

REVIEW ARTICLE



Concomitant mandibular hypo-hyperdontia: Report of two rarest cases with the literature review

N. B. Nagaveni, Meghna Bajaj, Kirthiga Muthusamy, P. Poornima, Suryakanth M. Pai, V. V. Subba Reddy

Department of Pedodontics and Preventive Dentistry, College of Dental Sciences, Davangere, Karnataka, India

Correspondence

Dr. N.B. Nagaveni, Department of Pedodontics and Preventive Dentistry, College of Dental Sciences, Davangere, Karnataka, India. Email: nagavenianurag@gmail.com

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Abstract

Concomitant occurrence of both hypodontia (congenital tooth agenesis) and hyperdontia (supernumerary tooth) in the same dental arch is an extremely rare dental anomaly. Literature search shows very few cases of this anomalous condition with all cases depicting the unilateral presence of supernumerary tooth. Therefore, the intention of the current article is to report two cases of concomitant occurrence of mandibular both hypo-hyperdontia. In that one case exhibited bilateral occurrence of mesiodens teeth in the midline of mandible with associated agenesis of permanent both central incisors and taurodontism in permanent molars, which is not published so far. The article also provides comprehensive literature review on this rarest clinical entity.

Keywords: Mandibular mesiodens, supernumerary tooth, tooth agenesis

Introduction

Thorough clinical knowledge pertaining to the tooth development and chronology of eruption is necessary to understand the occurrence of various dental anomalies. Supernumerary tooth (hyperdontia) is an extra tooth compared with the normal tooth number count and these can develop as single, multiple, can be seen unilateral or bilateral and be found in either maxillary arch or mandibular or both.^[1]

The frequency of occurrence of these teeth is 8.2-10 times more in the maxilla compared to mandible. In maxilla, the pre-maxillary region involving the incisor area (90%) is the frequently affected part. Other regions seen with a supernumerary tooth in decreasing order are distomolars (mandibular or maxillary fourth molars), premolars and lateral incisors.^[2] The existence of extra teeth in the mandibular arch is extremely rare and very few cases have been reported [Table 1].^[3-27]

Missing teeth due to tooth agenesis also known as hypodontia is one more developmental defect in the normal number of teeth and most commonly involving the permanent teeth in children.^[28] Congenital agenesis of mandibular central incisors is a rare finding, and few cases have been reported as given in Table 2.^[7-34]

The term "concomitant hypo-hyperdontia (CHH)" is used to state the occurrence of both different clinical entities within the

same patient. This condition is also called as "oligopleiodontia" as suggested by Nathanail.^[35] Later, Gibson^[27] used the new term only "hypohyperdontia" to overcome the familiarity with the term "concomitant."

The reported prevalence of hypohyperdontia is between 0.002% and 3.1%, and most of these were of CHH involving both the arches.^[27] Males were more commonly affected with this condition. However, there is no particular gender predominance associated with this condition. This dental numerical variation has been reported more commonly in a permanent dentition compared with the primary dentition.^[27]

The combination of both supernumerary teeth and agenesis of permanent teeth i.e. hypodontia in combination with hyperdontia (hypohyperdontia) in the mandibular arch is a rarest entity. Only countable number of cases have been reported showing the combination of both hypodontia (agenesis of central incisor) and hyperdontia (midline mesiodens) involving the mandibular arch as given in Table 3.^[19,28,33-40] Hence, the present article aims to describe two cases of mandibular midline supernumerary teeth with agenesis of both permanent central incisors. One case exhibited bilateral mesiodens along with agenesis of permanent both central incisors and taurodontism affecting permanent molars, which are not reported so far according to author's best knowledge.

