

CASE REPORT

Congenital bilateral agenesis of permanent mandibular incisors: case reports and literature review

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(Received 20 April 2009, revised manuscript accepted 27 September 2009)

Keywords

Central incisor,
congenital absence,
hypodontia,
tooth agenesis.

Abstract Hypodontia is the congenital absence of less than six teeth because of agenesis. The absence of teeth may be unilateral or bilateral. There are reports showing unilateral occurrence of permanent mandibular incisors. But agenesis of bilateral mandibular central incisors is not well documented in the literature and comprehensive review of literature shows paucity of data pertaining to this anomaly. As general dentist is the first member of the health team to diagnose and treat patients having unusual anomalies, the knowledge of congenital absence of permanent mandibular both central incisors is very essential to provide the most appropriate and comprehensive dental care and treatment possible. The aim of the present paper is to report, four cases of bilateral congenital missing of permanent mandibular central incisors and to review the literature regarding etiology, clinical implications and management.

Introduction

Hypodontia is the congenital absence of less than six teeth where as oligodontia refers to congenital lack of more than six teeth excluding third molars (Endo *et al.*, 2006a). Partial anodontia (hypodontia or oligodontia) involves one or more teeth and is a rather common condition (Endo *et al.*, 2006a). The most frequently occurring congenitally missing permanent teeth, excluding third molars, are the mandibular second premolar (3.4%) and the maxillary lateral incisor (2.2%) (Bäckman and Wahlin, 2001). The absence of teeth may be unilateral or bilateral. There are reports showing unilateral occurrence of permanent mandibular central incisors (Pfeiffer *et al.*, 1994; Newman and Newman, 1998). But agenesis of bilateral (both right and left) mandibular central incisors is not well documented and literature shows paucity of data pertaining to this anomaly. The first report of congenitally missing two mandibular incisors was given by Newman in 1967 (Newman, 1967). It has been reported that missing mandibular incisors is common in certain populations like Japanese, Korean and Chinese (Niswander and Sujaku, 1963; Davis, 1987).

Occurrence of this trait is very interesting to the orthodontist because of the potential development of malocclusion and its correction is challenging to obtain balanced occlusion, and to the geneticist as they contribute to one of the most widespread polymorphism in man. General dentist is not an exception to this. As a general practicing dentist is the first member of the health team to diagnose and treat patients having unusual anomalies, the knowledge of congenital absence of permanent mandibular both central incisors is very essential to provide the most appropriate and comprehensive dental care and treatment possible. The aim of the present article is to report, four cases of bilateral congenitally missing permanent mandibular central incisors and to review the literature citing its etiology, clinical implications and management.

Case reports

Case report 1

A 12-year-old Indian female patient reported to the Department of Pediatric dentistry, College of Dental Sciences, Davangere, complaining of space in the lower anterior teeth. Intraoral examination showed presence of retained deciduous mandibular central incisor in the midline having grade II mobility (Figure 1). The central incisor was distinguished from the lateral incisor by the distoincisor angle and incisal edge. The distoincisor angle is more rounded in lateral incisor compared to central incisor and

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the incisal edge is straight in central incisor whereas in lateral, it slopes toward the distal aspect of the tooth. Both permanent mandibular central incisors were missing clinically. Permanent maxillary centrals, laterals, left canine, left first premolar, first molars, mandibular laterals, canines, left first premolar and first molars were clinically present. Class I molar relation with absence of dental midline was evident. The child was born to non-consanguineous parents. The pregnancy and delivery were uneventful. There was no history of any severe systemic diseases, any history of trauma, or infections to the anterior region. Family history revealed no such finding in any members of the family. Panoramic examination revealed congenital agenesis of permanent mandibular both central incisors (Figure 2). As the deciduous central incisor was placed in the midline, there was a difficulty in diagnosing clinically, whether the central incisor is right or left. In the radiograph, the root of central incisor was placed left to the midline of mandible. So after the radiographic examination, the tooth was diagnosed as left central incisor. Because of the existing space the crown was tilted to the midline, resulting in difficulty in diagnosing the right or left incisor.



Figure 1 Intraoral photograph showing retained deciduous mandibular central incisor and missing permanent mandibular both central incisors.



Figure 2 Panoramic view of agenesia of permanent mandibular both central incisors.

Extraction of retained mobile deciduous central incisor was planned. To restore the missing incisors, fabrication of removable partial

denture was planned, as rigid fixed partial denture is contraindicated at this age. The other treatment option is the closing of the space by orthodontic treatment. Unfortunately patient was lost for further treatment.



Figure 3 Photograph showing retained deciduous mandibular central incisors.



Figure 4 Mandibular occlusal radiograph showing missing permanent mandibular both central incisor tooth buds. Permanent right canine is horizontally impacted.

