Case Report

A rare occurrence of nonsyndromic focal microdontia of primary teeth with hypodontia of permanent teeth in a pediatric patient

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Abstract

Dental anomalies in human dentition consist of a considerable variation in size, number, position, shape and structure of the tooth. Microdontia is defined as a condition in which teeth are abnormally smaller in size. Hypodontia is defined as developmental absence of 1–5 teeth excluding third molars. Simultaneous occurrence of multiple dental anomalies is most commonly seen in patients with some abnormality or syndrome; it can also occur in nonsyndromic patients. This case report describes about an unusual simultaneous occurrence of dental anomalies such as localized microdontia of primary teeth and hypodontia in a nonsyndromic 10-year-old male patient. Simultaneous occurrence of multiple dental anomalies in a nonsyndromic patient is rare. As the treatment of such cases involves multiple phases, identification and proper timely management is important to help the patient lead a better quality life.

Keywords: Focal microdontia, hypodontia, nonsyndromic, pediatric patient

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INTRODUCTION

Dental anomalies in human dentition consist of a considerable variation in size, number, position, shape and structure of the tooth. Such anomalies may occur due to genetic and environmental factors during morphodifferentiation or histodifferentiation stages of tooth development.[1]

Dental anomalies which involve variation in size of the tooth include microdontia and macrodontia. Microdontia is defined as a condition in which teeth are abnormally smaller in size. According to Shafer *et al.*, microdontia is classified as localized microdontia, relative generalized microdontia and true generalized microdontia. Microdontia is a rare condition. The frequency of microdontia in permanent dentition is 2.5%, whereas in primary dentition, it is <1%. The most common form of localized microdontia is peg laterals which involve microdontia of maxillary lateral incisors.[2]

Hypodontia is defined as developmental absence of 1–5 teeth excluding third molars. The prevalence of hypodontia in permanent teeth excluding 3rd molars is 3%–8.5%, whereas in primary teeth, it is 0.1%–0.7%. The most common teeth which are affected by hypodontia include last tooth of each series, i.e., third molars, second premolars and lateral incisors.[3] Congenitally missing deciduous teeth are not

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commonly seen, but when it occurs it involves maxillary lateral incisors followed by mandibular laterals and cuspids.[4] According to the literature, 15% of missing teeth are lower second premolars, 19% upper second premolars, 29% upper lateral incisors, 11% lower first premolars, 3% lower central incisor and 1% are lower lateral incisor.[5]

Although simultaneous occurrence of multiple dental anomalies is most commonly seen in patients with chromosomal alterations, multisystemic disease and patients with familial history of dental anomalies, it can also be seen in patients with no anomalies, no disease and with no positive dental history.[6]

This case report describes about an unusual simultaneous occurrence of dental anomalies such as localized microdontia of primary teeth and hypodontia in a nonsyndromic 10-year-old male patient.

CASE REPORT

A 10-year-old male patient reported to the Department of Pediatric and Preventive Dentistry, Mahatma Gandhi Dental College and Hospital, Jaipur, with a chief complaint of unerupted permanent teeth in the lower front region. Past medical and dental history of the patient was not relevant. Familial history of the patient was negative, i.e., no other family member showed any past medical history or similar or any other tooth anomaly. The developmental milestone of the patient was normal.

On physical examination, it was seen that the patient had normal appearance, height, weight and intelligence without any other abnormal signs and symptoms. The gait of the patient was normal. On extraoral examination, no evident abnormalities were seen in the eyes, ears, hairs and nails and the face was bilaterally symmetrical. On examination of temporomandibular joint, no clicking sound, crepitus, pain, deviation of mouth or restricted mouth opening was noticed. On examination of lymph nodes, lymph nodes were normal and not palpable.

On intraoral examination, the patient showed mixed dentition phase. Examination revealed the presence of the following teeth (according to FDI-Federation Dentaire Internationale numbering system).

16 55 54 53 12 11 21 22 63 64 65 26

46 85 84 83 42 81 71 32 73 74 75 36

The mandibular primary central incisors were smaller in size as compared to normal. The shape and size of other teeth were normal. The presence of calculus, stains and generalized marginal gingivitis was noted [Figure 1].

To verify whether the patient had any tooth deficiency, a panoramic radiograph was taken. The smaller size of lower primary central incisors was even evident on OPG - Orthopantomogram. Furthermore, the permanent tooth germs of bilateral mandibular central incisors and right mandibular second premolar were absent [Figure 2].

Based on clinical and radiographic findings, the patient was diagnosed with focal microdontia of primary teeth (71 and 81) along with hypodontia of permanent teeth (31, 41 and 45). Since the patient was completely normal with no other past medical history, it was diagnosed as nonsyndromic focal microdontia with hypodontia.

