

## CASE REPORT

## Pre-maxillary hypo-hyperdontia: report of a rare case

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### Abstract

Agenesis of bilateral maxillary canines is very rare and mesiodens is a commonly occurring supernumerary tooth type. Concomitant occurrence of both hypodontia and hyperdontia is extremely rare and it is a condition of mixed numeric variation in the same individual. The reported prevalence of this condition ranges between 0.002% and 3.1%. The purpose of this case report is to describe a rare occurrence of hypo-hyperdontia involving agenesis of both maxillary canines, mesiodens and associated with taurodontism.

**Keywords:** hypodontia, hyperdontia, hypo-hyperdontia, mesiodens, taurodontism.

### Introduction

Hypodontia is a term used for one or few teeth missing than the normal complement of teeth, whereas hyperdontia is a condition of having excess number of teeth to the normal series. Both hypodontia and hyperdontia are two opposite extremes and it is a condition of mixed numeric variation in the same individual [1].

Both environmental and genetic factors have been proposed to explain these anomalies in isolation. Disturbances in differentiation, migration, and proliferation of neural crest cells and interactions between the epithelial and mesenchymal cells during the initiation of odontogenesis have been recommended for this condition. This condition has also been reported in patients with cleft lip and palate, Down syndrome, Dubowitz syndrome, Ellis-van Creveld syndrome, fucosidosis, G/BBB syndrome, Marfan syndrome [2].

This condition may affect both the dentitions and may entail both dental arches [2–4]. The reported prevalence on the hypo-hyperdontia varies from 0.002% to 3.1% [3]. Supernumerary teeth occurs more in males whilst females were commonly affected by hypodontia [5, 6], and hypo-hyperdontia has been frequently reported in males [7].

Most commonly missing teeth are second premolars followed by lateral incisors excluding third molars while mesiodens is a common supernumerary tooth type, which affects anterior part of maxilla.

The purpose of this article is to report an occurrence of concomitant hypo-hyperdontia affecting pre-maxillary region in a 13-year-old female patient.

### Patient, Methods and Results

A 13-year-old female reported to the Department of Pedodontics and Preventive Dentistry (Narayana Dental College and Hospital, Nellore, AP, India) with the chief complaint, abnormal tooth in maxillary anterior region. Medical history was unremarkable and dental history

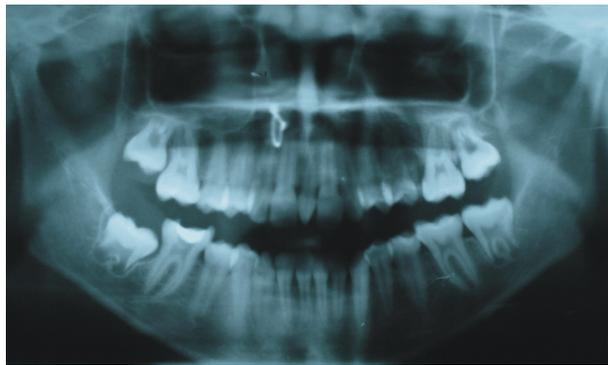
revealed that she had a composite resin restoration on tooth 46 two years ago. There was no history of trauma, infection or metabolic disorders, and extractions. Clinical examinations revealed that she was in permanent dentition with adequate oral hygiene. She brushed her teeth twice a day. The patient was the elder of two siblings born to parents of a non-consanguineous marriage. Intra-oral examination revealed presence of a conical shaped supernumerary tooth in the midline and both teeth 13, 23 and 47 were missing (Figure 1).

Radiographic examination revealed teeth 18, 13, 23, 28, 38, and 48 missing, tooth 47 showed delayed development, and taurodontism was evident in teeth 17 and 27 (Figure 2).

The premolars were well aligned in place on canines in maxillary arch. It was decided to extract mesiodens and refer the patient to orthodontic department for fixed orthodontic appliance treatment to close the space and harmonize the arches. Based upon the history, clinical and radiographic examination, a provisional diagnosis of pre-maxillary hypo-hyperdontia was made with the exclusion of third molars. The treatment plan was explained to parents. Since, the parents and patient declined for the treatment, patient was reviewed in the department until the eruption of tooth 47 and discharged.



