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Case Report

Bimaxillary Agenesis of Central Incisors Along with Impaction of Canine having Bifid Roots - A Rare Case Report

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Abstract

The occurrence of hypodontia, hyperdontia as well as impaction of teeth in the same individual is a rarely seen condition in dental practice. Thorough clinical and radiographical examination are essential in adequate treatment planning of such patients. This case report presents an unusual case of a nineteen-year-old male with the congenital agenesis of bimaxillary central incisors, with concomitant presence of a supernumerary tooth as well as impaction of canine having bifid roots. The condition has been reported to as concomitant hypo-hyperdontia. The erupted or unerupted tooth present in the anterior region of the jaws might cause aesthetic and/or functional problems. In this paper, the awareness about such condition; its diagnosis and treatment planning have been discussed in relation to the limited number of previously reported cases.

Keywords: Tooth Abnormalities; Hypodontia; Hypo-hyperdontia; Tooth Agenesis; Impaction

Introduction

Development of tooth need numerous genetic and molecular factors to determine tooth's type and performance [1]. Hypodontia and supernumerary teeth are contemplated as reciprocally exclusive conditions and are a result of interferences during tooth formation. Congenital absence of one or more teeth, primary or permanent, is referred to as hypodontia [2]. The prevalence of congenitally missing tooth varies from 0.2% to 16.2% in Asians [3]. The most frequently missing teeth are the third molars, upper lateral incisors, second premolars, and lower central incisors [4,5]. Agenesis of bilateral maxillary and mandibular central incisors is not documented and literature shows paucity of data pertaining to this anomaly. Therefore, the presentation is likely to be quite rare.

A supernumerary tooth is one that is additional to the normal series and can be found in any region of the dental arch. The most common supernumerary tooth is mesiodens that is typically present in maxilla between maxillary central incisors [1]. An erupted mandibular mesiodens in permanent dentition is a rare phenomenon.

On highly unusual occasions, hypodontia and supernumerary tooth can coexist within the same individual and the condition is referred to as 'concomitant hypodontia and hyperdontia (CHH)' [6], 'oligopleiodontia' [7], or 'hypo-hyperdontia' [8]. It is stated that CHH can be associated with various conditions like cleft lip/palate, cleidocranial dysplasia, G/BBB syndrome, Dubowitz syndrome, downs syndrome, marfan syndrome, ellisvan creveld syndrome and fucosidosis [9-11]. Non-syndromic CHH is sporadic and only countable number of cases have been reported (Table 1). It is therefore likely that the true incidence in the general population is significantly lower.

Author	Year	Gender	Age	Hypodontia	Hyperdontia	
					Maxilla	Mandible
Matsumoto., *et al*. [12]	2001	Female	8	UL5, LL2	UL2	
Sharma A [13]	2002	Female	12	UL3	Six supernumeraries	
Oliveira., *et al*. [14]	2002	Female	9	LL5, LR5	Mesiodens	
		Male	8	Gemination	Two mesiodens	
El-Bahannasawy., *et al*. [15]	2004	Female	4	URC	URD, UR5	
Patchett., *et al*. [16]	2006	Male	9	LL5, LR5	Mesiodens	
Anthonappa., *et al*. [17]	2008	Female	12	LL1, LR1	UL3	
		Male	9	UR5, LL5	UL1	
		Male	11	LL2	UR5	
		Male	5	LLA, LRA, LR2	Mesiodens	
		Male	7	LL1, LR1	Two mesiodens	
		Female	5	LLB, LL2	Mesiodens	
		Male	6	LL5, LR5	Mesiodens	
Sharma A [18]	2008	Male	7	UL5	Two mesiodens	
Varela., *et al*. [19	2009	Female	-	LL5	LR2	
		Male	-	LL5, LR5	Mesiodens	
		Male	-	LL5	UR2	
		Male	-	LL5, LR5		LL2
		Male	-	UL2	Mesiodens	
		Female	-	UL2	ULB	
		Female	-	UL2	URB, UR2	
Nuvvula., *et al*. [20]	2010	Female	15	LL1, LR1		Mesiodens
Nagaveni., *et al*. [21]	2011	Male	9	LL1, LR1		Two Mesiodens
		Female	13	LL1, LR1		Mesiodens
Marya., *et al*. [2]	2012	Male	20	LL1, LR1		Mesiodens
Verma., *et al*. [23]	2012	Male	15	LL1, LR2		Mesiodens
Nirmala., *et al*. [24]	2013	Female	13	UR4,UL3	Mesiodens	
Gupta., *et al*. [25]	2013	Male	11	LL2, LR2	Mesiodens	
Sawai M A [26]	2016	Female	26	LL1, LR1		Mesiodens
Tewari., *et al*. [27]	2017	Male	13	LL5	Two mesiodens	
		Female	10	LR5	Mesiodens	
Kariya., *et al*. [28]	2017	Male	10	LL1, LR1	Mesiodens	
Bharti., *et al*. [29]	2018	Male	13	LL1, LR1, LR7	UR1	
Sharma., *et al*. [30]	2018	Male	25	LL4	Four Supernumeraries	
Pandey N [31]	2019	Male	7	URA	Mesiodens	
Hlaing., *et al*. [32]	2020	Female	21	LL3, LR3	Two mesiodens	

