

## Oral rehabilitation of a preschool child with ectodermal dysplasia through fixed space maintainers in a case of 7 primary incisors oligodontia – Case report

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**Abstract:** **Introduction:** Oligodontia is a common manifestation of Ectodermal Dysplasia with severe implications for oral aesthetics and function. The adaptation of space maintainers can be challenging due to the complexity of missing teeth and facial growth in children. **Objective:** report and describe the manufacturing and adaptation of fixed dentomuco-supported space maintainers in the maxilla and mandible of a child with Ectodermal Dysplasia. **Case Report:** A 4-year-old patient with Ectodermal Dysplasia and oligodontia of seven primary incisors was subjected to adaptation of fixed space maintainers. For the maxilla, a modified Nance space maintainer was fabricated with an acrylic resin extension to the vestibular to simulate gingival tissue, and artificial primary teeth were fabricated using prefabricated acetate crowns. For the mandible, a lingual arch was fabricated with an acrylic resin extension to the vestibular in the region where the four lower primary incisors were missing to simulate gingival tissue, and artificial lower primary incisors were also fabricated. **Results:** The patient showed excellent adaptation to space maintainers, with improvements in masticatory function, posture, and tongue function. The family reported psychological improvement in the child, who had stopped attending school due to embarrassment and returned to regular school activities. **Conclusion:** Space maintainers are effective in restoring function and aesthetics in complex cases of oligodontia associated with Ectodermal Dysplasia.

**Key words:** Space Maintenance, Orthodontic; Anodontia; Ectodermal Dysplasia.

## Rehabilitación oral de un niño en edad preescolar con displasia ectodérmica mediante mantenedores de espacio fijos en un caso de oligodoncia de 7 incisivos primarios – Reporte de caso

**Resumen:** **Introducción:** La oligodoncia es una manifestación común de la Displasia Ectodérmica y presenta implicaciones severas para la estética y la función oral. La adaptación de los mantenedores de espacio se convierte en un desafío debido a la complejidad de los dientes ausentes y al crecimiento facial en niños. **Objetivo:** informar y describir la fabricación y adaptación de mantenedores de espacio fijos dento-mucosoportados en el maxilar y la mandíbula de un niño con Displasia Ectodérmica. **Reporte de caso:** un paciente de 4 años con Displasia Ectodérmica y oligodoncia de siete incisivos primarios fue sometido a la adaptación de mantenedores de espacio fijos. Para el maxilar, se confeccionó un mantenedor de espacio tipo Nance, modificado con una extensión de resina acrílica hacia el vestíbulo para simular el tejido gingival, y se fabricaron dientes primarios artificiales mediante coronas prefabricadas de acetato. Para la mandíbula, se confeccionó un arco lingual con una extensión acrílica hacia el vestíbulo, en la región de los cuatro incisivos inferiores ausentes, para simular tejido gingival, y también se fabricaron incisivos primarios inferiores artificiales. **Resultados:** el paciente presentó una excelente adaptación a los mantenedores de espacio, con mejoras en la función masticatoria, la postura y la función lingual. La familia reportó una mejoría psicológica en el niño, quien había dejado de asistir a la escuela por vergüenza y había retomado sus actividades escolares habituales. **Conclusión:** los mantenedores de espacio son efectivos para restaurar la función y la estética en casos complejos de oligodoncia asociada a Displasia Ectodérmica.

**Palabras clave:** Mantenedor de Espacio Ortodóncico; Anodoncia; Displasia Ectodérmica.

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## Reabilitação oral de criança pré-escolar com Displasia Ectodérmica através de mantenedores de espaço fixos em caso de Oligodontia de 7 incisivos decíduos – Relato de Caso

