

CASE REPORT

Solitary Maxillary Central Incisor along with Mesiodens—Report of a Rare Case with Literature Review

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ABSTRACT

Dental anomalies of tooth number in development of the permanent dentition are quite common than the primary dentition. However, the combined occurrence of hypodontia and hyperdontia is a rare phenomenon, especially in the same dental arch, congenital absence of maxillary central incisor is still the rarest entity. The purpose of this report is to describe a rare case of a solitary maxillary permanent central incisor in association with labially erupted mesiodens.

Keywords: Agenesis, Mesiodens, Solitary maxillary central incisor.

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INTRODUCTION

Agenesis of one or more teeth in primary or permanent dentition is known as hypodontia, while, hyperdontia is a condition of having extra to the normal complement of teeth. Hypodontia and hyperdontia are two extremes in the development of the dentitions. The occurrence of both these together in the same individual is a form of mixed numeric variation. Although, the existing literature shows exclusively either hyperdontia or hypodontia, however, only a few studies have accounted for the incidence of both the anomalies.¹ Gibson adopted the term “hypo-hyperdontia” to describe the occurrence in the same individual.² Hypo-hyperdontia may affect primary or permanent dentition and maxilla or mandible. The frequency of hypodontia in permanent dentition ranges from 2 to 10%, and it can also be associated with some other anomalies.³ The prevalence of hyperdontia is reported to be between 0.15% and 3.9% while the reported

prevalence of hypo-hyperdontia is 0.002 to 3.1% with no gender predilection.⁴

The etiology of hypo-hyperdontia remains masked and it is not evidently documented in the literature. It is inconclusive whether a specific gene or an enzyme defect has an effective role in the development of this condition. Although genetic and environmental factors have been proposed to explain these anomalies in isolation, disturbances in migration, proliferation, and differentiation of neural crest cells and interactions between the epithelial and mesenchymal cells during the initiation of odontogenesis have been suggested to give rise to such a condition.⁵ Excluding third molars, the most affected teeth from hypodontia are maxillary lateral incisors, mandibular and maxillary second premolars in previous studies. Absence of maxillary central incisors, maxillary and mandibular first molars and canines are very rare to be seen.⁶ Supernumerary teeth can be present in both the primary and the permanent dentitions. However, they are almost five times more frequently observed in permanent dentition.⁷⁻⁹ Supernumerary teeth mostly occur in the maxilla (80 to 90%), and 50% of them are found in the anterior region.^{10,11} Most commonly seen is the mesiodens which is majorly located in the maxillary central incisor region, having an overall prevalence between 0.15% and 1.9%.⁷⁻⁹ The presence of mesiodens has the capacity to affect patient esthetics and occlusion by the altering natural eruption path of permanent incisors. The etiology of mesiodens can be attributed to any intrusion injuries to deciduous teeth, genetic predilection, and hyperactivity of the dental lamina.⁹⁻¹² The most common tooth morphology of mesiodens is characterized by a conical crown and short root. Mesiodens having completely formed roots often erupt into the oral cavity.¹³

Agenesis of one of the maxillary central incisor, presenting in the form of solitary maxillary central incisor is very rare. A similar condition is solitary median maxillary central incisor (SMMCI) syndrome, which involves mutations in Sonic hedgehog gene. It involves midline structures of the face, emerges in the midline, occurs in both primary and permanent dentitions. Patients with SMMCI presents, short philtrum, arched upper lip, narrow palate, absent intermaxillary sutures, absent upper labial frenum, and incisive papilla.¹⁴

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Early diagnosis of SMMCI is necessary as it might be associated with other syndromes and anomalies. Unlike SMMCI, solitary maxillary central incisor presents a common shape, with distinct mesial and distal thirds of the facial surface, and is located lateral to the midline (i.e., left or right).¹⁵ It may also not occur in both dentitions.

Additionally, the incisive papilla and upper labial frenum are present.¹⁶

The purpose of this manuscript is to report a very rare case of a child with only one maxillary permanent central incisor along with the presence of mesiodens at the place of missing counterpart central incisor. Correct diagnosis of SMMCI should be made in a child as if detected early can be referred for genetic counseling.

