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Generalized Odontodysplasia: A Rare Case with 19-Year Follow-up

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Authors' contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Aims: Regional Odontodysplasia is a dental anomaly with an unknown, non-hereditary etiology, which affects the dental tissues of both dentitions and commonly affects only one quadrant. When it affects multiple quadrants and goes beyond the midline, it is called Generalized Odontodysplasia.
Presentation of Case: This case report is of a patient diagnosed with Generalized Odontodysplasia and followed for 19 years from diagnosis to treatment. The patient presented to the dental office with changes in the shape and absence of some upper and lower teeth. The pathology was diagnosed by combining clinical findings and complementary tests, such as radiographic examination and histopathological analysis. The treatment involved several specialties.

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Discussion: The treatment of Generalized Odontodysplasia depends on each case, taking into account age, extent of involvement and patient and family expectations. After pubertal growth is complete, rehabilitation with definitive implants can be considered.
Conclusion: When diagnosed early, it is possible to have good results with oral rehabilitation in patients with Generalized Odontodysplasia.

Keywords: Regional odontodysplasia; generalized odontodysplasia; dental dysplasia; agenesis.

1. INTRODUCTION

Regional Odontodysplasia is a non-hereditary dental anomaly that compromises the development of the enamel, dentin and pulp of deciduous and permanent teeth (Neville et al., 2012; Volpato et al., 2008). Its manifestation can lead to the absence of tooth eruption, most frequently affecting female sex and the anterior region of the maxilla. It usually affects only one quadrant and does not cross the midline. When the affected area is bilateral, multiquadrant and crosses the midline, it is called Generalized Odontodysplasia (Kahn, Hinson, 1991).

The etiology of Odontodysplasia is uncertain. There may be an association between various factors, such as the use of medication during pregnancy, radiotherapy, local trauma, genetic factors and vascular problems, but no theory can explain this pathology (Neville et al., 2012).

Diagnosis involves a combination of clinical findings, such as the absence of teeth and anomalies in tooth color and shape. The radiographic examination reveals characteristics as the appearance of “ghost teeth” (Cho, 2006; Magalhães et al., 2007; Volpato et al., 2008).

Case reports can clarify the wide range of clinical presentations of Odontodysplasia, such as teeth with defective enamel, abnormal tooth morphology and incomplete tooth eruption. Documenting these variations can help researchers understand the full spectrum of the disorder. The aim of this article is to report a case of Generalized Odontodysplasia in a child aged 3 years and 11 months, presenting the treatment alternatives used over 19 years until prosthetic rehabilitation with implants. This case report is of great importance to the scientific community, as it offers updated information on the diagnosis, treatment options and prognosis of Odontodysplasia, opportunely as a reference for other professionals in the field.

2. PRESENTATION OF CASE

A female patient, 3 years and 11 months old, leukoderma, with no relevant medical history, presented with delayed eruption of the teeth, maxillary and mandibular atresia and changes in the shape of the teeth, which had a pointed and narrow appearance. According to the mother, there were no family members with this anomaly and there had been no eventualities during pregnancy.

The first panoramic X-ray at 3 years and 11 months revealed the absence of tooth 51 and the absence of the germs of teeth 21, 41, 42, 43 and 44. It was difficult to see the germs of teeth 11, 12 and 13 due to possible anatomical changes. Teeth 52, 53, 81, 82, 83 and 85 also had changes in shape. Quadrant 3 was the only one not affected.

Tooth 85 was extracted due to extensive decay and sent for histopathological examination, which revealed reduced thickness and deficient mineralization of the enamel and dentin. The enamel presented irregular structure and the dentin presented dysplasia, with a small amount of dentinal tubules. According to the clinical, radiographic and histopathological characteristics, the diagnosis was Generalized Odontodysplasia. The treatment was carried out with the aim of preserving the erupted teeth for as long as possible, to allow better chewing, phonation and development of the dental arches until the beginning of the definitive rehabilitation treatment.

A new panoramic radiograph was taken at 7 years and the clinical examination showed marked maxillary atresia, posterior and anterior crossbite, absence of element 51 and changes in the shape and size of elements 52, 53, 81, 82 and 83 (Fig. 1).

A McNamara appliance was used for 7 months to correct the crossbite (Fig. 2). Then, a removable retaining appliance was installed in the upper arch.

At the age of 9, a panoramic radiograph showed the tooth germs of elements 11, 12 and 13, which had not erupted in the oral cavity and showed changes in shape and size. The teeth were small and malformed with less radiopacity of the enamel and crowns with appearance of corrosion. There was also a delay in the development of the Nolla stages of teeth 41, 42, 43 and 44, while the homologous teeth were already at a more advanced stage of development.

At this point, a removable partial denture with upper anterior teeth was made in order to improve the patient's phonation, aesthetics and social interaction, as well as acting as a space maintainer.

At the age of 13, the patient was referred to begin surgical rehabilitation treatment. The unerupted teeth 11, 12 and 13 were extracted, followed by the installation of temporary implants to support a temporary fixed prosthesis for elements 11, 12, 13 and 21 to restore aesthetics (Fig. 3).

