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CASE REPORT

NON-SYNDROMIC OLIGODONTIA WITH MULTIPLE RETAINED DECIDUOUS TEETH: A RARE CASE REPORT

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ABSTRACT

Oligodontia is a congenital tooth agenesis with the absence of six or more permanent teeth. This clinical report describes a patient with non-syndromic partial oligodontia, with retained deciduous teeth and the absence of 19 permanent teeth. The absence of permanent teeth in young patients may cause aesthetic, functional and psychological problems. Considering the level of complexity in the management of oligodontia patients, treatment should start with proper diagnosis to prevent future functional and esthetic problems.

Key words:

Anodontia, Hypodontia,
Developmental Anomaly,
Oligodontia.

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INTRODUCTION

Tooth development is a complex process, in which sequential interactions between epithelial and mesenchymal cells regulate cell activities like proliferation, condensation, migration and differentiation. Dental agenesis is the common developmental anomaly in human which can occur in an isolated fashion or as a part of syndrome. Isolated cases of missing teeth can be sporadic or familial in nature. Various terminologies have been used to describe the agenesis of teeth in the primary or permanent dentition, like Hypodontia - agenesis of one or few teeth (up to six), Oligodontia - agenesis of six or more teeth excluding the third molars and anodontia is the extreme of oligodontia where there is total absence of any dental structure. The prevalence of hypodontia in the primary dentition ranges from 0.08% to 1.55% and in the permanent dentition ranges from 2.3% to 11.3% depending on the population investigated².

Oligodontia has prevalence rate of 0.3% in permanent dentition and females are more frequently affected than males by a ratio of 3:2³. The mandibular second premolar is the most frequently missing tooth after the third molar, followed by the maxillary lateral incisor and the maxillary second premolar. Agenesis of maxillary central incisors, canines or first permanent molars seems to be rather exceptional⁴. Oligodontia is often associated with specific syndromes or severe systemic abnormalities, while anodontia is commonly seen in severe cases of ectodermal dysplasia^{5,6}. The etiology of oligodontia is unknown; however, both environmental and genetic factors can cause a failure of tooth development. Environmental factors include children treated with irradiation at tooth developing stages or those in whom chemotherapeutic agents have been administered^{3,7}. Genetic factors are constituted by two mutated genes, MSX-1 and PAX-9 in humans^{8,9}. Oligodontia of permanent teeth is usually associated with retained primary teeth.

The purpose of this article is to report a rare case of non-syndromic oligodontia with agenesis of nineteen permanent teeth excluding the third molars and multiple retained deciduous teeth in a 22-year old male patient.

Case Report

A 22-yr male patient reported with the chief complaint of multiple unerupted teeth, difficulty in chewing and psychosocial problems. The patient gave insignificant family history. General examination revealed no abnormalities of the skin, hair or nails. Patient had a history of allergy on hands and sole of feet. Extra oral examination revealed symmetric face and a straight profile and normal vertical dimension. Intra oral examination (Fig: 1) revealed the presence of eighteen retained deciduous teeth and only 12 permanent teeth (oligodontia). The present teeth were 11, 53, 55, 16, 17, 18, 61, 62, 63, 65, 26, 27, 28, 71, 72, 73, 74, 75, 36, 37, 38, 81, 82, 84, 85, 46, 47 and 52 was missing.



Fig. 1. Intraoral photograph showing single upper front permanent tooth i.e 11 and retained deciduous teeth



Fig. 2. Intraoral photograph of maxillary arch showing missing 52 and root stumps wrt 54 and 64

The retained deciduous teeth were morphologically well formed except root stumps wrt to 54, 64, 83 and caries wrt 53, 54, 62, 74, 75 and 84. Slight attrition seen among lower anterior deciduous teeth and 61. Single permanent tooth found in a upper front region i.e 11. Generalized spacing was evident in upper and lower front region. Maxillary and mandibular arches were properly developed. Examination also revealed anterior deep bite and normal posterior bite or class I molar occlusion.

Since the patient reveals lesser no. of permanent teeth than normally expected for the chronological age, so an Opg (orthopantograph) was advised to the patient to know about the developing dentition. Opg (Fig:4) revealed the absence of nineteen permanent teeth excluding the impacted 48. Generalized External root resorption was seen in the retained deciduous teeth.



Fig. 3. Intraoral photograph of mandibular arch showing retained deciduous teeth and well erupted permanent molars bilaterally



Fig.4. Opg reveals the absence of 12-15,21-25,31-35and 41-45 with impacted 48

DISCUSSION

Oligodontia is the medical term used commonly in describing the phenomenon of congenitally missing teeth. Oligodontia is classified as isolated or non-syndromic and syndromic hypodontia¹⁰. Although oligodontia can occur over with 60 different syndromes, these anomalies can occur without any syndrome or systemic disease. Syndromes commonly associated are anhidrotic ectodermal dysplasia, Vander-Woude syndrome, Down's syndrome, Pierre Robin syndrome and Ehler Danlos syndrome. However, oligodontia is seen more common in non-syndromic or familial form than syndromic form⁴. The biological basis for the congenital absence of permanent teeth is partially explained by the failure of the lingual or distal proliferation of the tooth bud cells from the dental lamina. The causes of hypodontia are attributed to environmental factors such as irradiation, tumours, trauma, hormonal influences, rubella, and thalidomide or to hereditary genetic dominant factors, or to both⁵. MSX1 and PAX9 genes play an important role in early tooth development. PAX9 is a paired domain transcription factor that plays a critical role in odontogenesis.

All mutations of PAX9 identified to date have been associated with nonsyndromic form of tooth agenesis⁸. Oligodontia condition should not be neglected as it may result in various disturbances like dearranged occlusion, altered facial appearance which may cause psychological distress, difficulty in mastication and speech. Treatment depends on extent of hypodontia and should consist of interdisciplinary approach. Therefore early diagnosis is important in such conditions. Case of tooth agenesis should be recorded with complete clinical history including medical and radiological investigations to rule out any syndrome¹¹. There may be various treatment options for retained deciduous teeth: either retain, retain and modify extraction and prosthetic replacement, extraction and space closure. In this case, treatment plan i.e implant supported fixed prosthesis or removal prosthesis was explained to the patient.

Conclusion

Oligodontia is mostly considered to be associated with several syndromes but non syndromic aspect of Oligodontia should also be taken into consideration. This case reports the rare occurrence of nonsyndromic hereditary oligodontia. Complete case history with detailed medical history and relevant radiographic investigations are required to eliminate association with any syndrome. Correct diagnosis and proper treatment planning to achieve both prosthetic and aesthetic functionality of teeth.

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