CASE REPORT

A 15-year-old girl reported to the Department of Pedodontics and Preventive Dentistry, College of Dental Sciences, Davangere, Karnataka, India with a chief

complaint of the presence of a small tooth in upper front tooth region. She was a normal healthy child with non-contributory medical history. The mother gave a history of normal delivery and a child was born to non-consanguineous parents. The family history reveals sibling with no such occurrence. Parents gave no history of trauma and normally erupted primary teeth. Intraoral examination showed the presence of permanent dentition with proclined left maxillary central incisor with mesiodens in place of right maxillary central incisor with right maxillary central incisor clinically missing (Figs 1 to 3). Anterior incisor space was reduced due to mesial drift of erupted permanent left central incisor and distopallatally rotated right maxillary lateral incisor. There was the presence of angle's class II molar relation bilaterally and generalized mild fluorosis was noted. An intraoral radiograph was advised which revealed the presence of mesiodens in place of a right maxillary central incisor with congenitally missing permanent right maxillary central incisor (Fig. 4). The radiograph indicated the pres-



Fig. 1: Preoperative frontal view showing single left central incisor and missing right central incisor



Fig. 2: Preoperative maxillary occlusal view



Fig. 3: Maxillary cast depicting conical mesiodens



Fig. 4: Intraoral periapical radiograph showing conical shaped mesiodens in place of missing right central incisor



Fig. 5: Postoperative frontal view depicting esthetic composite build up of mesiodens



Fig. 6: Postoperative maxillary occlusal view



Fig. 7: 18 months follow-up frontal view

ence of well-formed, labially placed mesiodens, with its long axis vertical in the alveolus, with a conical crown and long and straight root. The mesiodens had a flat surface with a mesiodistal width of 5.5 mm. Considering the age of the patient, clinical and radiographic findings, it was decided to carry out aesthetic restoration of mesiodens with anterior composites (Figs 5 and 6). The patient was recalled for follow up after every 6 months. At 18 months follow up satisfactory acceptable esthetics was seen and the patient was happy with the treatment (Fig. 7).

DISCUSSION

Anterior maxillary teeth are one of the most important features contributing to the aesthetics of an individual. Missing anterior teeth can cause psychological stress in children, during their growing years and can impede their behavioral and personality development.

The present case report revealed solitary maxillary left central incisor in permanent dentition with the presence of labially placed mesiodens in the space of missing right central incisor. This case is unique as it shows the

presence of supernumerary tooth in association with unilateral agenesis of the permanent maxillary central incisor, which is very rare. Etiologies proposed for hypodontia include any failure of tooth bud proliferation from dental lamina due to trauma, infections like rubella, osteomyelitis or use of drugs like thalidomide, genetic factors like mutation of muscle segment homeobox (MSX1) and paired box genes (PAX9), chemotherapy at very young age and presence of syndromes like ectodermal dysplasia and nonsyndromic conditions like cleft lip alveolus with or without cleft palate. Tooth agenesis is probably caused by various independent defective genes either acting alone or coupled with others to produce a specific phenotype.³

A similar condition SMMCI syndrome (phenotype) is a unique developmental abnormality, arising from an unknown events occurring between the 35th and 38th days in utero, and involving midline structures of the head including the cranial bones, the maxilla and its associated dentition (specifically the central incisor tooth germs), the nasal airways (choanal atresia, midnasal stenosis or congenital pyriform aperture stenosis), and sometimes the brain (holoprosencephaly), along with other midline structures of the body.¹⁷ The characteristics of tooth present in this syndrome are: solitary occurring central incisor in the maxilla present precisely in the midline of maxillary alveolus which might have unusual crown form. Hence, the acronym SMMCI syndrome. The circumstances where only one central incisor tooth is present and is not SMMCI includes failure of normally developing incisor to proceed beyond cellular development stage where remaining normal tooth develops to one side of midline, but may erupt in or near the midline; traumatic loss of one incisor; mesiodens of conical form erupting in midline but developing to one or other side of midline in permanent dentition only.¹⁷ Patients with SMMCI did not present a distinct philtrum, maxillary

labial frenulum, or incisive papilla. The fact that the tooth was not exactly in the midline, and the presence of the incisive papilla, philtrum, and the maxillary labial frenulum confirmed the diagnosis of agenesis of the right permanent maxillary central incisor. Knowledge of the clinical features of these two entities is crucial because the presence of an SMMCI can be linked to several important systemic manifestations and can be passed on to the patient's offspring. Unfortunately, most dentists treat cases of SMMCI as agenesis. These patients may present undiagnosed malformations and substantial mutations in their genome. Since both conditions occur in the anterior region of the oral cavity leading to poor esthetics, the dentist is often the first professional sought by patients and should, therefore, be able to make the differential diagnosis between an SMMCI and agenesis of the maxillary central incisor. In cases of diagnosis of an SMMCI, the patient should be referred for a thorough medical evaluation, including neurological, cardiac, endocrine, and genetic assessment.¹⁸ In our case, all the syndromic features were ruled out and diagnosed as nonsyndromic. Table 1 shows the list of cases of a solitary maxillary central incisor in the literature till date.¹⁹⁻⁵⁷