Case report 2

An 11-year-old Indian male patient reported for oral prophylaxis. On clinical examination, retained deciduous mandibular both central incisors with no mobility were found (Figure 3). Permanent maxillary centrals, laterals, canines, mandibular laterals and left canine had already erupted. Patient had class III molar relation. Suspecting the congenital agenesia of permanent lower central incisors, mandibular occlusal radiograph was taken which confirmed the provisional diagnosis (Figure 4). Both permanent laterals were abnormally angulated with crown deviating laterally and roots deviating medially. Along with agenesia of centrals, horizontal impaction of right canine was also evident on the radiograph (Figure 4). Patient had been informed about the absence of permanent incisors. As the deciduous centrals were still firm without evidence of root resorption, no treatment was done at present. More over patient was also not willing for any

treatment. For this case, in the future, once the primary incisors undergo exfoliation, fabrication of removable or fixed partial prosthesis or orthodontic closure of the space could be done. Patient was kept under observation, but he did not turn up.



Figure 5 Photograph showing missing permanent mandibular both right and left central incisors. Note permanent maxillary centrals, laterals and mandibular laterals have already erupted. But primary mandibular right central incisor is still not exfoliated.



Figure 6 Periapical radiograph showing bilateral absence of permanent mandibular central incisors.

Case report No. 3

A 9-year-old Indian female patient reported, with a complaint of missing lower front tooth and space in the anterior region. Patient's mother gave a history of shedding of left primary anterior tooth 2 years back and after that, no permanent tooth erupted in that space. On intraoral examination, permanent mandibular left central incisor was missing (Figure 5). Primary mandibular right central incisor was retained with grade III mobility. Both permanent mandibular laterals and maxillary centrals and laterals had already erupted. There was no history of trauma to the anterior region. The teeth were in normal shape, size and color. Patient had class II molar relation. Suspecting the congenital absence of permanent left central incisor, a periapical radiograph was taken, which revealed the absence of both permanent central incisor tooth buds (Figure 6). Permanent lateral incisors were slightly angulated may be

because of the pressure exerted by erupting permanent canines on roots of lateral incisors. Patient's mother was informed about the condition and possible treatment options needed in the future. The mother was worried more about the esthetics, hence extraction of the mobile deciduous incisor and fabrication of removable partial denture was planned to restore missing teeth and esthetics.



Figure 7 Photograph of mandibular arch with midline spacing because of missing both central incisors.



Figure 8 Periapical radiograph showing absence of both (right and left) permanent mandibular central incisors.

Case report No. 4

A 13-year-old Indian female patient reported, complaining of spacing in lower anterior region and wanted to get it closed. Patient's mother gave a history of presence of four milk teeth and after their exfoliation; the permanent teeth had not erupted in their place. Intraoral examination revealed, complete permanent dentition with class II molar relation. Both permanent mandibular central incisors were missing clinically (Figure 7). Patient did not express features of any syndromes or systemic diseases. The remaining teeth were in normal shape, size and color and there were no relevant family history and history of trauma. Periapical radiograph showed the absence of

permanent mandibular both central incisors (Figure 8). Patient was advised to undergo orthodontic therapy for closure of the lower midline spacing. Unfortunately patient missed the appointment as she moved to another place.

Discussion

Etiology

Although the exact etiology of congenital agenesis of both central incisors is unknown, several factors like trauma, radiation, infection, metabolic disorders and idiopathic are the possible etiologic factors (Endo *et al.*, 2006a). Newman and Newman (1998) have given four main theories mainly for the cause of agenesis of incisors. Heredity or familial distribution is the primary cause. Second, anomalies in the development of the mandibular symphysis may affect the dental tissues forming the tooth buds of the lower incisors (Newman, 1977). Third, a reduction in the dentition regarded as nature's attempt to fit the shortened dental arches (an expression of the evolutionary trend) (Lavelle and Moore, 1973) and finally, localized inflammation or infections in the jaw and disturbance of the endocrine system destroying the tooth buds (Newman and Newman, 1998). It has also been reported that genes MSX1, TGFA and PAX9 interaction sometimes play a role in human tooth agenesis (Vieira *et al.*, 2004).

Class III malocclusion is sometimes associated with agenesis of mandibular incisors (Endo *et al.*, 2004; Endo *et al.*, 2006b). In all the four cases presented here, definite etiology was not found. The cause could be idiopathic in all these cases.

Very strong correlation exists between the agenesis of permanent teeth and agenesis of primary teeth (Meon, 1992; Davis and Darvell, 1993). In the study by Grahnen and Granath (1961), most of the cases with hypodontia in primary dentition showed the same condition in the permanent dentition. But no such finding was found in our four cases. In one study (Davis and Darvell, 1993), the four permanent mandibular incisors were missing but with the presence of four normal primary incisors. Pfeiffer *et al.*, (1994) have shown association of single mandibular incisor in a patient with del (18p) anomaly. Fukawa (1993) has reported a case with congenitally missing lower central incisor in twins.