**Resumo: Introdução:** Oligodontia é uma frequente manifestação da Displasia Ectodérmica com severas implicações na estética e função oral. A adaptação de mantenedores de espaço se torna um desafio devido às inúmeras ausências dentárias e ao crescimento facial em crianças. **Objetivo:** relatar e descrever detalhadamente a confecção e adaptação de mantenedores de espaço fixos dento muco suportados na maxila e mandíbula de uma criança portadora de Displasia Ectodérmica. **Descrição:** paciente de 4 anos de idade portador de Displasia Ectodérmica com oligodontia de 7 incisivos decíduos foi submetido à adaptação de mantenedores de espaço fixos. Para maxila foi confeccionado um mantenedor de espaço tipo Botão de Nance modificado com extensão de resina acrílica até vestibular para simular o tecido gengival, e foram confeccionados dentes decíduos artificiais. Na mandíbula foi confeccionado arco lingual com extensão de resina acrílica até vestibular na região da ausência dos 4 incisivos decíduos inferiores para simular o tecido gengival e também foram confeccionados incisivos decíduos inferiores artificiais com o uso de moldeiras de acetato pré-fabricadas. **Resultados:** paciente apresentou excelente adaptação aos mantenedores de espaço com melhora da função mastigatória e de postura e função de língua. Família relatou melhora psicológica da criança que não frequentava mais a escola por vergonha e voltou às atividades escolares rotineiramente. **Conclusão:** mantenedores de espaço são eficazes para reestabelecer função e estética em casos complexos de oligodontia associada à Displasia Ectodérmica.

**Palavras-chave:** Mantenedor de Espaço em Ortodontia; Anodontia; Displasia Ectodérmica.

### Introduction

Dental caries, dental trauma, tooth impaction, or the congenital absence of tooth germs can cause tooth absence in children.<sup>1-3</sup> Among the consequences of missing teeth in children are the reduction of the arch perimeter due to mesial drift of adjacent teeth—which may even result in impaction of the permanent successors—decreased masticatory efficiency, psychological distress associated with esthetic impairment, reduced quality of life, and the development of atypical tongue function during swallowing, resting, and speech.<sup>2,4,5</sup>

When tooth absence occurs in children, whether due to premature tooth loss or hypodontia, space maintainers should be considered. Space maintainers are removable or fixed appliances fabricated from stainless steel and/or acrylic resin, and they may include natural or artificial teeth as part of their design. Their main objectives are to prevent the migration of adjacent and opposing teeth into the edentulous space, to

preserve the mesiodistal space, to maintain vertical dimension, and to restore esthetics and function.<sup>6</sup>

In this clinical case, we describe the fabrication and placement of upper and lower space maintainers in a child presenting with oligodontia involving 7 missing incisors and Ectodermal Dysplasia (ED). Ectodermal dysplasia is a rare, heterogeneous group of genetic disorders that affect the development of ectodermal tissues, including hair, nails, skin, teeth, and sweat glands. The genes most commonly affected in this syndrome belong to the EDA/NF- $\kappa$ B signaling pathway, while mutations in WNT10A, TP63, and KRT17 are less frequent.<sup>7</sup> The incidence of ED varies according to the specific subtype, as several clinical variants have been described. The global prevalence of ectodermal dysplasia syndromes is estimated at approximately 6–9 per 10,000 individuals.<sup>8</sup> Hypohidrotic Ectodermal Dysplasia (HED) is the most common form, affecting at least 1 in 5,000–10,000 live births.<sup>8</sup>

## Case Report

A 4-year-old male patient, mixed race, was referred by the Genetics Department of the University Hospital of the Federal University of Espírito Santo (UFES) with a presumptive diagnosis of Ectodermal Dysplasia (ED) and multiple missing teeth, for evaluation and treatment at the university's Pediatric Dentistry outpatient clinic.

The child's parents were informed about the treatment procedures and signed the Informed Consent and Assent Forms, authorizing both the dental treatment and the scientific publication of the case. Among the main complaints from the patient and family, the psychological aspect stood out, as the child no longer wished to attend school due to the absence of his upper and lower incisors.

Extraoral examination revealed facial symmetry, passive lip seal, and good maxillomandibular relationship in the sagittal plane. The patient presented sparse and thin hair and eyebrows, corroborating the previously established diagnosis of ED (Figure 1).