Table 1: Reported cases of mandibular mesiodens in the literature

Author	Year	Cases reported
De Jonge ^[3]	1965	Mandibular mesiodens
De Jonge ^[4]	1966	Partially erupted mandibular mesiodens
Boer ^[5]	1968	Partially erupted mandibular mesiodens
Furman and Williams ^[6]	1970	Mandibular lateral incisor
Spyropoulos <i>et al.</i> ^[7]	1979	Mandibular incisor
Ranta ^[8]	1983	Supplemental 72, 32
Pilley ^[9]	1989	Mandibular mesiodens
Macpherson ^[10]	1991	Supernumerary teeth in 83, 82, 72 region
Peyrano and Zmener ^[11]	1995	Mandibular mesiodens
Chow and O'Donnell ^[12]	1997	Supernumerary teeth in 32 and 42 region
Tanaka <i>et al.</i> ^[13]	1998	Bilateral mandibular mesiodens
Heathcote ^[14]	1999	Bilateral mandibular mesiodens
Sharma ^[15]	2001	Mandibular mesiodens
Cassia <i>et al.</i> ^[16]	2004	Mandibular mesiodens
Oncag <i>et al.</i> ^[17]	2005	Mandibular mesiodens
Alencar <i>et al.</i> ^[18]	2005	Mandibular mesiodens
Yokose <i>et al.</i> ^[19]	2006	Two cases: Mandibular mesiodens
Cho ^[20]	2006	Two cases: Supplemental mandibular mesiodens
Das <i>et al.</i> ^[21]	2006	Partial anodontia and mandibular mesiodens
Cho ^[22]	2006	Mandibular mesiodens
Zengin <i>et al.</i> ^[23]	2007	Mandibular mesiodens
Schmuckli <i>et al.</i> ^[24]	2010	Mandibular mesiodens
Nagaveni <i>et al.</i> ^[25]	2010	Mandibular mesiodens
Bargale and Kiran ^[26]	2011	Mandibular mesiodens
Naganahalli <i>et al.</i> ^[27]	2013	Supplemental mesiodens

Case Reports

Case report 1

A 9-year-old male patient accompanied by his father, visited to the Department of Pedodontics and Preventive Dentistry complaining of decayed tooth in lower left back region and grossly decayed tooth in upper right and left back region of jaw along with loosening of teeth in lower front region of jaw, with no history of pain associated with any of the mentioned findings. Medical history and family history were unremarkable. There was no history of consanguineous marriage. History of any trauma, infections or systemic disorders was not found. Intra-oral examination revealed that the patient was in mixed dentition period. Oral hygiene was adequate and there were over retained deciduous mandibular central incisors (FDI 71 and 81) [Figure 1] and mandibular permanent both central incisors (FDI 31 and 41) appeared clinically missing [Figure 2].

Table 2: Documented cases of agenesis of mandibular central incisors

Author/year	Age (in years)/gender	Tooth	Other significant dental findings
Low (1977) ^[28]	7 year/Male	31, 41	
Gibson (1979) ^[29]	6 year/Female	31, 41	-
Symons (1992) ^[30]	8/Male	31, 41	Supernumerary teeth in 11 and 13 region
Das (2006) ^[18]	8 year/Female	31, 41	-
Anthonappa <i>et al.</i> (2008) ^[31]	7/Male 11/Male 12/Female	31, 41 32 31, 41	Two supernumerary teeth in 11, 21 area Supplemental 15 Supplemental 23
Nagaveni and Umashankara (2009) ^[32]	12 year/Female 11 year/Male 9 year/Female 13 year/Female	31, 41 31, 41 31, 41 31, 41	- - - -
Marya <i>et al.</i> (2011) ^[33]	20 year/Male	31, 41	Agenesis of 18, 28 and 48
Nagaveni <i>et al.</i> (2011) ^[34]	12 year/Male	31, 41	Mandibular permanent left canine transmigration



Figure 1: Intraoral photograph showing over retained primary central incisors and lingually erupting conical shaped supernumerary teeth (arrows)

On radiographic examination, the panoramic radiograph revealed that conical shaped teeth were present without complete root formation and with no periapical changes; congenital agenesis of permanent right and left central incisors was also confirmed [Figure 3]. Taurodontism was evident in all four permanent first molars of the patient. Complete physical examination and clinical investigations were carried out to rule out any syndromic features.

