

# Temporary rehabilitation with mini-implants in a child with ectodermal dysplasia

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• **Conflicts of interest:** none declared.

## ABSTRACT

**Objective:** to present the versatility of mini-implants for temporary rehabilitation of patients with hypohidrotic ectodermal dysplasia (HED). **Case Report:** this case describes the rehabilitation started on April/2015 in an eight-year-old boy with HED and family history of the disease. In view of the large number of cases reporting the difficult adaptation of removable dentures in children, the oral rehabilitation of this patient was planned based on the fabrication of dentures supported on orthodontic mini-implants. **Conclusion:** this alternative approach facilitates the use and adaptation of dentures, promotes preservation of the alveolar bone and improves the functional, aesthetic and psychosocial conditions of the patient. There should be annual radiographic and quarterly clinical monitoring until the end of growth spurt, for re-evaluation and renovation of the dentures whenever required according to the alveolar ridge.

**Keywords:** Child; Ectodermal dysplasia; Dental prosthesis; Rehabilitation.

## Introduction

**H**ypohidrotic ectodermal dysplasia (HED) is part of a group of hereditary diseases that affect ectodermal structures. It is a rare congenital disease, occurring in 1 in every 100,000 births, which is linked to the X chromosome and affects predominantly males.<sup>1-3</sup>

Patients with HED usually present specific characteristics of the syndrome, such as partial or total absence of sweat glands, hypotonicity of the perioral muscles, scant scalp hypodontia and, quite often, vertical loss of occlusion.<sup>2</sup> Other signs and symptoms include dry eyes, xerostomia, dry skin and decreased number of mucous glands.<sup>2-5</sup>

Removable dentures are often the treatment of choice for oral rehabilitation of patients with HED.<sup>2,5,6</sup> However, alternatives such as the use of temporary prostheses supported on mini-implants have been proposed in order to further improve the denture stability, preservation of the alveolar bone, as well as patient comfort and compliance to the treatment.<sup>1,7</sup>

This case report describes the oral rehabilitation of a patient with HED using a mini-implant-supported maxillary denture and a mandibular denture connected to orthodontic bands cemented to molars.

## Case Report

An eight-year-old boy was brought to the Orthodontics Clinic of the School of Dentistry of UFMG by his mother. During the interview, the mother reported that the patient had difficulties in feeding, speech and interpersonal relationships. On physical examination, the patient presented prominent forehead and lips, hypotonicity of perioral muscles, clear and sparse eyelashes and eyebrows, reddish eyes,

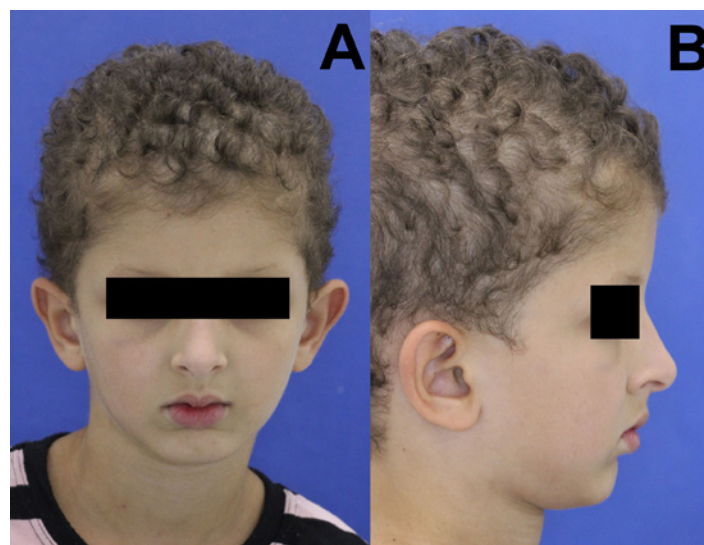
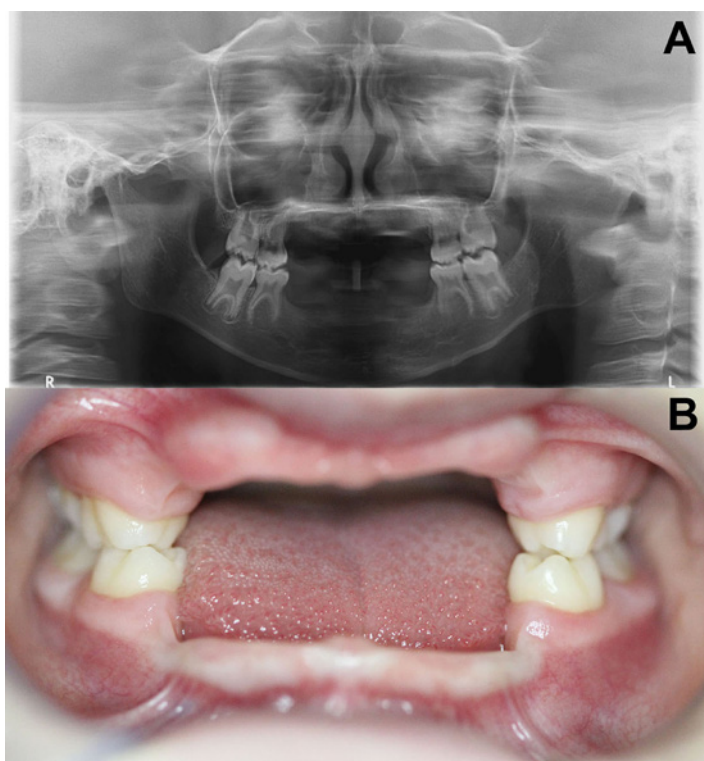


