;22(3):268-276.
8. Stanford CM, Guckes A, Fete M, Srun S, Richter MK. Perceptions of outcomes of implant therapy in patients with ectodermal dysplasia syndromes. *Int J Prosthodont*. 2008;21(3):195-200.
9. Pigno MA, Blackman RB, Cronin RJ, Cavazos E. Prosthodontic management of ectodermal dysplasia: a review of the literature. *J Prosthet Dent*. 1996;76(5):541-545. doi:10.1016/s0022-3913(96)90015-3
10. Bergendal T, Eckerdal O, Hallonsten AL, Koch G, Kurol J, Kvint S. Osseointegrated implants in the oral habilitation of a boy with ectodermal dysplasia: a case report. *Int Dent J*. 1991;41(3):149-156.
11. Bhattacharjee B, Saneja R, Bhatnagar A, et al. A comparative evaluation of neurophysiological activity, active tactile sensibility and stereognostic ability of complete denture prosthesis, and implant-supported prosthesis wearer—a pilot study. *Clin Implant Dent Rel Res*. 2022;24(4):510-521. doi:10.1111/cid.13094
12. Sermsiripoca K, Pisarnturakit PP, Mattheos N, Pimkhaokham A, Subbalekha K. Comparing pre- and post-treatment patients' perceptions on dental implant therapy. *Clin Implant Dent Relat Res*. 2021;23(5):769-778. doi:10.1111/cid.13036
13. Chatrattanarak W, Aunmeungtong W, Khongkhunthian P. Comparative clinical study of conventional dental implant and mini dental implant-retained mandibular overdenture: a 5- to 8-year prospective clinical outcomes in a previous randomized clinical trial. *Clin Implant Dent Rel Res*. 2022;24(4):475-487. doi:10.1111/cid.13098
14. Menini M, Pesce P, Delucchi F, et al. One-stage versus two-stage technique using two splinted extra-short implants: a multicentric split-mouth study with a one-year follow-up. *Clin Implant Dent Relat Res*. 2022;14:602-610. doi:10.1111/cid.13113
15. Agarwal N, Kumar D, Anand A, Bahetwar SK. Dental implants in children: a multidisciplinary perspective for long-term success. *Natl J Maxillofac Surg*. 2016;7(2):122-126. doi:10.4103/0975-5950.201362
16. 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(4):282-285.
17. Misch CE, Perel ML, Wang HL, et al. Implant success, survival, and failure: the International Congress of Oral Implantologists (ICOI) Pisa consensus conference. *Implant Dent*. 2008;17(1):5-15. doi:10.1097/ID.0b013e3181676059
18. Papaspyridakos P, Chen CJ, Singh M, Weber HP, Gallucci GO. Success criteria in implant dentistry: a systematic review. *J Dent Res*. 2012;91(3):242-248. doi:10.1177/0022034511431252
19. Kramer FJ, Baethge C, Tschernitschek H. Implants in children with ectodermal dysplasia: a case report and literature review. *Clin Oral Implants Res*. 2007;18(1):140-146. doi:10.1111/j.1600-0501.2006.01180.x
20. Guckes AD, Scurria MS, King TS, McCarthy GR, Brahim JS. Prospective clinical trial of dental implants in persons with ectodermal dysplasia. *J Prosthet Dent*. 2002;88(1):21-25.
21. Kearns G, Sharma A, Perrott D, Schmidt B, Kaban L, Vargervik K. Placement of endosseous implants in children and adolescents with hereditary ectodermal dysplasia. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 1999;88(1):5-10. doi:10.1016/s1079-2104(99)70185-x
22. Rossi E, Andreasen JO. Maxillary bone growth and implant positioning in a young patient: a case report. *Int J Periodontics Restorative Dent*. 2003;23(2):113-119.
23. Dean D. Facial growth, 3rd ed. By D. H. Enlow. Philadelphia: W. B. Saunders. 1990. 576 pp. $79.00 (cloth). *Am J Phys Anthropol*. 1991;86(1):90-92. doi:10.1002/ajpa.1330860111
24. Bergendal B, Ekman A, Nilsson P. Implant failure in young children with ectodermal dysplasia: a retrospective evaluation of use and outcome of dental implant treatment in children in Sweden. *Int J Oral Maxillofac Implants*. 2008;23(3):520-524.
25. Lesot H, Clauss F, Manière MC, Schmittbuhl M. Consequences of X-linked hypohidrotic ectodermal dysplasia for the human jaw bone. *Front Oral Biol*. 2009;13:93-99. doi:10.1159/000242398
26. Silthampitag P, Klineberg I, Jones AS, et al. Ultramicroscopy of bone at oral implant sites: a comparison of ED and control patients. Part 1—defining the protocol. *Int J Prosthodont*. 2011;24(2):147-154.
