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Mandibular midline supernumerary tooth associated with agenesis of permanent central incisors: A diagnostic conundrum

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SUMMARY

Concomitant hypo-hyperdontia is a rare mixed numeric anomalous condition. The presence of this condition in the same area of dental arch and specifically in the mandibular anterior region is reported very infrequently. This case report presents a case of 20 years old male with congenitally missing permanent mandibular central incisors in conjunction with a mandibular midline supernumerary tooth. Only 3 cases have been documented in English literature till date. The article focuses on the review of mandibular mesiodens and the clarity regarding the usage of the terminology "mesiodens".

Key words: agenesis, incisors, mandibular, mesiodens, supernumerary.

INTRODUCTION

The term 'concomitant hypo-hyperdontia' (also known as oligopleiodontia) describes the condition where developmental absence of teeth (Hypodontia) and supernumerary teeth (Hyperdontia) are co-existing in same individual (1). The Probability of both numeric dental anomalies co-existing is between 0.0008 and 0.0015 (2). Most of the cases of hypo-hyperdontia have been documented in maxillary arch (1). The etiopathogenesis of Hypo-Hyperdontia is unknown. Various theories like developmental disturbances in proliferation, migration and differentiation of the neural crest cells and interactions between the epithelial and mesenchymal cells during odontogenesis have been proposed (3).The most common supernumerary tooth is mesiodens that is typically present in anterior maxilla between maxillary central incisors. An erupted mesiodens in mandible is a rare phenomenon. But when the mandibular mesiodens co-exists with agenesis of mandibular central incisors, it becomes a unique and debatable entity. Only three similar cases have been reported to occur in mandibular anterior arch in English literature (1, 4, 5). Here we report a case of mandibular midline supernumerary tooth with agenesis of permanent mandibular central incisors. The present article focuses on review of mandibular mesiodens and the clarity of use of terminology "mesiodens" as permanent mandibular central incisors in this case are missing.

CASE REPORT

A 20-year-old male reported to dental O. P. D for the chief complaint of abnormal shape of lower front teeth and spacing between them. The intra-oral examination revealed that both the mandibular permanent central incisors i.e. teeth 24 and 25 were missing and a conical supernumerary tooth was present in midline between permanent right and left lateral incisors i.e. teeth 42 and 32 (Fig.1). The supernumerary tooth was centrally placed, causing a spacing of 3 mm with right and left mandibular lateral incisors. The supernumerary tooth was not causing any problem in the occlusion. Mandibular right first permanent premolar was filled with temporary restoration from a private practitioner a few days ago. His medical history was unremarkable. A family history of supernumerary or congenitally missing teeth was negative. No history of oro-facial

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Fig. 1. Clinical photograph (occlusal view) showing the presence of a conical supernumerary tooth [mesiodensoid] and missing bilateral mandibular central incisors (mirror view)

Fig. 2. Intraoral periapical radiograph showing the presence of a conical supernumerary tooth positioned between mandibular right and left permanent lateral incisors

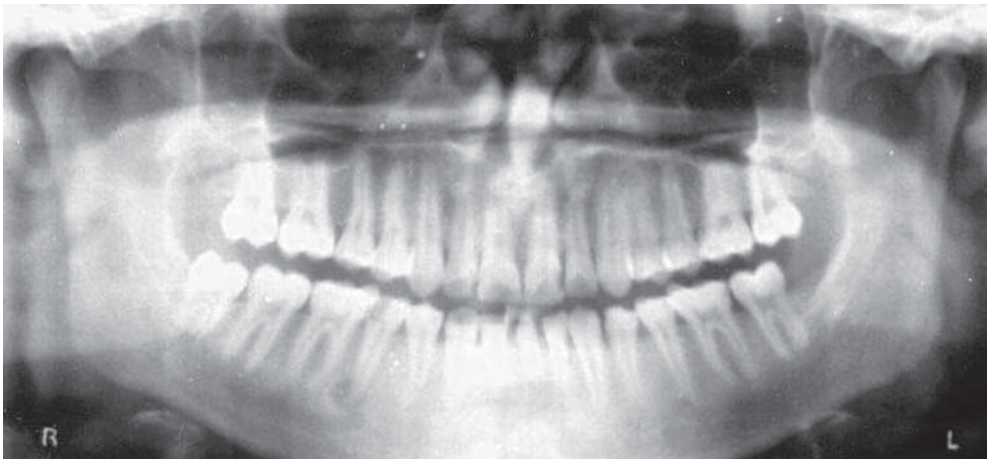

Fig. 3. Panoramic radiograph revealing the absence of mandibular right and left central incisors and presence of a conical supernumerary tooth

trauma and dental extractions could be elicited. There were no other associated signs or symptoms suggesting a syndrome association.