Based on the appearance and size the teeth were diagnosed as bilateral mandibular supernumerary teeth. Other findings were normal considering the patient's age. Based on history, clinical and radiographic examinations diagnosis of concomitant

Table 3: Reported cases of CMHH

Case no.	Author/year	Age (in years)/gender	Hypodontia	Hyperdontia
1.	Low (1977) ^[28]	7/Male	31, 41	Unilateral mesiodens
2.	Gibson (1979) ^[29]	8/Female	31, 41	Unilateral mesiodens
2.	Das <i>et al.</i> (2006) ^[18]	8/Female	31, 41	Unilateral mesiodens
3.	Raghavan (2009) ^[35]	8.5/Female	31, 41	Unilateral mesiodens
4.	Nuvvula <i>et al.</i> (2010) ^[36]	15/Female	31, 41	Unilateral mesiodens
5.	Nayak <i>et al.</i> (2010) ^[37]	15.5/Male	31, 41	Unilateral mesiodens
6.	Venkataraghavan <i>et al.</i> (2011) ^[38]	9/Female	31, 41	Unilateral mesiodens
7.	Verma <i>et al.</i> (2012) ^[39]	9/Male	31, 41	Unilateral mesiodens
8.	Marya <i>et al.</i> (2012) ^[33]	20/Male	31, 41	Unilateral mesiodens
9.	Nirmala <i>et al.</i> (2013) ^[40]	10.9/Male	41	Unilateral mesiodens
		9.2/Male	41	Unilateral mesiodens
		8.5/Female	41	Unilateral mesiodens
10.	Present authors (2014)	9/Male	31, 41	Bilateral mesiodens
		13/Female	31, 41	Unilateral mesiodens

CMHH: Concomitant mandibular hypo-hyperdontia

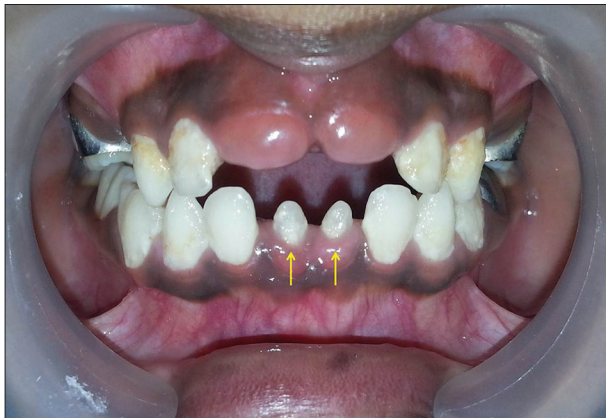


Figure 2: Photograph showing bilateral conical shaped mesiodens after extraction of primary incisors (arrows)

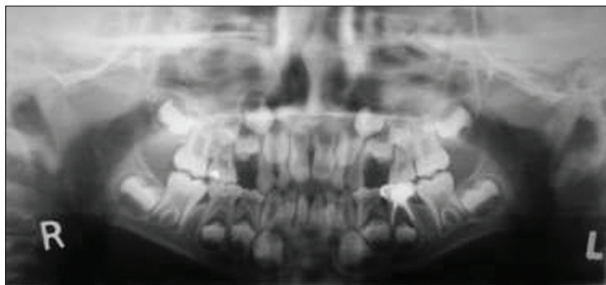


Figure 3: Orthopantomograph illustrating congenital agenesis of permanent mandibular both central incisors, bilateral mesiodens and taurodontism in permanent molars

mandibular hypo-hyperdontia was made. The patient’s parents were informed about the condition. Esthetics was given priority and was advised rehabilitation for the anterior teeth with composite resin once it erupts completely.

