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

Priyanka Sunil Lekhwani, Nikhil Marwah, Yashi Sharma, Pooja Yadav
Department of Pedodontics and Preventive Dentistry, Mahatma Gandhi Dental College and Hospital, MGUMST, Jaipur, Rajasthan, India

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

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%–8.3%, 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

Access this article online
Quick Response Code:
Website: www.jomfp.in
DOI: 10.4103/jomfp.jomfp_62_21

How to cite this article: Lekhwani PS, Marwah N, Sharma Y, Yadav P. A rare occurrence of nonsyndromic focal microdontia of primary teeth with hypodontia of permanent teeth in a pediatric patient. J Oral Maxillofac Pathol 2022;26:S22-5.

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com
commonly seen, but when it occurs it involves maxillary lateral incisors followed by mandibular laterals and cuspids. 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.

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.

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.

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.

Although the exact mechanism for variation in the teeth is not known, many studies have shown that environmental...
Lekhwani, et al.: A rare occurrence of nonsyndromic focal microdontia of primary teeth with hypodontia of permanent teeth in pediatric patient

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%.\[9\]

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.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

REFERENCES
1. Temilola DO, Folyan MO, Fatusi O, Chukwumah NM, Onyejaka N, Oziegbie E, et al. The prevalence, pattern and clinical presentation of developmental dental hard-tissue anomalies in children with primary and mix dentition from Ile-Ife, Nigeria. BMC Oral Health 2014;14:125.
2. Tahmassebi JF, Day PF, Toumba KJ, Andreadis GA. Paediatric dentistry in the new millennium: 6. Dental anomalies in children. Dent Update 2003;30:534-40.
3. Peker I, Kaya E, Darendeliler-Yaman S. Clinic and radiographical evaluation of non-syndromic hypodontia and hyperdontia in permanent dentition. Med Oral Patol Oral Cir Bucal 2009;14:e393-7.
4. Shafer WG, Hine MK, Levy BM. A Textbook of Oral Pathology. 5th ed. New Delhi: Elsevier Publishers; 2005.
5. Gupta N, Kadu A, Marya CM, Nagpal R, Oberoi S, Dahiya V. Congenitally missing permanent mandibular central incisors and maxillary second molars in conjunction with a supernumerary mandibular central incisor. J Oral Health Community Dent 2017;11:44-7.
6. Suprabha BS, Sumanth KN, Boaz K, George T. An unusual case of non-syndromic occurrence of multiple dental anomalies. Indian J Dent Res 2009;20:385-7.
7. Chen Y, Zhou F, Peng Y, Chen L, Wang Y. Non-syndromic occurrence of true generalized microdontia with hypodontia: A case report. Medicine (Baltimore) 2019;98:e16283.
8. Ezoddini AF, Sheikhha MH, Ahmadi H. Prevalence of dental developmental anomalies: A radiographic study. Community Dent Health 2007;24:140-4.
9. Guttal KS, Naikmasur VG, Bhargava P, Bathi RJ. Frequency of developmental dental anomalies in the Indian population. Eur J Dent 2010;4:263-9.
10. Al-Ani AH, Antoun JS, Thomson WM, Merriman TR, Farella M. Hypodontia: An update on its etiology, classification, and clinical management, Biomed Res Int 2017;8-16.
11. Bargale SD, Kiran SD. Non-syndromic occurrence of true generalized microdontia with mandibular mesiodens – A rare case. Head Face Med 2011;7:19.
12. Anjali PG, Varma B, Kumar JS, Kumar P. Concurrent occurrence of bilateral hypodontia and microdontia in a nonsyndromic paediatric patient: A case report. J Clin Diagn Res 2020;14:3-6.