Intraoral examination revealed good oral hygiene, absence of carious lesions or periodontal disease, but absence of the deciduous teeth: maxillary right lateral incisor, maxillary right central incisor, maxillary left central incisor, mandibular left lateral incisor, mandibular left central incisor, mandibular right central incisor, and mandibular right lateral incisor. The maxillary right canine, maxillary left lateral incisor, maxillary left canine, mandibular left canine, and mandibular right canine exhibited a conical morphology (Figure 2).



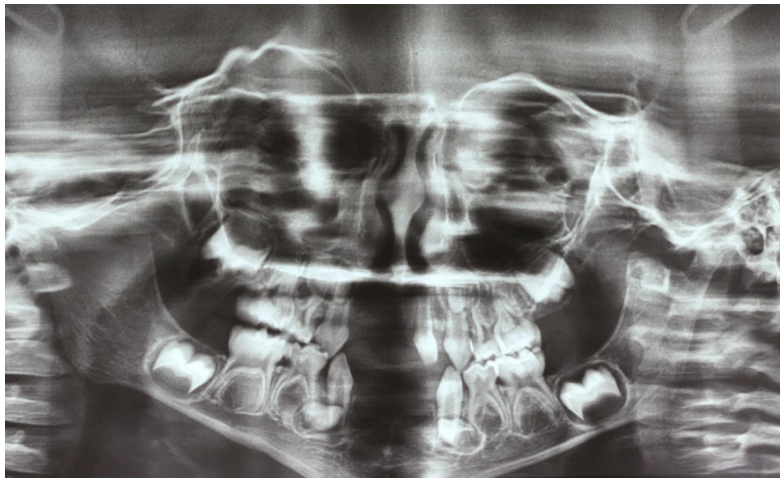
Figure 1. Initial extraoral photographs.



Figure 2. Initial intraoral photographs.

Radiographic examination confirmed oligodontia involving the same teeth and the congenital absence of the permanent successors: maxillary right lateral incisor, maxillary right central incisor, maxillary left central incisor, maxillary left lateral incisor, mandibular left lateral incisor, mandibular left central incisor, mandibular right central incisor, and mandibular right lateral incisor (Figure 3).

The proposed treatment plan consisted of the installation of a fixed dento-mucosa-



**Figure 3.** Panoramic radiographic examination.

supported esthetic and functional space maintainer in both arches, incorporating acrylic base plates with composite resin artificial teeth and orthodontic band anchorage on the second primary deciduous molars.

An alternative treatment plan involved the adaptation of removable acrylic plates with artificial teeth as space maintainers for both arches.

The advantages of the first option included better stability and retention, lack of dependence on patient cooperation, and elimination of the risk of appliance loss—factors particularly relevant for a child of this age. The main disadvantage considered was the potential for appliance fracture; therefore, the family was instructed to ensure the child avoided applying force to the artificial teeth of the maintainers. The family agreed with the proposed treatment plan.

The first clinical step was the placement of separating elastics in the mesial aspects of the maxillary right and left second primary molars. After one week, the elastics were

removed, orthodontic bands were fitted on these teeth, and an alginate transfer impression was taken. The bands were repositioned in the impression, and new separators were placed.

In the laboratory, a Nance button appliance was fabricated using gingiva-colored acrylic resin extending to the vestibular region corresponding to the deciduous maxillary right lateral incisor, maxillary right central incisor, and maxillary left central incisor.

For the fabrication of the artificial teeth corresponding to those incisors, prefabricated acetate crown forms matching the primary incisor morphology were used. The composite resin was inserted incrementally in 2 mm layers, light-cured after each increment. The composite resin color used was BW. The central portion of each artificial tooth was left hollow to allow the acrylic resin from the base to penetrate the structure, achieving mechanical retention in addition to chemical adhesion between the teeth and the vestibular acrylic extension of the Nance button (Figures 4 and 5).



**Figure 4.** Fabrication of artificial primary teeth using composite resin.



**Figure 5.** Fixed esthetic and functional dento-mucosa-supported space maintainers for maxillary and mandibular incisors.

Before cementation, a clinical try-in was performed and minor adjustments were made to relieve pressure from the acrylic in the upper labial frenum area.