Complete oral prophylaxis along with extraction of over retained mandibular deciduous central incisors (FDI 1 and 81) was done in first appointment and also extraction of 54 and 64 (FDI tooth notation) was done in next appointment followed by space maintainer; pulpectomy, followed by stainless steel crown in relation to 75 was done in consecutive appointments and the patient has been kept under regular follow-up.

Case report 2

A 13-year-old female patient visited to the Department of College of Dental Sciences, with the chief complaint of labially placed upper front teeth. The medical history was not significant, and the general health was found normal. On intra-oral examination, patient exhibited complete permanent dentition (excluding second and third molars), with the presence of two supernumerary teeth in the mandibular anterior region exactly in the midline. On careful examination, we also noticed the absence of permanent mandibular central incisors [Figure 4]. The Orthopantomograph showed the agenesis of mandibular two central incisors (both right and left) and the presence of a supernumerary tooth in the midline that was of conical shape with fully developed root [Figure 5]. The patient was advised oral prophylaxis and orthodontic treatment for the anterior teeth proclination. As the patient did not show any concern for the mandibular mesiodens, no treatment was done.

Discussion

According to the detailed survey done by Gibson, in 1979,^[29] hypo-hyperdontia can be divided into pre-maxillary, maxillary, mandibular and bi-maxillary hypo-hyperdontia based on the site of occurrence. Among these, co-existence of hypo-hyperdontia in the mandibular anterior region is rarely seen clinical phenomenon. This is most commonly observed in the



Figure 4: Photograph showing conical shaped mesiodens with missing permanent both central incisors

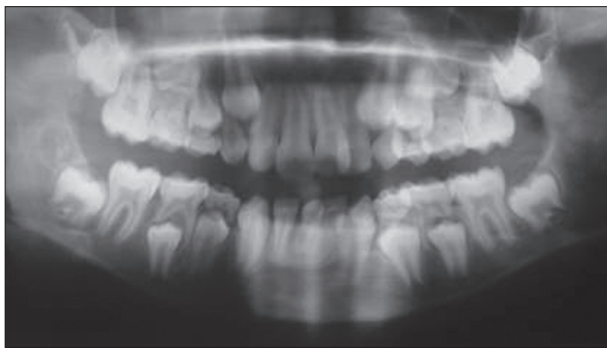


Figure 5: Orthopantomograph showing mesiodens in the midline of mandible with agenesis of both central incisors

permanent dentition than the primary dentition.^[29] The first report on this rarity was given by Low in 1977.^[28] After this, only eight clinical reports are available in the literature. All cases are reported in the mixed dentition except Marya *et al.*^[33] case, which is reported in a permanent dentition. Recently, Nirmala *et al.*^[40] reported three cases of CHH in Indian patients. Our two cases found in mixed dentition. However, our first case exhibited an incomplete root formation in the mesiodentes. This finding is also reported in a case published by Das *et al.*^[29] This case is unique as it shows bilateral presence of supernumerary tooth in association with bilateral agenesis of permanent central incisors and taurodontism involving all permanent molars.

There is no exact etiology for CHH and it is not clearly mentioned in the literature. However, environmental and genetic factors have been suspected to explain the occurrence of these anomalies.^[31] A recent study from Poland^[38] suggests that CHH is a rare phenomenon in association with predominant hypodontia. In addition, CHH has been found in patients with Dubowitz syndrome, Ellis–van Creveld syndrome, Down syndrome, G/BBB syndrome, Marfan syndrome, cleft lip and palate and fucosidosis.^[18,20] However, all published cases on the hypo-hyperdontia involving mandibular anterior region including the present cases does not show any significant

systemic abnormality or syndrome.^[19,28,36-40] This suggests that these cases are sporadic and may be due to a specific gene mutation that may be enabling the hypodontia of novel dentition and encouraging odontogenesis of the supernumerary teeth.