Figure 1. Frontal (A) and lateral (B) facial features commonly observed in patients with HED

pale and dry skin and generalized hypotrichosis and hypohidrosis (Figure 1A and B).

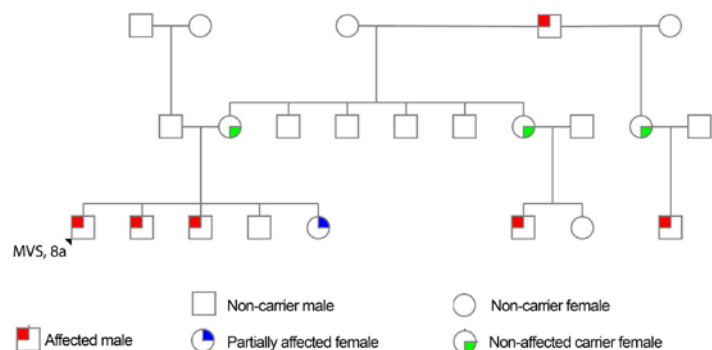
The intraoral examination revealed regular salivation and atypical swallowing with anterior tongue thrust. The patient presented only the permanent first molar (PFM) and the deciduous second molar (DSM) in each hemi-arch. The teeth were sound and in good conditions of hygiene and there was no loss of vertical dimension in the lower third of the face. According to the mother, those teeth were the only ones that the patient had ever presented, which certainly contributed to the bone loss observed mainly in the mandibular arch and underdeveloped alveolar bone in the edentulous areas. The radiographic examination showed absence of tooth germs of the missing teeth (Figure 2A and B).



**Figure 2.** Panoramic radiograph without evidence of embedded teeth or tooth germs (A); intraoral photograph showing absence of multiple teeth (B)

The patient had no medical and dental history, thus he had not been diagnosed previously. The family history reported by the mother indicated a hereditary factor of maternal origin, in which the affected males exhibited complete clinical characteristics of the disease and the females presented absence or only minimal clinical manifestations or minor characteristics, such as conical teeth and hypohidrosis, as shown in the heredogram (Figure 3). The patient was referred to the Pathology Clinic of the School of Dentistry of UFMG and a diagnosis of HED was established.