Clinical implications

Mandibular incisor agenesis has a large effect on mandibular symphysis growth and morphology. Buschang *et al.*, (1992) demonstrated that, vertical and horizontal growth changes during childhood and puberty, were most pronounced in the upper half of the mandibular symphysis and tooth eruption plays a critical role in continuous growth of the mandibular symphysis, resulting in an increase

in the height of the mandibular body. Hence patients with absence of mandibular both central incisors, exhibit significantly smaller mandibular symphysis area than the normal patients. They have also reported that, the growth of alveolar bone is also associated with continuous eruption of the dentition (Buschang *et al.*, 1992). Thus the congenital absence of lower incisors can result in minimal volume of bone for the placement of end-osseous implants in locations favorable for subsequent restorations. Endo *et al.* (2007) have concluded from their study that, before planning/implementing orthodontic treatment on a patient with congenital missing incisors, some factors like retroclination of alveolar bone and reduced mandibular alveolar bone area should be taken into consideration, as these may affect the treatment outcome.

Some orthodontists (Kokich and Shapiro, 1984; Canut, 1996) say that congenital absence of both mandibular central incisors is advantageous, as the extraction of mandibular central incisors is sometimes considered as the treatment of choice in crowded class I malocclusion, especially when a preexisting tooth-size discrepancy (severe mandibular excess) prevents the achievement of an acceptable occlusion.

The other consequences of agenesis of both mandibular incisors are disturbance in tongue-lip pressure balance and lack of lingual support. Severe malocclusion usually class II Div I malocclusion is also seen with severe anterior deep bite and absence of dental midline or sometimes wide spacing in the anterior region exists resulting in unaesthetic appearance for a child (Endo *et al.*, 2007).

The other problem encountered with congenital absence of incisors is that, difficulty in identification of teeth. Because of the existing space resulting from missing teeth, the adjacent teeth move to this space, leading to difficulty in identification of incisors. Thus for correct diagnosis of teeth, radiographic examination is mandatory in order to see the exact position of the root.

From the four cases, it was also found that retained deciduous incisors exceeding normal exfoliation time or space in anterior region, justify the importance of radiographic examination in every patient with a retained deciduous teeth or abnormal spacing, for early diagnosis and early intervention of congenital missing of incisors.

Treatment modalities

Treatment strategies used in treating missing mandibular incisors include, various restorative and orthodontic procedures to improve aesthetics and function. Although any treatment could not be done in the four cases presented here, definite treatment includes multidisciplinary management consisting of various specialists like pediatric dentist, prosthodontist and orthodontist, and oral and

maxillofacial surgeons to restore aesthetics and function. From orthodontists' point of view, techniques for treating malocclusions stemming from agenesis of mandibular incisors vary by case and clinician (Newman and Newman, 1998). Usually their absence creates a diagnostic decision involving three options of treatment. In the adolescent period, one of the possible treatment procedures involves, obtaining a functionally adapted occlusion by protracting the mandibular canines and posterior teeth forward. A second treatment modality includes the creation of space and up righting and aligning the mandibular laterals and canines to receive a fixed prosthesis. Finally, the third method involves the removal of maxillary premolars or lateral incisors to balance tooth material resulting from the absence of mandibular incisors.

Restorative procedure involves fabrication of removable partial denture as an immediate and temporary treatment to restore the missing teeth and esthetics. After the growth completion, fabrication of fixed partial prosthesis is the other treatment modality, if malocclusion is not a major problem.

Considerable research supports, the efficacy of rehabilitating a completely or partially edentulous mandible using prosthesis supported by implants anchored in the anterior mandible (Adell, 1983; Guckes *et al.*, 1998). But the congenital absence of teeth can result in minimal volume of bone for the placement of endosseous implants in locations favorable for subsequent restorations. Also, craniofacial growth will necessitate remake and redesign of the prosthesis as growth occurs. But there are other reports showing good results with implants (Guckes *et al.*, 1991; Guckes *et al.*, 1997). If implant-supported prosthesis were shown to have positive effects on craniofacial growth, social development, self image, and food choice of a patient, their use in the anterior mandible might be routinely recommended in younger patients with bilateral absence of mandibular central incisors. With the advent of new designs in dental implants and their abutments, it is possible to consider replacing missing incisors with implant borne prosthesis (Guckes *et al.*, 1998).

Treatment in all the presented four cases includes multidisciplinary management to restore aesthetics and function. Initially, removable partial acrylic denture can be given in case one and three, as rigid fixed prosthesis is contraindicated at this age. As patients reach adolescence, conservative fixed prosthetic replacement of missing both central incisors, comprehensive orthodontic treatment to close space and advanced treatment strategy such as use of Osseo-integrated implants are the treatments of choice in all the cases. Thus it is the general dentist's responsibility to identify these patients for early referral to receive multidisciplinary treatment before any complications can occur.

Finally to conclude, agenesis of mandibular incisors can lead to compromised dental and facial aesthetics and therefore requires appropriate treatment (Endo *et al.*, 2007); however, due to the rarity of hypodontia of mandibular incisors, general dentist do not always have the necessary experience to embark upon the treatment of affected children. Careful treatment planning is important, because there is a need to deal with not only the immediate, but also the long-term adverse implications. Hence, multidisciplinary treatment planning which takes account of established and emerging techniques needs to be practiced. And also the subsequent development of different treatment options, that take account of growth and development of the dentition and of the compliance of the child can lead to a treatment plan that can produce pleasing interim results, which do not compromise any future treatment.

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