Case Report

Permanent Canine Agenesis: A Case Report

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Abstract

Introduction: Congenital syndromic or non-syndromic absence of maxillary permanent canine teeth is a rare occurrence. This anomaly can adversely affect esthetics and masticatory function. Other dental anomalies that can be associated with tooth agenesis include tooth malposition, insufficient bone height, and periodontal disease. According to the statistics, the incidence of canine agenesis ranges from 0-4.7% with a prevalence rate of 3.0%. Non-syndromic canine agenesis combined with other types of tooth agenesis, like mandibular central incisor agenesis, has been occasionally described in the literature.

Case Presentation: This case report presents a nine-year-old male patient with three canine agenesis combined with both mandibular central incisors and unilateral maxillary lateral incisor agenesis.

Results: Tooth agenesis is a common dental anomaly causing significant esthetic impairment. Its proper management requires early detection and a multidisciplinary approach.

Key Words: Cuspid; Hypodontia; Dentition; Permanent

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Introduction

Approximately 60,000 to 20 million people worldwide suffer from impaired development of at least one of their teeth (1). The prevalence rate of congenital absence of permanent teeth among Asians ranges from 2.2% to 16.2% (2). Hypodontia refers to the absence of one or more teeth, which is a common dental anomaly. Moreover, according to previous studies, its prevalence is growing (1-3). Congenital absence of teeth has been observed in different syndromic and non-syndromic forms, and in most cases, it affects permanent teeth (4).

Canine agenesis refers to failure in formation of permanent canine teeth, which are considered to be the most stable teeth in humans. According to the Butler's theory in 1939, the mesial tooth of any group of dentition is the most stable (5). In 1975, Bailit divided the permanent dentition into stable and unstable teeth; accordingly, maxillary canine, maxillary central incisor, first premolar, and first molar teeth are in the stable group (6). Therefore, maxillary canine agenesis is uncommon. Moreover, maxillary canine agenesis is less common and mandibular canine agenesis is rare (5). In a systematic review conducted in 2020, it was stated that the global incidence of canine agenesis ranges from 0 to 4.7% with a prevalence rate of 3.0%. Results of the aforementioned study also indicated that canine agenesis is most prevalent among Asians;

furthermore, it was found that it has a higher prevalence in males and is mostly reported in the maxilla (7).
The purpose of this case report is to describe a case of canine agenesis.

Case Presentation

A nine-year-old male patient presented to a pediatric dental office in Ardabil, Iran in February 2021 for a routine dental examination. He was healthy with no history of systemic disease or any syndrome. Extraoral examination revealed no abnormalities of the skin, hair, or nails. Clinical examination revealed retained primary maxillary lateral incisors in the right side along with both lower central incisors. Lateral incisor microdontia was also detected in the left side of his maxilla. Obvious spacing was noted among teeth in the anterior maxilla and mandible, and all remaining anterior primary teeth had attrition (Figure 1). Radiographic examination showed two missing permanent teeth in the right side of the maxilla, canine and lateral incisor, and four other missing teeth in the mandible, including the right and left canine and both mandibular central incisors (Figure 2). There were no other dental abnormalities. Clinical examination of the occlusion showed an end-to-end molar relationship as well as 4 mm of overbite and 2 mm of overjet in the anterior teeth. No trauma history or early irradiation of tooth germ was recalled. The patient was the only child of the family and there were no noticeable anomalies in his familial dental history except maxillary lateral incisor microdontia in his mother.

Figure1. Intraoral photograph showing retained primary maxillary lateral incisors in the right side along with both mandibular central incisors

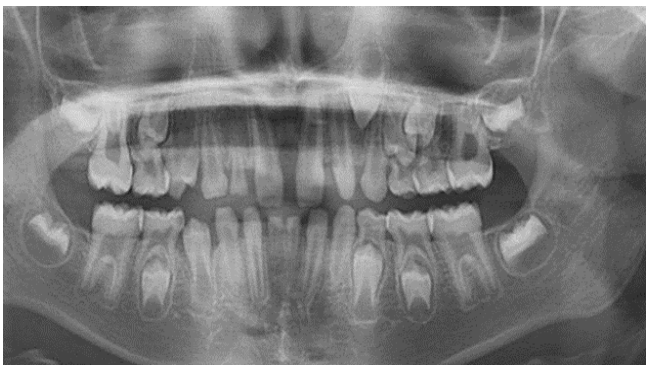

Figure 2. Panoramic radiograph showing congenitally missing maxillary permanent canines (right side) mandibular permanent canines (bilateral), and mandibular central incisors (bilateral)