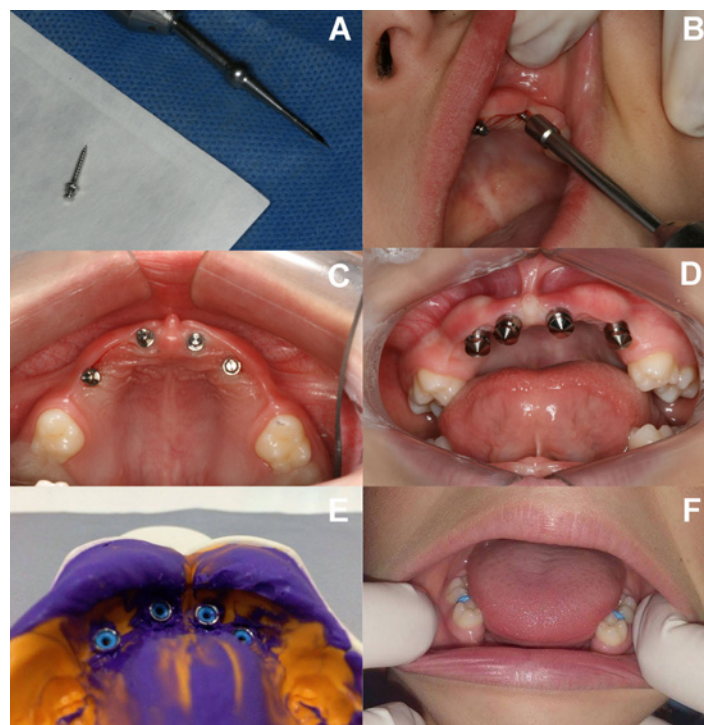
**PEDIGREE CHART**



**Figure 3.** Heredogram indicating the maternal origin of the syndrome

The treatment of choice was the installation of a dental prosthesis supported by mini-implants in the maxillary arch. In the mandible, the dental prosthesis was connected to orthodontic bands cemented on the PFMs. Four 8-mm

mini-implants (Morelli, Sorocaba, SP, Brazil) were placed on the maxillary alveolar ridge with the four corresponding o’rings (Emfils, Itu, SP, Brazil) connected to them (Figure 4A-D). Then, a single-step addition silicone (Zhermack, Badia Polesine, RO, Italy) impression with obtained and the o’rings were corrected positioned (Figure 4E). The mandibular work model, the maxillary impression housing the o’rings and four Morelli mini-implant as analogues were sent to the laboratory.



**Figure 4.** Placement of mini-implants (A, B and C) and o’rings (D) for adaptation of the denture in the maxilla; impression with addition silicone in maxillary arch and correct positioning of the o’rings in the impression (E); Placement of orthodontic separators for banding the mandibular permanent first molars (F)

After fabrication, the maxillary denture was sectioned through its midline to prevent any possible restriction of transversal growth in the area, considering the long time that the temporary denture would be in use. The denture was evaluated for stability, occlusal adjustment and internal wear to eliminate areas of tissue ischemia.

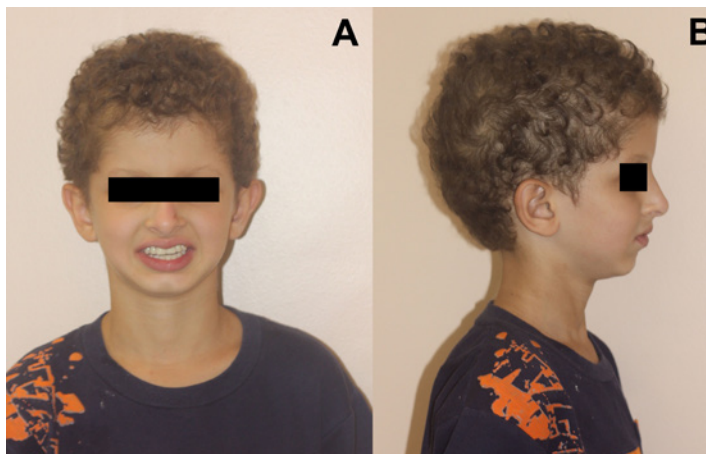
Next, orthodontic separators were placed between the mandibular PFM and DSM on the left and right sides (Figure 4F). In view of the lower stability and retention of temporary compared with definitive fixed dentures, a decision was made to maintain the dentures in infraocclusion to avoid the application of heavy occlusal load (Figure 5). Installation of the dentures was completed in three weekly appointments.





**Figure 5.** Temporary fixed dentures kept in infraocclusion to avoid the application of heavy occlusal load

The patient and the mother received full instructions on general oral care and were advised about the need for a long-term follow up until the end of the growth spurt, which included quarterly visits for evaluation of the dentures. In addition, an interdisciplinary approach was established with the aid of the Departments of Ophthalmology and Speech Therapy. The patient presented a good response to the treatment and an improvement of functions like speech and swallowing. The mother reported an increase in self-esteem and interpersonal relationships at school and family environment after denture placement (Figure 6 A and B).