27. Montanari M, Tallarico M, Vaccaro G, et al. Two years after loading performance of implant-supported overdenture with metal bar and low-profile attachments: a prospective case series multi-center clinical study. *Int J Dent*. 2020;2020:1-8. doi:10.1155/2020/8830722
28. Pozzan MC, Grande F, Mochi Zamperoli E, Tesini F, Carossa M, Catapano S. Assessment of preload loss after cyclic loading in the OT bridge system in an "all-on-four" rehabilitation model in the absence of one and two prosthesis screws. *Materials*. 2022;15(4):1582. doi:10.3390/ma15041582
29. Grande F, Pozzan MC, Marconato R, Mollica F, Catapano S. Evaluation of load distribution in a mandibular model with four implants depending on the number of prosthetic screws used for OT-bridge system: a finite element analysis (FEA). *Materials*. 2022;15(22):7963. doi:10.3390/ma15227963
30. Grande F, Cesare PM, Zamperoli EM, Gianoli CM, Mollica F, Catapano S. Evaluation of tension and deformation in a mandibular Toronto bridge anchored on three fixtures using different framework materials, abutment systems, and loading conditions: a FEM analysis. *Eur J Dent*. 2023. Epub ahead of print. doi:10.1055/s-0042-1758785
31. Pera F, Menini M, Alovisi M, et al. Can abutment with novel superlattice CrN/NbN coatings influence peri-implant tissue health and implant survival rate compared to machined abutment? 6-Month results from a multi-center split-mouth randomized control trial. *Materials*. 2022;16(1):246. doi:10.3390/ma16010246
32. Scarano A, Piattelli M, Vrespa G, Caputi S, Piattelli A. Bacterial adhesion on titanium nitride-coated and uncoated implants: an in vivo human study. *J Oral Implantol*. 2003;29(2):80-85. doi:10.1563/1548-1336(2003)029<0080:BAOTNA>2.3.CO;2
33. Azpiazu-Flores FX, Lee DJ, Mata-Mata SJ, Zheng F. Rehabilitation of a patient with mandibular flexure using contemporary glass-infiltrated high performance CAD-CAM polymers: a clinical report with 1-year follow-up. *J Prosthet Dent*. 2023. Epub ahead of print. doi:10.1016/j.prosdent.2022.12.014
34. Mijiritsky E, Shacham M, Meilik Y, Dekel-Steinkeller M. Clinical influence of mandibular flexure on oral rehabilitation: narrative review. *Int J Environ Res Public Health*. 2022;19(24):16748. doi:10.3390/ijerph192416748
35. Zarone F, Apicella A, Nicolais L, Aversa R, Sorrentino R. Mandibular flexure and stress build-up in mandibular full-arch fixed prostheses supported by osseointegrated implants. *Clin Oral Implants Res*. 2003;14(1):103-114. doi:10.1034/j.1600-0501.2003.140114.x
36. Reddy MS, Geurs NC, Wang IC, et al. Mandibular growth following implant restoration: does Wolff's law apply to residual ridge resorption? *Int J Periodontics Restorative Dent*. 2002;22(4):315-321.
37. Mizoguchi I, Toriya N, Nakao Y. Growth of the mandible and biological characteristics of the mandibular condylar cartilage. *Jpn Dent Sci Rev*. 2013;49(4):139-150. doi:10.1016/j.jdsr.2013.07.004
38. Krarup S, Darvann TA, Larsen P, Marsh JL, Kreiborg S. Three-dimensional analysis of mandibular growth and tooth eruption. *J Anat*. 2005;207(5):669-682. doi:10.1111/j.1469-7580.2005.00479.x

39. Fudalej P, Kokich VG, Leroux B. Determining the cessation of vertical growth of the craniofacial structures to facilitate placement of single-tooth implants. *Am J Orthod Dentofacial Orthop*. 2007;131(4 Suppl): S59-S67. doi:10.1016/j.ajodo.2006.07.022
40. Cronin RJ, Oesterle LJ, Ranly DM. Mandibular implants and the growing patient. *Int J Oral Maxillofac Implants*. 1994;9(1):55-62.
41. Johnson EL, Roberts MW, Guckes AD, Bailey LJ, Phillips CL, Wright JT. Analysis of craniofacial development in children with hypohidrotic ectodermal dysplasia. *Am J Med Genet*. 2002;112(4): 327-334. doi:10.1002/ajmg.10654
42. Alcan T, Basa S, Kargül B. Growth analysis of a patient with ectodermal dysplasia treated with endosseous implants: 6-year follow-up. *J Oral Rehabil*. 2006;33(3):175-182. doi:10.1111/j.1365-2842.2005.01566.x

How to cite this article: Montanari M, Grande F, Lepidi L, Piana G, Catapano S. Rehabilitation with implant-supported overdentures in preteens patients with ectodermal dysplasia: A cohort study. *Clin Implant Dent Relat Res*. 2023;1-10. doi:10.1111/cid.13258