Occurrence of supernumerary tooth in the mandibular anterior segment is a rare entity. Primosch^[41] described two types of mesiodens as supplemental and rudimentary. Supplemental mesiodens resemble normal tooth and are also named as incisiform. Rudimentary mesiodens are again categorized as three types based on morphology as conical, tuberculate and molariform. Conical mesiodens are generally solitary, have completely formed roots and are usually seen palatal to maxillary central incisors. However, Nagaveni *et al.* in 2010, reported a unique case of multilobed mesiodens in the maxillary arch.^[42] Ribbons^[43] reported that the most of the supernumerary teeth seen in the mandibular anterior part are of supplemental type (2%). The same finding was also showed in the cases published by Marya *et al.*^[33] and Nirmala *et al.*^[40] However, the maximum frequency of occurrence of mesiodens in the mandibular front region reported belong to conical shape (56%). The patients described in the current report too exhibited conical shaped mesiodens. Moreover, the first case is different from other previous reported cases as it shows bilateral midline supernumerary teeth in association with missing both central incisors.

There is a tendency for dental agenesis to occur more unilaterally than bilaterally. Literature review shows few cases of bilateral agenesis of mandibular central incisors than unilateral cases [Table 2].^[28,34] In 2009, Nagaveni and Umashankara,^[32] reported four cases of bilateral agenesis of permanent mandibular central incisors in Indian patients.

Many authors have researched the association between taurodontism and hypodontia. Seow and Lai^[44] reported 34.8% of patients showed association between hypodontia and taurodontism. Whereas two Indian reports^[33,40] shows association between taurodontism and CHH. Similarly, our first case too, along with hypohyperdonita exhibited taurodontism of both permanent and primary molars. Contrarily, reports published by Marya *et al.*^[33] and Nuvvula *et al.*^[37] shows agenesis of maxillary and mandibular third molars. Unfortunately, this finding cannot be compared with the present as well as all other previous reports because the age in other published cases varies from young to old age (5-20 years). Apart from this, most recently double tooth and dens evaginatus have been reported in association with CHH.^[13,45]

The diagnosis of CHH is critical sometimes especially in a growing child. In the first case presented here, at first clinical examination, it was difficult to diagnose the condition as the tooth appeared entirely different in size and shape from normal appearance of mandibular central incisor. It created dilemma whether the tooth is a normal central incisor or a conical mesiodens with agenesis of permanent incisors. Careful clinical examination in addition to thorough radiographic evaluation helped us to diagnose finally as a mesiodens tooth not the central incisor. Moreover, literature evidence revealed no data on the

unusual appearance, i.e., microdontic or distinct conical shape of the mandibular central incisors, except for mesiodens tooth.

CHH in the mandibular anterior region can interfere with dental and facial aesthetics by causing a shift in the midline and hence requires correct treatment. The management of CHH is quite challenging as it requires a multidisciplinary team approach and treatment depends on the various stages of developing dentition and varies from individual to individual.^[19,28,36-40] Therefore, careful treatment planning is highly essential in order to deal with immediate, as well as long-term complications.

Although literature shows no standardized treatment protocols, in the current scenario, extraction of mesiodens and closing the space with fixed orthodontics, no treatment, extraction of mesiodens, followed by placing a Maryland bridge as short-term appliance, composite build up to restore as incisor and implants in the future are the suggested rehabilitation options in the literature. In the first case no treatment was carried out as mesiodentes were still in erupting stage.^[33,40] The possible treatment options were explained to the parents and patient is still under follow-up. In the second patient, parents did not show any concern for the mesiodens. Therefore, no treatment was carried out.

Conclusion

- The occurrence of both hypodontia (agenesis of bilateral permanent mandibular central incisors) and bilateral midline supernumerary tooth in the same dental arch especially in the mandibular anterior segment is an uncommon and extremely rare dental anomaly
- Careful clinical examination along with detailed radiographic evaluation enhances the diagnosis of this condition, which ultimately helps to provide appropriate treatment for an individual.

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