An intra oral periapical radiograph revealed bilateral absence of the permanent mandibular central incisors and presence of a conical supernumerary tooth with completely formed root between mandibular lateral incisors (Fig 2).Panoramic radiograph did not detect any other supernumerary tooth or any other dental anomalies (Fig 3). The only significant finding observed in panoramic radiograph was periapical radiolucency in mandibular right first premolar (44). There was also agenesis of maxillary third molars and mandibular left third molar (18 and 38). A diagnosis of mandibular midline supernumerary tooth associated with agenesis of permanent mandibular central incisors was achieved.

The patient was explained about the condition. The patient was presented the option to extract the mandibular midline supernumerary tooth followed by orthodontic treatment to create arch space in the anterior mandibular region. After the completion of orthodontic treatment dental implants were to be placed in space created in place of permanent mandibular central incisors. However, the patient refused treatment because of financial and time constraints and was ultimately lost for follow-up.

DISCUSSION

Agenesis of teeth is the most common developmental craniofacial anomaly in Humans (6). The etiology of congenital absence of teeth includes physical disruption or obstruction of the dental lamina, reduced arch space, functional abnormalities of the dental epithelium and the failure of initiation of the underlying mesenchyme. Mutations in Genes PAX9, MSX1 and AXIN2 have been implicated as probable cause for the agenesis in non-syndromic hypodon-

Table. List of the documented cases in the literature similar to this case

Authors	Age/gender	Hyperdontia(in addition to mandibular midline supernumerary tooth)	Agenesis	Other significant dental findings
Low (1977)	7/M	-	31, 41	-
Gibson (1979)	6/F	-	31, 41	-
Das (2006)	8/F	-	31, 41	-
Present authors (2011)	20/M	-	31, 41	Agenesis of 18, 28 and 48

tia (6). The usual mode of inheritance of hypodontia is Autosomal dominant, but occasionally autosomal recessive and X-linked and polygenic/multifactorial models of inheritance have also been reported (6).

Agenesis of teeth is more common in permanent dentition and is also more prevalent in females (1.37:1; F:M) (7). Studies have demonstrated that hypodontia tends to be more common in mandible than maxilla (7). Hypodontia of teeth in familial cases has also been reported to be associated with colorectal polyposis and cancer and also with ovarian pathology (6). The prevalence of agenesis of teeth in decreasing frequency involves third molars followed by mandibular second premolars in turn followed maxillary lateral incisors and canines (7). There is a tendency for dental agenesis to occur more unilaterally than bilaterally (7).

Supernumerary teeth are defined as teeth present in addition to the normal set of teeth in the dentition. The prevalence of hyperdontia varies approximately from0.1-3.8% (8). The most common location for the presence of supernumerary teeth is in the maxillary anterior region. "Mesiodens" is a midline supernumerary tooth characteristically positioned in maxillary arch between maxillary central incisors. The prevalence of mesiodens in various populations has ranged from 0.45% in Caucasians, 0.4% in Finnish, 1% in Indians, 0.3% in Turkish and 1.4% in Norwegians (9). Most mesiodens remain impacted but about 25% of mesiodentes erupt (9). Mesiodens can also be associated with various complications such as disturbances in tooth eruption, midline diastema or axial rotation of permanent mandibular central incisors and resorption of adjacent teeth and development of pathology like dentigerous cysts (9).

Numerous theories have been suggested as etiologies of mesiodens such as Phylogenetic reversion theory (Atavism), splitting of developing tooth bud to create two teeth and hyperactivity of dental lamina. The most widely accepted theory among the above is hyperactivity of dental lamina that states that the rests of the dental lamina are initiated to develop into an additional tooth bud, which manifests itself as a supernumerary tooth. Due to various case reports of mesiodens in siblings, twins and families, the role of genetics have also been considered. An autosomal dominant inheritance with incomplete penetration has also been proposed (8).

According to Primosh mesiodens are of two types: Supplemental and rudimentary. Supplemental mesiodens resemble normal tooth and are also termed as incisiform (10). Rudimentary mesiodens can be classified as three types according to morphology as conical, tuberculate and molariform. Conical mesiodens are generally solitary, have completely formed roots and are usually palatal to maxillary central incisors. Inverted mesiodens have also been reported (11). A tuberculate mesiodens is short, occupies a more palatal position and shows incompletely formed roots. Molariform mesiodens presents a premolar-like crown and has completely formed roots. A rare multilobed variety of mesiodens has also been recently reported (12).