Discussion

Hypodontia is the most prevalent maxillofacial malformation (8), and its prevalence varies based on gender, dentition, jaw, and race. Moreover, based on a previous study, the worldwide prevalence of hypodontia is high in Caucasians and low in Asians and Oceanians while canine agenesis is more prevalent in Asians and less prevalent in Africans and Europeans. Besides, canine agenesis is less common in the maxilla and rare in the mandible (7).
Amini et al, in 2012 conducted a study on 3,374 panoramic radiographs of people aged 10-20 years and found that the prevalence of hypodontia was 5.21%. They reported that one out of every 20 Iranian orthodontic patients might have some missing permanent teeth. An important finding of their study was that hypodontia was more prevalent in females and mostly affected the maxilla (9). Based on the results of a study performed in 2018, the incidence of dental developmental anomalies in permanent dentition was 20.2% in Ardabil, Iran (10). According to the terminal reduction theory, canines are considered as the key teeth of dentition and their missing is rare (11).
Hypodontia is accompanied by several syndromes, such as Van der Woude syndrome, ectodermal dysplasia, oral-facial-digital syndrome (type I), holoprosencephaly, Rieger syndrome, and cleft lip and palate (12). No etiological factors have been identified for

non-syndromic agenesis or selective tooth agenesis. However, there has been evidence in twins with mirror-like unilateral agenesis which proves its genetic origin (11). It appears that it has a dominant autosomal inheritance with variable expressivity. Other kinds of inheritance have also been reported (13). Other factors that may influence agenesis are dental trauma, irradiation, or surgical procedures.

In some studies, it has been emphasized that a combination of environmental and genetic factors affect agenesis, and it is suggested that missing posterior teeth are sporadic while anterior agenesis is genetic (2, 14). In 2017, Yamaguchi et al. used exome sequencing and showed that novel variants in previously reported causal genes appear to mainly contribute to tooth agenesis of the anterior region and suggested the association between FAM65A, NFATC3, and CDH23 or SMIA as genetic factors (15).

Tooth agenesis can adversely affect esthetics and masticatory function. In addition, speech development depends on normal dentition, and most importantly, tooth agenesis can emotionally affect adolescents (9). Other dental anomalies that can be associated with tooth agenesis are dental malposition, insufficient bone height, and periodontal disease (16). Unilateral canine agenesis is more frequent than bilateral agenesis and is usually accompanied by other dental anomalies, such as congenital absence of other teeth (17) which was also seen in the present case.

Unlike unilateral occurrence of permanent mandibular incisors, agenesis of bilateral mandibular central incisors has not been well documented in the literature. Knowledge about congenital absence of bilateral permanent mandibular central incisors and its treatment is highly important (18). In 2006, Endo et al. reported that Class III malocclusion is sometimes associated with agenesis of mandibular incisors. They noted that hypodontia results in counterclockwise rotation of the mandible that creates the appearance of prognathism. These changes are mostly due to soft tissue imbalance and must be considered in orthodontic treatments (19). However, in the present case, the etiology was unknown.

Continuous eruption of lower incisors causes significant growth changes in the upper part of the mandibular symphysis during childhood and puberty (18). Therefore, mandibular incisor agenesis has an important impact on the growth of mandibular symphysis. Another important consequence is a disturbance in the tongue-lip balance and lack of tongue support. In 2007, Endo et al. reported class II division I severe malocclusion due to this disturbance. It results in severe anterior deep bite, absence of dental midline, wide spacing in the anterior region, and unesthetic appearance (20). It should be mentioned that spacing and unaesthetic appearance were detected in the present case as well.

Tooth agenesis requires a complicated treatment plan; in most cases, orthodontic and prosthodontic consultations are needed. Timely extraction of primary teeth for space alignment followed by reshaping of premolars and keeping primary canines with a suitable restoration are two common treatment plans. Maintenance of the primary canines has the advantage of avoiding alveolar resorption for implant placement until the patients reach the appropriate age which also helps avoid bone grafting. It must be noted that different individuals receive treatments tailored for their specific situation (11, 16). In the present case, mandibular right primary second molar received a direct restoration treatment. All remaining primary teeth without permanent successors should receive caries preventive treatments, such as fluoride therapy, and undergo regular dental visits to save these teeth until bone maturation and preserve maximum bone density for subsequent implant placement. Considering the obvious spaces in the dentition of the present case, he should receive orthodontic treatment for space management before implant treatment.

An association has been reported between hypodontia and dental anomalies such as peg-shaped lateral incisors, impaction, transposition, taurodontism, and ectopic

eruption (21). In 2018, Jafari et al. assessed the relationship between the prevalence of dental anomalies and tooth agenesis among 9-20 year-old individuals in Ardabil city, Iran, and found that there was no significant relationship between them. However, the most commonly affected teeth were mandibular second premolars, maxillary second premolars, and maxillary lateral incisors (22). Lateral incisor microdontia was seen in the present case. As mentioned earlier, hypodontia is regarded as an autosomally inherited dominant condition with varying expression and incomplete penetrance. Studies show that pig-shaped lateral incisors can be a mild variant of hypodontia (23). In the present case, hypodontia in the patient and a peg-shaped lateral incisor in his mother were detected.

Evidence shows that mineralization of third molars is a population-specific process that varies according to age in different ethnic groups. In the present case, initial mineralization of the third molar was not observed on the panoramic radiograph at the age of nine, while it should be seen averagely at 8.87 years of age (24-27). This phenomenon may be considered a population-specific process.

The authors did not perform any treatment because the patient required a follow-up until the proper age.

Conclusion

Tooth agenesis is a common dental anomaly associated with significant esthetic problems. Its proper management requires early detection by clinicians besides a multidisciplinary approach.

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