Case Report

Bilateral Agenesis of Permanent Mandibular Central Incisors: Two Familial Case Reports

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Abstract

Agenesis of one or more teeth is the most common anomaly of tooth development. Several factors like infection, trauma, metabolic disorders, radiation, environmental, genetic factors and idiopathic are possible etiological factors of congenital agenesis. Hypodontia usually requires comprehensive and complex treatments, ranging from single restorations to surgery and multiple restorations, associated with lifelong maintenance.

A 9 year-old female and a 9 year-old male patient have reported to the clinic and we have detected agenesis of bilateral mandibular incisors with an oligodontia and a hypodontia with familial transmission.

Although in many cases mentions oligodontia, congenitally missing bilateral mandibular central teeth cases has not been documented adequately. The purpose of this article is to present two cases of agenesis of bilateral mandibular incisors with hypodontia and oligodontia. Pediatric dentists must be capable of diagnosing because early diagnosis and guidance with regard to treatment are necessary for the problems that may affect the behavioral pattern and social life of these persons.

Keywords
Agenesis; Congenital missing; Hypodontia; Non-syndromic; Oligodontia

Introduction

Agenesis of one or more teeth is the most common anomaly of tooth development. Congenital tooth agenesis may be either hypodontia (agenesis of fewer than six teeth excluding third molars) or oligodontia (agenesis of more than six teeth excluding third molars). Oligodontia is a rare condition that can occur with genetic syndromes, or as a non-syndromic isolated familial trait, or as a sporadic finding [1].

Several factors like infection, trauma, metabolic disorders, radiation, environmental, genetic factors and idiopathic are possible etiological factors of congenital agenesis [2]. Genetic factors like MSX1 and PAX9 genes have an important role in the development of incisors. Disturbances in the interaction between these environmental and genetic factors are also main etiologic factors for congenital missing teeth [3].

The prevalence of hypodontia ranges from 1% in African Black and Australian Aborigines to 30% in Japanese people, and studies of have reported higher frequencies of hypodontia among females [4]. The prevalence of oligodontia in permanent dentition was reported as 0.14% in the literature [5].

The third molars are the most frequently missing teeth, but they do not need to be replaced; nevertheless, mandibular second premolars (2.8%), maxillary lateral incisors (1.6%), and maxillary second premolars and mandibular incisors (0.23%-0.08%) are the most frequently missing teeth that require some type of treatment [6].

Hypodontia usually requires comprehensive and complex treatments, ranging from single restorations to surgery and multiple restorations, associated with lifelong maintenance [7].
Several treatment solutions have been presented in the dental literature [8].

The necessary treatment generally depends on the pattern of tooth absence, the amount of residual spacing, the presence of malocclusion and patient attitudes. One of the most important key factor for the successful treatment of patients with hypodontia is the interdisciplinary intervention, which involves the close work of a committed team (general dental practitioner, pediatric dentist, orthodontist), where each member contributes with a different expertise to achieve optimal results for the patient [9].

Although in many cases mentions oligodontia, congenitally missing bilateral mandibular central teeth cases has not been documented adequately. And 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 review of literature paucity of data pertaining to this anomaly. The purpose of this article is to present two cases of agenesis of bilateral mandibular incisors with hypodontia and oligodontia.

**Case Report 1**

A 9-year-old female patient reported to the clinic with main complaint concerning about having spaces between the lower teeth (Figure 1). Physical examination was completely normal and the patient did not present any history of systemic disease, trauma or tooth extraction. She was born premature. On intraoral examination presence of retained deciduous mandibular central incisor in the midline having mobility Grade I is detected. In this patient; upper and lower second pre-molar, upper first molar, upper and lower second molar was also missing (Figures 2-4).

According to the patient's history; the patient, her siblings and father had congenitally missing bilateral mandibular centrals (Figures 4 and 6-9). According to the radiographic examination, # 54, 63, 64, 73, 74, 83 and 84 were extracted based on exfoliation times (Figure 4). We have restored # 65, 36, and 85 with composite resin. And we have restored the
retained deciduous mandibular central with composite resin esthetically (Figure 5).

Case Report 2

A 9 year-old male patient reported to the clinic with chief complaint concerning non-erupting lower anterior incisors (Figures 10-12). In general, physical examination was completely normal. Intraoral examination showed presence of retained left deciduous lower central incisor (Figure 10). According to the patient's history, the patient and his father had congenitally missing bilateral mandibular centrals (Figures 12-14). We have extracted # 71, 75, 84 based on exfoliation times (Figures 10, 12 and 15). We have restored # 53, 63, 73, 83, 16, 26, 36, 46, 11, 21, 22 with composite resin (Figures 16 and 17). And finally we have prepared partial removable prosthesis (Figure 17). We have consulted the patient to orthodontics for clinical follow-up.