The management of mesiodens is generally treatment planned according to the stage of dentition; primary, mixed and permanent. Extraction of supplemental mesiodens in the primary dentition is not recommended because of the risk of displacing or damaging the tooth bud of the developing permanent central incisors. However, in early mixed dentition extraction of mesiodens may allow normal eruption pattern of central incisors (8). The later the extraction of the mesiodens, the greater the chance that the permanent central incisor will not erupt. There is also possibility of permanent lateral incisors erupting and getting mesially drifted, hence leading to space arch loss. An orthodontic management will be required in such cases (8).

In our case report the possibility of a microdont central incisor associated with the congenital absence of the contra lateral mandibular central incisor cannot be excluded. However, the distinct conical shape of the midline tooth that bears no resemblance to central incisor and also reduced root formation as compared to mandibular lateral incisors are the possible rationale why the tooth cannot be contemplated as central incisor. The presence of the conical shaped tooth in exact midline also favors the diagnosis of supernumerary tooth. Therefore, the diagnosis of mandibular midline supernumerary tooth associated with agenesis of mandibular central incisors was performed.

As there was agenesis of mandibular permanent central incisors in our case the dental management of the midline supernumerary tooth becomes different, as our case is unique. The possibility of fixed prosthesis following the extraction of midline supernumerary tooth was not considered feasible because of poor esthetics. Therefore, the best treatment possible in our case report was extraction of the midline supernumerary tooth and orthodontic treatment to create arch space followed by the placement of dental implants due to absence of congenitally missing central incisors. The above case report suggests the need for a careful intra-oral screening of the patients at an earlier age and need for panoramic radiograph at an early stage in case of altered numeric dental anomaly on clinical examination to rule out any congenitally missing tooth/teeth or any other dental anomaly.

Concomitant hypodontia and hyperdontia (also known as hypohyperdontia and oligopleiodontia) have been found more often in the permanent dentition than in the primary dentition (3). Oligopleiodontia is also associated with Cleft Lip/palate, Ellis-van creveld (13) and Down syndromes (14). The occurrence of both hypodontia and supernumerary tooth in the same arch especially in the mandible is an uncommon dental anomaly. Till date only three case reports have been published where mandibular central incisors were absent and a midline supernumerary tooth was present.

The scarcity of case reports of mandibular midline supernumerary tooth associated with agenesis of permanent incisors prevents the authors to do a noteworthy analysis. The only three cases that have been documented, all were in mixed dentition, as compared to our case, which has been found in permanent dentition (Table). However, there was an incomplete root formation of conical supernumerary incisor in the case reported by Das (5). This is slightly different from our case where complete root formation had occurred. There was also agenesis of maxillary third molars and mandibular left third molar. However, this finding cannot be compared to all other similar cases reported, as the age in the other reported cases was very young (6-8 years) (1, 4, 5).

In this case report, the use of terminology "Mesiodens" becomes contentious as the supernumerary tooth is positioned in midline but the permanent central incisors are missing. In view of the above case report and few case reports published before there is uncertainty surrounding the terminology of "mesiodens" in these types of cases, so we suggest a minor alteration in the definition of "mesiodens". The definition of mesiodens should therefore be altered to "Supernumerary tooth positioned in anterior maxilla characteristically *but not always* between permanent central incisors".

The authors suggest a new terminology" Mesiodensoid" (Mesiodens-like) for such unique condition.

The literature review of the hypo-hyperdontia in anterior mandibular arch does not reveal any significant systemic abnormality/syndrome. This suggests that these cases are sporadic and maybe due to a specific gene mutation that may be enabling the hypodontia of normal dentition and encouraging odontogenesis of the supernumerary tooth. This relatively harmless condition can provide to be boon to genetics research as there is a unique subset of both the supernumerary tooth and hypodontia co-existing in the same individual.

CONCLUSION

The above article focuses on the diagnosis and management of an innocuous looking "mesiodens". Greater the delay in diagnosis, the more complicated is the management of "mesiodens". Mandibular mesiodens is an uncommon occurrence and its association with agenesis of mandibular incisors is an even rarer event. This condition should be managed with a multi-disciplinary team approach involving Oral & Maxillofacial Radiologist, Oral & Maxillofacial Surgeon, Orthodontist, Prosthodontist and the General Dentist acting as the quarterback.

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