**Figure 1 – Intra-oral view showing erupted mesiodens and congenitally missing teeth 13 and 23.**



**Figure 2 – Panoramic radiograph illustrating erupted mesiodens and missing teeth 13 and 23.**

## Discussion

Gibson AC [8] divided hypo-hyperdontia, four subdivisions; pre-maxillary, maxillary, mandibular, and bimaxillary hypo-hyperdontia based on the site of occurrence. The present case serves as good example for pre-maxillary hypo-hyperdontia, since both hypodontia (agenesis of maxillary canines) and hyperdontia (mesiodens) were noted excluding the absence of third molars in both the arches. If third molars (18, 28, 38, and 48) were taken into account, it should be coined as bimaxillary hypo-hyperdontia. Garn SM *et al.* [9] described the association of third molar agenesis with missing teeth from other classes of teeth. The authors concluded that the association among the reduction in the number of other teeth and third molars hysteresis the hypothesis of a field of variable intensity, which, in its greatest degree of expression, eliminates all four third molar teeth.

Hypodontia is commonly seen in females than males with prevalence of 2.91% to 3.22% excluding third molars [10]. The most common missing teeth after third molars were mandibular second premolars in Caucasians [10] while, mandibular incisors are frequently missing in southern Chinese [6]. In the present case, rare occurrence of agenesis of maxillary canines was evident and the reported prevalence of missing canines ranges between 0.06 and 0.45% [11]. A study from Hong Kong reported that, only nine cases (0.012%) were affected by agenesis of bilateral canines in maxilla in 70 000 school children [12], while six cases (0.13%) in 4417 Hungary population [13] and a Japanese study reported six cases (0.16%) in 35 937 subjects [11].

Hyperdontia is common in males and commonly seen in pre-maxillary region. Supernumerary tooth located in maxillary central incisor region is called mesiodens and the prevalence of mesiodentes is between 0.15% and 1.9% [14]. This may occur in single or as multiples, may appear unilaterally or bilaterally, and often do not erupt [15]. In the present case, there was evidence of mesiodens in the anterior region of the maxilla.

Taurodontism has been frequently reported incidence rate ranging from 0.25% to 18% [16]. The association among taurodontism and hypodontia has been studied by several authors [17, 18]. Seow WK and Lai PY [17] reported 34.8% of patients effected by hypodontia had taurodontism, while, Gomes RR *et al.* [18] found one third of their patients. In the present case, there was

evidence of taurodontism in maxillary second permanent molars and this might be coincidental.

The management of hypo-hyperdontia is challenging and needs a multidisciplinary approach, because no standard treatment protocols have been discussed in the literature. In the present case, the treatment options included referral to orthodontics for the management following extraction of mesiodens, however, patient and parents declined for the treatment. The patient was reviewed until the eruption of tooth 47 and discharged from the department. Early diagnosis is a key for the successful management, since it permits the dentist to implement the most appropriate treatment options for the patient to minimize consequences. The treatments may differ from individual to individual. Panoramic radiographs are essential for the diagnosis of hypodontia and/or supernumerary teeth [19].

The case described in the present article represents a good example of pre-maxillary hypo-hyperdontia. Although several hypotheses have been speculated on the occurrence of concomitant hypo-hyperdontia, nevertheless, the etiology of this condition still remains unknown [2–4]. It has been reported that males were commonly affected by this condition than females [7, 8], and it is not affected by gender because of the disparities in gender distribution of the studies [4]. Moore R [20] reported similar findings (pre-maxillary hypo-hyperdontia) in an 11-year-old male patient with agenesis of bilateral maxillary permanent canines and presence of a conical shaped supernumerary tooth in right lateral incisor however, third molars were excluded. The position of supernumerary tooth was different in both cases, so the present case stands for its own uniqueness. In the present case, taurodontism was evident in maxillary permanent second molars, to the best of our knowledge this is first time in the literature to notice in a hypo-hyperdontia patient.

## Conclusions

Concomitant hypo-hyperdontia is an unusually condition which might be diagnosed by regular clinical and radiographic examination. Agenesis of bilateral maxillary canines is very rare in association with mesiodens in the same individual. Our report highlights a rare occurrence of hypo-hyperdontia in an unusual site. The present case is unique case of hypo-hyperdontia with agenesis of both maxillary canines and presence of mesiodens, which is probably rare occurrence. Furthermore, this is only case report describing the evidence of taurodontism in association with hypo-hyperdontia.

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