At the age of 14, corrective orthodontic treatment was started to improve occlusion and tooth positioning. Later, at the age of 16, the teeth 41, 42, 43 and 44, which had not erupted in the oral cavity, were extracted.

At the age of 18, single implants were installed in the region of teeth 11, 12, 13 and 21. At this point, the upper fixed appliance was removed, and the lower fixed appliance was maintained until the lower implants were installed, in order to prevent tooth movement. During the same period, the third molars were extracted.

Even with maxillary expansion in childhood, there was no satisfactory mandibular growth. Minimally invasive orthognathic surgery was indicated and performed at the age of 20, with mandibular advancement only. At the age of 22, lower implants were installed for teeth 41, 42 and 43 (Fig. 4).

The case was completed with the installation of single-unit prostheses on implants and ceramic veneers, seeking to establish harmony throughout the smile (Fig. 5).

Fig. 1. Panoramic radiograph and frontal view at 7 years and 1 month
Source: Patient files

Fig. 2. Frontal view after maxillary disjunction with McNamara appliance
Source: Patient files

Fig. 3. Panoramic radiograph and frontal view after the installation of temporary implants and prosthesis at the age of 13
Source: Patient files

Fig. 4. Panoramic radiograph after the installation of the upper and lower implants, third molar extraction and orthognathic surgery
Source: Patient files

Fig. 5. Intraoral result after cementation of the ceramic veneers and installation of the crowns on implants
Source: Patient files

3. DISCUSSION

The clinical characteristics of teeth affected by Odontodysplasia include hypoplastic defects, resulting in changes in the shape and size of the teeth, which tend to be smaller than normal, with a yellowish or brownish color and an appearance of erosion (Cho, 2006; Magalhães et al., 2007 and Volpato et al. 2008).

Radiographic examination showed the appearance of “ghost teeth”, with no precise demarcation between enamel and dentin and less radiopacity (Neville et al., 2012; Tervonen et

al., 2004). In all the cases reported by Alotaibi et al. (2019), the “ghost teeth” were present, with different degrees of alteration of the dental structure.

The decision to extract the teeth affected by this anomaly must be made considering the specifics of each case (Cho, 2006). When the patient is very young, the affected teeth should be kept for as long as possible for the maxilla and mandible to develop satisfactorily (Kahn et al., 1991; Volpato et al., 2008), as occurred in the case presented.

In this patient, the affected upper teeth were extracted, and a temporary acrylic prosthesis was installed, as reported by Kahn et al. (1991) and Tervonen et al. (2004). On the other hand, the lower teeth, which were intraosseous, were only extracted when the patient was 16 years old, as they helped to maintain the thickness and height of the bone (Cho, 2006). When erupted, the affected teeth can be maintained through pulpectomy and restorative treatment, which is a more conservative approach, especially in situations where these teeth have not been so severely affected. In this case, extractions were performed only when the teeth were not viable to be kept in the mouth.

Orthodontic treatment was carried out to enhance future oral rehabilitation using implants and implant-supported prostheses. It is worth noting that the installation of definitive implants can only be carried out after pubertal growth has been completed (Cunha et al., 2020). This factor is a major challenge for the rehabilitation stage, since the absence of anterior teeth can cause significant psychological damage, especially to young people. In the observed case, temporary implants were installed when it was not yet possible to install a permanent implant.

There is no single way to treat this anomaly, as many issues must be taken into consideration, such as the expectations of the patient and parents, the conditions for carrying out the treatment and age, among others (Volpato et al., 2008). Many cases have been conducted in a similar way to this study. Lopes-Delphino, Assao and Almeida (2023) reported a case in which rehabilitation was carried out with a temporary prosthesis, proservation, orthodontics, surgery, and rehabilitation with a prosthesis on an implant. Carreira et al. (2011) documented a case in which rehabilitation was carried out only with a temporary prosthesis, due to the patient's young age. Cho (2006) opted for a more conservative approach by maintaining the teeth to stimulate proper development of the maxilla and mandible, and monitoring the case over the time.

Although there have been a few studies on Generalized Odontodysplasia reported in the literature, none of them have shown a 19-year follow-up involving several specialties.

4. CONCLUSION

The treatment of Generalized Odontodysplasia is challenging for both the dental surgeon and the patient, but when diagnosed early and with the help of various specialties, it is possible to achieve satisfactory and effective oral rehabilitation.

CONSENT

All authors declare that written informed consent has been obtained from the patient for publication of this study.

ETHICAL APPROVAL

All the authors declare that all the experiments were examined and approved by the Unioeste Human Research Ethics Committee (CEP), CAAE number 77751924.5.0000.0107, and were therefore carried out in accordance with the ethical standards established in the 1964 Declaration of Helsinki.

DISCLAIMER (ARTIFICIAL INTELLIGENCE)

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc.) and text-to-image generators have been used during the writing or editing of this manuscript.

COMPETING INTERESTS

Authors have declared that they have no known competing financial interests or non-financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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