**Figure 6.** Frontal (A) and lateral (B) of the patient after placement of the dentures

## Discussion

HED is a X-chromosome linked recessive syndrome, which affects mostly males, like the patient of this case, and has the mother as the unavoidable carrier of the syndrome.<sup>3,4,8</sup>

Although hypotrichosis is one of the major clinical manifestations of HED,<sup>2,3</sup> our patient did not exhibit a decreased amount of scalp hair, but he had scant eyebrows and eyelashes. It is usual to have a deficiency or absence of the mucous and sweat glands,<sup>3,4</sup> which in our patient resulted in reddish

eyes and a dry and pale skin. Another common finding in HED is the decrease of occlusal vertical dimension due to oligodontia,<sup>2</sup> but it did not apply to our patient because the two first molars in each arch were in vertical and transverse occlusal stability.

The ideal surgical and prosthetic approach should consider the patient's age, skeletal and dental maturity and available bone volume. Dentures made at an early age might lead to significant improvements in self-esteem, speech and chewing, as observed in this case. The restoration of a natural and pleasant appearance is important for the future social integration of children with HED.<sup>9,10</sup> Prosthetic treatment is generally recommended since the age of five, although dentures can be installed even earlier, at three and four years of age.<sup>6,11,12</sup>

Due to family's lack of information and deprived economic condition, the oral rehabilitation of this patient started later than usually recommended, which prevented him to benefit from the early installation of dentures. It also explains the presence of the narrow alveolar crest, especially in the anterior mandibular region, which led to the choice for an alternative temporary denture supported on the PFMs.

Patients with hypodontia are often treated with partial or complete removable dentures.<sup>6,11,12</sup> However, the use of mini-implants have been indicated for the treatment in HED cases, considering that these patients may have smaller bone volume and mini-implants require fewer surgical procedures surgeries and have a shorter healing time compared with conventional implants.<sup>7,9,10</sup> The treatment proposed in this case report could also be indicated for young patients with other variations of ectodermic dysplasia who have some kind of tooth agenesis and could benefit from the improvements achieved. However, this treatment is contraindicated for patients with very thin alveolar bone crest (as observed in the mandibular arch of our patient); children below 3 years of age due to difficulty of performing this procedure in such young patients; and patients who cannot attend regular clinical and radiographic follow-up.<sup>6,7,10</sup>

One of the major reasons for using mini-implants in oral rehabilitation of patients with HED is to preserve the alveolar bone in order to improve the prognosis of the future definitive denture with the benefits of having increased retention and stability, which ultimately results in improved function and aesthetics.<sup>6,9,13</sup>

However, further long-term clinical studies are necessary to evaluate and validate this alternative treatment for the rehabilitation of children with HED. Prostheses for young patients should be closely monitored for adjustments or replacements and therefore re-evaluations at three- to six-month intervals are highly recommended.<sup>10,14,15</sup>

Despite the favorable outcomes observed so far, the treat-

ment presented in this case report is recent and has yet a short follow-up period to be considered as successful. A quarterly monitoring interval was established to observe routine factors related to denture use, such as oral hygiene and fitting. In addition, an annual radiographic follow-up will be performed.

## Conclusion

The use of mini-implant supported fixed dentures is a viable alternative for patients in the growing phase. This approach aims to facilitate denture fitting and improve functional aspects such as speech, mastication and swallowing, as well as aesthetics. However, a meticulous long-term follow-up is required until the end of the growth spurt.

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## Mini Curriculum and Author's Contribution

- Natalia Luiza Lini Ferreira – DDS. Contribution: data collection, manuscript writing, manuscript review and clinical care of the patient.
- Fernanda Silva Viana – DDS. Contribution: clinical care of the patient, manuscript writing, manuscript review.
- Leonardo Foresti Soares Menezes - DDS and MSc. Contribution: data collection, manuscript writing, manuscript review and preparation of the patient's treatment plan.
- Esdras de Campos França - DDS and MSc. Contribution: coordination of the clinical care of the patient, data collection, photographs of the case, manuscript review.
- Henrique Pretti - DDS and PhD. Contribution: manuscript review and preparation of the patient's treatment plan.
- Patrícia Maria Zarzar - DDS and PhD. Contribution: manuscript advisor, coordinated the development and critical review of the manuscript and preparation of the patient's treatment plan.

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