The occurrence of the supernumerary tooth in the maxillary anterior segment is a common entity. Primosch¹⁰ described two types of mesiodens as supplemental and rudimentary. Supplemental mesiodens resemble normal tooth and are also named as incisoriform. Rudimentary mesiodens can be categorized into three types based on their morphology as conical, tuberculate and molariform. Conical mesiodens are generally solitary, have completely formed roots and are commonly seen palatal to maxillary central incisors.⁴ The patient described in the current report too exhibited conically shaped mesiodens with completely formed root but was seen in a labial position which is a deviation from its palatal occurrence. About 90% of the mesiodens reported are situated palatal to the associated permanent tooth. Very rarely a mesiodens is encountered on the labial aspect of the permanent tooth and in 28 to 63% of cases, mesiodens is noted to cause an ectopic eruption, displacement or rotation of a permanent central incisor.¹⁷ In a situation such as this, a mesiodens in a favorable position could be considered to replace a permanent missing tooth. Most literature available describes the need for early recognition of the mesiodens and its extraction, to avoid any displacement, developmental anomalies or delayed eruption of the associated permanent tooth. Early extraction of a supernumerary or mesiodens in the mixed dentition is recommended, to allow for optimal yet minimal treatment for the permanent successor. However, in the

Table 1: Summary of reported solitary maxillary central incisor in the literature¹⁹⁻⁵⁷

Sl. No.	Author/year	Syndromic	Non syndromic
1.	Rappaport EB, 1977		Yes
2.	Wesley RK, 1978		Yes
3.	Boudailliez B, 1983	Yes	
4.	Marechaux SC, 1986		Yes
5.	Liberfarb RM, 1987		Yes
6.	Suss J, 1990	Yes	
7.	Miura M, 1993		Yes
8.	Kocsis SG, 1994	Yes	
9.	Roger K. Hall, 1997	Yes	
10.	M. Harrison, 1997	Yes	
11.	F. S. Lo, 1998	Yes	
12.	Triona McNamara, 1999	Yes	
13.	Parentin F, 2003	Yes	
14.	Tubbs RS, 2004	Yes	
15.	Hehr U, 2004	Yes	
16.	Garavelli L, 2004	Yes	
17.	Yang HC, 2005	Yes	
18.	Talmant J, 2006		Yes
19.	S. Y. Cho, 2006		Yes
20.	Garcia de Paula e Silva FW, 2007	Yes	
21.	Jinda Iertsirivorakul, 2008	Yes	
22.	Tabatabaie F, 2008		Yes
23.	Michele Bolan, 2009	Yes	
24.	Elizabete da Silva Viana, 2010	Yes	
25.	Kate blackmore, 2010	Yes	
26.	Jesus Gimento hernandaz , 2010	Yes	
27.	Catania P, 2010	Yes	
28.	Michele Bolan, 2010		Yes
29.	Ashok Utreja, 2011	Yes	
30.	Gurunathan D, 2011		Yes
31.	F. S. Van dijk, 2011	Yes	
32.	Da silva viana, 2011		Yes
33.	Nanni L, 2011	Yes	
34.	Kjaer, 2012		Yes
35.	Ahmet Ercan Sekerci, 2012	Yes	
36.	G. Shilpa, 2012		Yes
37.	N. N. Lygidakis, 2013	Yes	
38.	Delforge A, 2013		Yes
39.	Lutz Günther, 2014	Yes	
40.	Fabrizio Kitazono de Carvalho, 2014		Yes
41.	Holla G, 2014	Yes	
42.	Deepak Chandrasekaran, 2015		Yes
43.	Simon Poelmans, 2015	Yes	
44.	Sara Yang, 2016	Yes	
45.	Ilhan O, 2018	Yes	

event of a malformed, impacted permanent successor, all efforts should be taken to analyze and identify the path of eruption and location of the impacted mesiodens relative to the adjacent structures, and development of an appropriate treatment plan evolved to preserve and guide it to the desired position. Once a mesiodens have been diagnosed, the clinician must focus on early treatment to prevent further sequelae.¹⁸ This paper is unique

in that a very rarely seen labially placed mesiodens was detected in a favorable position and was used to replace a missing central incisor providing the patient with satisfactory esthetics.

CONCLUSION

The occurrence of both hypodontia (agenesis of unilateral permanent maxillary central incisors) and supernumerary tooth in the maxillary dental arch is an uncommon and extremely rare dental phenomenon. A careful history, clinical examination with detailed radiographic evaluation enhances the diagnosis ultimately helping to provide an appropriate treatment plan.

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