DISCUSSION

Development of tooth is a complex process. It results from the interaction of the epithelium which is derived from first arch and ectomesenchymal cells which is derived from neural crest cells. Ectodermal components include the enamel organ which forms enamel of the tooth while the ectomesenchymal components include dental papilla which helps in formation of dentin and pulp and dental follicle which forms the supporting tissue of the tooth.[7]

The physiologic process of tooth development involves various stages such as initiation, proliferation, histodifferentiation, morphodifferentiation and apposition. Any changes occurring in the normal tooth development process may lead to developmental anomalies of the tooth. The absence of single or multiple teeth may result due to lack of initiation. Any disturbance during morphodifferentiation or histodifferentiation stages of tooth development results in variation in size and form of the tooth.[8]

Although the exact mechanism for variation in the teeth is not known, many studies have shown that environmental

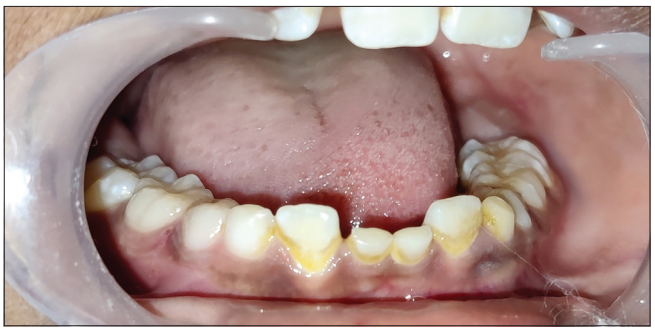

Figure 1: Intraoral view of mandibular arch showing smaller size of primary central incisors

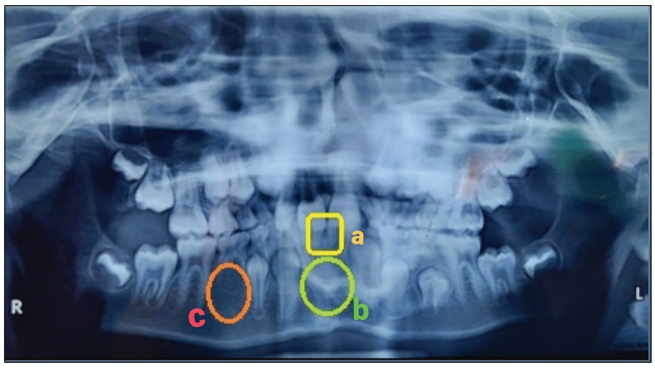

Figure 2: Orthopantomogram showing: a – Microdontia of primary mandibular central incisors; b – Missing permanent tooth germs of mandibular central incisors; c – Missing permanent tooth germ of right mandibular second premolar

factors, genetic mutations and epigenetics lead to developmental anomalies of the teeth.[7]

Microdontia is the variation in size of the teeth in which the teeth are smaller in size than normal. It can occur in both primary and permanent dentition. However, the prevalence in primary dentition is low as compared to permanent dentition. Microdontia occurs more frequently in females than males. The most common teeth affected with microdontia are maxillary lateral incisors; such teeth are referred to as "peg-shaped teeth" or "conical teeth."[9] Microdontia is often related to other syndromes such as Rieger anomaly, orofaciodigital syndrome, oculo-mandibulo-facial syndrome and pituitary dwarfism, also in children with a history of chemoradiotherapy. Microdontia also occurs without any syndrome or positive medical history, however, such occurrence is rare.[7]

Hypodontia is the condition in which there is developmental failure of 1–5 teeth excluding third molars. It is considered as most prevalent dental malformation in humans. Hypodontia is not commonly seen in primary teeth. Mandibular second premolars and the maxillary lateral incisors have been reported to be the most likely affected teeth with hypodontia. Hypodontia can occur as an isolated anomaly or it can occur simultaneously with other dental anomalies such as microdontia, supernumerary teeth, short roots, enamel hypoplasia and taurodontism. Hypodontia is often nonsyndromic, but it can occur in patients with cleft, in patients with Down syndrome and ectodermal dysplasia. Pathogenesis of hypodontia is unclear, and it is considered as multifactorial anomaly. Genetic and environmental factors during tooth development are considered as the cause of such anomalies.[10]

It has been reported that more than 300 genes regulate and control the process of tooth development. The genes related to hypodontia include AXIN2, MSX1 and PAX9. Environmental factor leading to hypodontia includes maternal rubella virus infection, maxillofacial trauma and the use of thalidomide and chemotherapy drugs.[7]

In our case, the patient was nonsyndromic as no abnormalities were seen, and the past medical history of the patient was not relevant. There was simultaneous occurrence of localized microdontia of primary teeth affecting mandibular central incisors and hypodontia affecting permanent central incisors and right mandibular second premolar.

Chen *et al*. reported a case of 9-year-old female patient with nonsyndromic occurrence of true generalized microdontia with hypodontia and variation in maxillary first molar with a single root and single canal.[7] Bargale and Kiran reported the case on a 12-year-old male patient with nonsyndromic occurrence of true generalized microdontia with mandibular mesiodens. Involvement of the entire dentition is usually rare.[11] Anjali *et al*. reported a rare case of bilateral occurrence of hypodontia and microdontia in nonsyndromic pediatric patients.[12]

This case presents an unusual simultaneous occurrence of microdontia of primary teeth and hypodontia of permanent central incisors. Such occurrence is rare as the prevalence of microdontia in primary teeth is <1%[2] and the prevalence of hypodontia affecting mandibular central incisors is 3%.[5]

After formulation of treatment plan for the patient, preventive therapy protocol including oral prophylaxis was performed and oral hygiene instructions were given to the parent and the patient. The patient was also informed about the multidisciplinary therapy plan that will be planned in future including oral surgery, dental implants and orthodontics for the treatment of the patient.

CONCLUSION

Simultaneous occurrence of multiple dental anomalies in nonsyndromic patients is rare. As such cases are not related to any syndrome, identification of such cases becomes difficult. It is very important to identify such cases as it requires multidisciplinary approach for the treatment. Proper treatment plan should be formulated as the treatment of such cases is a long-span process including preventive, corrective and rehabilitative phases. Identification and treatment of such cases should be done timely as it will help maintaining the integrity of the arch and will aid the patient leading a better quality life.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initial s will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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