Table 1: Published literature (from 2001-2020) on hypo-hyperdontia.

In addition to the above stated conditions, impaction of unilateral permanent canine in both maxilla and mandible along with retained multiple deciduous teeth makes this case report unique and a debatable entity. Incidentally, there is a gap in orthodontic literature with respect to the presence of bifid roots in impacted mandibular canine and have been reported by only Pallavi., *et al*. till date [33].

The aim of the present article is to report an extremely rare case of bilateral agenesis of both maxillary and mandibular central incisors, associated with presence of mandibular mesiodens and impacted maxillay canine and mandibular canine having bifid roots in a non-syndromic adult who was referred to the department as part of a routine orthodontic referral. Hence, this is a first of its kind case report of such an association and is discussed to provide a review to minimize the clinicians challenge in diagnosing such cases and thus helpful in providing a multidisciplinary approach in treating the patient.

Case Report

A 19 year old male presented to the department of orthodontics and dentofacial orthopaedics, for an orthodontic opinion for the chief complain of unesthetic appearance and missing teeth in both upper and lower arches. Prior and current medical and family history were non-contributory. No extraoral abnormalities on physical examination were elicited (Figure 1).

Figure 1: Extra-oral photographs.

The lips were competent with normal vertical facial proportions. An assessment of the temporomandibular joint was unremarkable. There were no other signs or symptoms suggesting a syndrome association. Intraoral examination revealed that there were presence of multiple retained deciduous teeth, i.e., 51,61,63,81,83 and absence of permanent both maxillary and mandibular central incisors, maxillary left canine and mandibular right canine, and maxillary and mandibular left third molars (Figure 2). On clinical examination, no bulge could be palpated or seen for any missing permanent teeth. In addition, there was presence of a conical supernumerary tooth in the midline of the mandibular arch. All retained deciduous teeth were attrited and fractured, amongst which tooth number 51 had blackish discoloration. In occlusion, the patient had a Class I molar relationship. There was presence of bilateral scissor bite in relation to teeth number 14 and 24. The patient's oral hygiene was fair with small amounts of plaque being present in the cervical margins of the posterior dentition. Small, opaque, paper white areas were scattered onto the surfaces of the teeth indicating dental fluorosis.

Figure 2: Intra-oral photographs.

A dental pantomogram revealed agenesis of bimaxillary and bilateral central incisors and presence of a conical supernumerary tooth in the midline of the mandibular arch. In addition, maxillary left canine and mandibular right canine were impacted. The roots of all retained deciduous teeth were resorbed but there was no obvious root resorption in relation to the permanent teeth. A root stump was evident in mandible between the supernumerary tooth and right lateral incisor. There were no reported cases of any missing teeth in the parents or other family members (Figure 3).

Figure 3: Orthopantomagram.

For diagnostic confirmation and to ascertain the position of impacted canines along with proper visualisation of root apices of the anterior teeth, a cone beam computed tomography (CBCT) in relation to both the arches was obtained. The CBCT confirmed the previous findings for the anterior superior and inferior region and revealed presence of root bifurcation in relation to impacted mandibular right canine (Figure 4 and 5).

Figure 4: CBCT images of maxillary arch.

Figure 5: CBCT images of mandibular arch.

Based on these physical and radiographic examination findings, a diagnosis of anomaly of tooth number was made. The patient presented partial anodontia with concomitant presence of a supernumerary tooth, thus characterizing hypo-hyperdontia of the permanent dentition along with bimaxillary canine impaction.

The patient was explained about the condition. Oral hygiene instruction, diet advice, and tooth brushing instruction were given. Given the reasonable amount of space present in both maxillary and mandibular arches, patient was advised to undergo treatment on a non-extraction basis. Extraction of all deciduous teeth was requested to allow disimpaction of the permanent canines. Levelling and alignment of the teeth through fixed orthodontic therapy was advised followed by prosthodontic rehabilitation of bimaxillary anterior teeth. Removal of supernumerary tooth was not advised due to its good periodontal health but the decision was made to regular monitor the root apex of the tooth.