Discussion

Several hypotheses are included for the etiology of oligodontia such as traumatic injury during tooth development, endocrine disturbances, infections, also radiation or chemotherapy during childhood cancer therapy.
Figure 10: Pre-op buccal aspect of the patient 2.

Figure 11: Mandibular occlusal aspect of the patient 2.

Figure 12: Orthopantomography of the patient 2.

Figure 13: Intraoral aspect of father of the patient 2.

Figure 14: Orthopantomography of father of the patient 2.

Figure 15: Maxillary occlusal aspect of the patient 2.
Furthermore, specific genes have been associated with tooth agenesis, particularly the MSX1, PAX9 and AXIN2 mutations [11].

Familial tooth agenesis can occur as an isolated anomaly or as part of a genetic syndrome [9] and is transmitted as an autosomal dominant, recessive, or X-linked condition [12]. MSX1 and PAX9 genes play a role in early tooth development [13]. PAX9 plays a role in the absence of third molars in some human populations [14] and mutations in MSX1 is related with nonsyndromic cleft lip with/without cleft palate [15] and autosomal dominant hypodontia. MSX1-associated oligodontia typically includes missing maxillary and mandibular second premolars and maxillary first premolar [16].

In familial oligodontia, the type of inheritance in majority of the families seems to be autosomal dominant with incomplete penetration and variable expressivity. An autosomal recessive model of inheritance is also possible mutations in transcription factors MSX1 and PAX9 have been identified in families with an autosomal dominant oligodontia. A new mutation has been detected that causes familial oligodontia is in the b-catenin binding protein AXIN2 [17].

Genetic factors like MSX1 and PAX9 are associated with the development of incisors. The main etiologic factor of congenital missing teeth is disturbances in the interaction between these factors [3].

The exact etiology of congenital agenesis of both central incisors is unknown but various factors like radiation, trauma, infection, metabolic disorders and idiopathic are the possible factors [18]. New man has given four theories mainly for the cause of agenesis of incisors. The primary cause is the heredity and second one is; disturbances of the development of the mandibular symphysis that may affect the tissues forming the tooth buds [19]. Thirdly, a reduction in the dentition to fit the shortened dental arches that is by attempt of nature [20] and finally, disturbance of the endocrine system and localized inflammation or infections in the jaw and affects the tooth buds [21].

Mandibular incisor agenesis has a large effect on growth and morphology of mandibular symphysis. It has demonstrated that, patients with absence of bilateral mandibular central incisors exhibit significantly smaller mandibular symphysis area than the healthy patients [22]. The other results of agenesis of bilateral mandibular incisors are lack of lingual support and disturbance in tongue-lip pressure balance and, malocclusion mostly Class II Div I malocclusion and severe anterior deep bite and absence of dental midline or wide spacing in the anterior region that results unaesthetic appearance for a child [23].

The adjacent teeth move to the space, leading to difficulty in identification of incisors. Radiographic examination is necessary in order to see the exact position of the root [2]. Hypodontia usually requires comprehensive and complex treatments, ranging from single restorations to surgery, and multiple restorations, associated with lifelong maintenance. The optimal therapy should include an interdisciplinary team approach, and rely on positive interaction between pediatric dentists, orthodontists, oral and maxillofacial surgeons and prostodontics [24]. The treatment purposes are to keep the remaining teeth, recover the masticatory function and aesthetics, speech improvement, and build the emotional and psychological well being [9].

The purpose of this study was to present two cases of bilateral agenesis of permanent mandibular central incisors with an oligodontia and a hypodontia with their treatments as it suggested in literature and their clinical follow-up regularly.

Conclusion

The congenital absence of teeth may occur as an isolated condition or may be associated with a systemic condition or syndrome. Clinical and radiographic examinations are the
best way for the clinicians may detect anomalies in primary and permanent dentition. When dental agenesis is diagnosed, treatment should provide the restoration of the masticatory function and aesthetics.

In summary, tooth agenesis causes a clinical and public health problem, because patients in these conditions may suffer a reduction in their masticatory capacity, malocclusions, phonoaudiological problems, and compromised esthetics. Pediatric dentists must be capable of diagnosing because early diagnosis and guidance with regard to treatment are necessary for the problems that may affect the behavioral pattern and social life of these persons. Careful treatment planning and current understanding of the condition with the genetic background are important to prevent esthetic, functional and psychological problems under the supervision of a multidisciplinary team.

References