For cementation of the maxillary space maintainer, relative isolation was achieved using a lip retractor, and acid etching with 37% phosphoric acid was performed on the buccal surfaces of the maxillary right

and left second primary molars for 30 seconds.

After cementation, the morphology of the maxillary left lateral incisor was recontoured using direct composite resin restoration (Figure 6).

For the mandibular space maintainer, orthodontic bands were fitted on the mandibular left and right second primary molars, and a transfer impression was taken. In the laboratory, a lingual arch with acrylic extension toward the vestibular region of the mandibular left and right lateral and central incisors was fabricated, along with composite resin artificial teeth created using prefabricated lower primary incisor crowns, following the same incremental technique.



**Figure 6.** Adaptation of the upper space maintainer and reanatomization of the maxillary left lateral incisor (tooth #62).

## Treatment Results

The results achieved were satisfactory, with good adaptation and acceptance of the space maintainers by the patient and no reports of discomfort or complications. The appliances allowed proper tongue function during speech and swallowing, restored the dental and gingival esthetics of the smile, and improved lip support (Figures 7 and 8). After 90 days, the space

maintainers demonstrated good stability, and the patient continues to be followed up through periodic clinical evaluations.

The child's family reported a significant improvement in psychological well-being, as the patient became more willing to participate in routine school and sports activities and felt more confident and happy to smile.



*Figure 7. Final intraoral photographs.*



*Figure 8. Final extraoral photographs.*

## Discussion

The child's parents reported that their primary motivation for seeking care at the University Hospital was concern about the absence of tooth formation, because this condition not only affected the esthetics of the child's smile and compromised masticatory function, but also had a considerable emotional impact. The parents related that the child no longer wished to attend school or interact with other children before treatment. Unfortunately, this situation is not uncommon, and previous studies have demonstrated that poor dental esthetics is a major cause of bullying among children.<sup>9,10</sup> In this context, the treatment performed was highly effective in improving the child's psychological and emotional condition. The patient expressed great joy at having teeth for the first time when smiling and reported feeling eager to show them to classmates and family members.

Although successful outcomes with removable space maintainers have been reported in similar cases of oligodontia in children with Ectodermal Dysplasia (ED)<sup>11</sup>, our decision to use fixed space maintainers instead of removable ones was justified by the patient's young age, reduced likelihood of cooperation, and the risk of appliance loss.

Regarding the potential restriction of maxillary transverse growth, the patient will be monitored every six months, and if necessary, the maxillary maintainer will be removed and expanded in the region of the orthodontic bands to compensate for any growth limitation. It is important to emphasize that, according to Enlow,<sup>12</sup> the alveolar growth pattern of the maxilla follows a "V-shaped" direction—posteriorly and laterally—which is not affected by the

cementation of the fixed space maintainer. Similarly, mandibular alveolar growth also follows a "V-shaped" pattern and is not compromised by the cementation of the lower space maintainer.

Furthermore, the peak period of maxillomandibular development occurs around eight years of age<sup>12</sup>, at which time new space maintenance strategies should be considered, such as a modified Haas appliance with artificial teeth, given the congenital absence of the permanent incisors.

The overall treatment plan involves maintaining the current space maintainers until approximately six to seven years of age, followed by an assessment of facial growth and possible orthopedic intervention if any transverse or sagittal discrepancy is identified. From the age of eight, Temporary Anchorage Devices (TADs) should be installed in the anterior region to support the placement of provisional crowns made of pressed composite resin over mini-implants.<sup>13</sup> Upon completion of craniofacial growth in adulthood, implant-supported prosthetic rehabilitation with porcelain crowns is expected to provide an esthetically and functionally satisfactory long-term outcome, suggesting a favorable prognosis for this case.

## Conclusions

Fixed esthetic and functional dento-mucosa-supported space maintainers proved to be a satisfactory treatment alternative for patients with the absence of primary maxillary and mandibular incisors. These appliances were effective in restoring both esthetics and function, resulting in significant improvement in the

emotional and psychological well-being of children with missing teeth.

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### Conflict of Interest and Funding Statement

No artificial intelligence tools were used at any stage of the article's production.

The authors declare no conflicts of interest related to this case report.

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