Discussion

Concomitant hypo-hyperdontia is a rare condition. The reported prevalence of hypo-hyperdontia is between 0.002% and 3.1% [28]. Despite a strong genetic basis behind both hypodontia and supernumerary teeth, a study looking at nine patients with CHH found that none of the family members also suffered from CHH [20]. Whilst hypodontia is more common amongst females and supernumerary amongst males, CHH has been reported to affect both genders equally [2,17]. The aetiology of CHH is unknown. Genetic contribution, environmental influence, and a combination of both have been anticipated as the possible etiological factors of this condition [17,34]. According to Gibson, hypo-hyperdontia can be divided into pre-maxillary, maxillary, mandibular and bimaxillary hypo-hyperdontia based on the site of occurrence [8]. Among these, the present case is a unique entity of bimaxillary hypo-hyperdontia.

In humans, the most common developmental craniofacial anomaly is of the congenital agenesis of teeth. The etiology of this dental anomaly includes physical disruption of the dental lamina, reduced dental arch space, functional abnormalities of the dental epithelium and the failure of initiation of the underlying mesenchyme [35]. Mutations in specific genes such as PAX9, MSX1, AXIN2, EDA, WNT10A and LRP6 have been implicated as probable cause for hypodontia in novel dentition of non-syndromic individual [22,32,36]. Hypodontia of teeth in familial cases has also been reported to be associated with colorectal polyposis and cancer and also with ovarian pathology [35]. 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 [4,5]. Most of the studies done so far have not found congenitally missing maxillary central incisor or the prevalence is very low, making it the least possible tooth to display

agenesis. A study found congenitally missing unilateral maxillary central incisor in a sample of 1236 orthodontic individuals [37]. In the presented case report, the possibility of congenital agenesis of lateral incisors was ruled out by morphological and anatomical features of the respected teeth. The overall dimension of the erupted tooth was smaller than the expected dimensions of normal central incisors. The mesioincisal angle and distoincisal angle were also more rounded. Hence, the presented case is an extremely rare tooth agenesis. Although some anecdotal case reports [38] have dealt with the management of congenitally missing maxillary central incisor, an in-depth epidemiological study to identify the prevalence is not available. However, hypodontia of mandibular permanent centrals with the presence of midline supernumerary tooth in mandible was reported by many authors.

The prevalence of hyperdontia varies approximately from 0.1-3.8% [39]. 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. Due to various case reports of mesiodens in siblings, twins and families, the role of genetics have also been considered [39]. In the presented case report the possibility of a microdont central incisor associated with the congenital absence of the contralateral mandibular central incisor cannot be excluded. However, the distinct conical shape of the midline tooth that bears no resemblance to central incisor is the possible rationale why the tooth cannot be contemplated as central incisor.

The impacted canines are part of the group of position dental anomalies, while hypodontia or hyperdontia is part of the number dental anomalies. Certain discrete malpositions of the human canine tooth and agenesis of at least one tooth are abnormalities known to occur together, one of the situations being the association between agenesia with palatal displaced canine [40]. Overall, the incidence of impacted maxillary canine is suggested to be 0.8-2.8% [41]. But the incidence for mandibular canine impaction is at least 20 times lower than that of maxillary canine impaction [42]. Various factors including bifid root influence treatment planning in the management of impacted teeth. For instance, till date only a single case report [33] has been published reporting the presence of bifid root of an impacted canine while diagnosing the patient for orthodontic treatment. Thus, the present case report depicts the rare presence of bifid root of impacted mandibular canine which the orthodontists often overlooks. Orthodontists are trained to look at the position and inclination of the teeth rather than the number of roots. However, the number of roots determines the anchorage potential of a tooth by increasing the overall root surface area and may cause anchorage loss in cases where critical/maximum anchorage is required. Thus, the diagnosis of such a variation is of paramount importance [33].

The presence of deciduous and supernumerary teeth and congenital absence of other teeth in the esthetic zone may lead to social and psychological disturbances. Early diagnosis of the cases similar to the one described herein can reduce orthodontic complications and assist in treatment planning.

Conclusion

The association of multiple dental anomalies is rarely met, specially in a non-syndromic healthy individual. The scarcity of case reports of congenital agenesis of all four central incisors along with presence of mandibular mesiodens and canine impactions prevent the authors to do a note- worthy analysis. A careful intraoral screening of the patients and need for panoramic radiograph at an early stage should be mandatory when addressing these dental anomalies. This relatively harmless condition can provide to be boon to genetics research as well.

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