Case Report

Bilateral Second Premolars Agenesia Together with a Unilateral Canine Radiculomegaly

Abstract

Congenitally missing teeth is a common feature for the third molars. However, missing teeth, macrodontia and radiculomegaly occurring in a single patient is very rare. This article describes a case of agenesis of mandibular second premolars, radiculomegaly with dilacerations of a canine tooth together with elongated roots of other canines. All these features had been discerned through diagnostic radiographs taken during a routine treatment planning.

Keywords: *Dental agenesia, dental defects, dilacerations, radiculomegaly*

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Dental Anomaly

There are a number of dental anomalies in existence that affect the tooth structure, shape, and size. While the etiology of these anomalies is generally not clear, certain factors associated with them are thought to affect the tooth at the histo- and morpho-differentiation stages of its development. Some of the etiological factors are genetic or environmental related.[1] Among the factors thought to cause dental anomalies are chemotherapy, radiotherapy, trauma, drugs, and infections, which exert their influence during the stage of the tooth bud cell proliferation. It will be important to note that the whole developmental process leading to dental anomaly appears to be complex and could be involving complex interactions between the genetic, epigenetic, and environmental factors.

Given that the process of tooth development is progressive, the reiterative signaling patterns between the ectoderm and crest cells' derived mesenchymal cell layer take place over a period during the sequential process of development. If this process is interrupted, it could lead to clinical changes that would demonstrate variations in the number, size, and the form of the teeth involved.[2] In the case where the epigenetic factors are involved, the influencing factors can lead to the alteration of the gene expression without necessarily showing changes in in the nucleotide sequencing of the cells,[3] and can wholly or singly influence the dimensional changes in the dentition. On the other hand, when the environmental factors such as trauma and diseases are involved, then the influence will be felt most on the dimensional growth of the dentition. Furthermore, apoptosis taking place in the enamel knot during the dental morphogenesis can also play a greater role in the regulation of the tooth size and shape.[4]

Agenesis

Dental agenesis can be described as the failure of a tooth to develop, and it is the most common developmental abnormality in the humans, with a prevalence rate of 20.7%–25% of the population.[5] While dental agenesis is usually a genetic problem,[6] other factors have been associated with it. These factors include environment, chemotherapy, radiotherapy, trauma, drugs, and infections. Dental agenesis has been reported to be lower in the African populations, with the females reportedly being more affected than the males.[7] Hypodontia has been reported to have a varying prevalence between 2.3% and 10.1%,[8] and when the third molars are excluded, the teeth that are most commonly affected by this anomaly include the mandibular second premolars (7.8%) followed by the maxillary lateral incisors and maxillary second premolars.[9] A unilateral occurrence of this abnormality

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is predominant, except in the case of the maxillary lateral incisors, for which the bilateral occurrence is more frequent than the unilateral.[5,10] The recently introduced tooth agenesis code procedure can help map out the frequency at which these teeth are missing in individuals, thus allowing for the efficient and unequivocal expression of the human dentition with respect to the number and location of the missing teeth.[11] Early diagnosis of agenesis is fundamental for the prevention of maxillary/mandibular disorders, allowing for the establishment of clinical and orthodontic management to achieve occlusal, functional, and esthetic harmony.

Macrodontia

Macrodontia or megadontia is a rare dental anomaly, when the size of a single tooth or a number of teeth is increased.[12] One of the well-characterized syndrome that is associated with large teeth is the otodental dysplasia, an autosomal dominant microdeletion of chromosome 11q13 that causes grossly enlarged canines and molars, and is also associated with eye defects and certain degree of hearing loss. This syndrome is very different from the radiculomegaly reported in oculofaciocardiodental (OFCDS) syndrome. This syndrome is a rare X-linked dominant syndrome characterized by canine teeth with extremely large roots (radiculomegaly), dental abnormalities, congenital cataracts, dysmorphic facial features, and congenital heart disease.[13] While the diagnosis of this syndrome is usually difficult, there are unique and specific symptoms, diagnosable characteristics associated with the dental, skeletal, ocular, and cardiac structures, which are usually present. Apart from radiculomegaly in OFCDS, other characteristics are the delayed eruption, malposition, root dilaceration, and oligodontia.[13]

While the prevalence of macrodontia has been estimated to be 1%–2% in males and 0.9% in females,[14] cases of nonsyndromic radiculomegaly are rare as only twenty cases have been documented as at 2010. A study by Maden *et al*.[15] found that radiculomegaly of the canine resulted in the change of the morphology of the tooth with the tooth occasionally having two root canals, root dilacerations, increased root length, and open apices. The present case report describes a nonsyndromic localized radiculomegaly of the canine and bilateral agenesis of mandibular second premolars.

Case Report

A 17-year-old girl, in good general health with no previous history of dental treatment, was referred to a pediatric dental clinic of a local University in Kenya, with a complaint of crowding in the upper anterior arch. Her previous dental history indicated the patient incurred trauma involving 21 and 22, with accompanying laceration of the upper lip. The injury had been due to collision. An oral examination of the patient at the time of the consultation revealed no remnant extraoral injuries or defects. However, intraorally, the patient had generalized mild dental plaque covering most of the teeth, lower second premolars (35 and 45) were missing, distolingual rotation of lower right canine, (43) mesiolingual rotation of lower first premolars (44 and 34), proclined upper central incisors (11 and 21), retroclined upper left second incisor (22). She had a V-shaped maxillary arch, Class III bilateral molar relationship, Class II canine relationship, an increased overjet of 10–12 mm, and an overbite of approximately 40%.

Apart from the dental study models that were fabricated, orthopantomogram and intraoral radiographs were also taken. The study models were analyzed for dental crowding using Carey's analysis, and the results showed a space discrepancy of 7.5 mm and 8.5 mm in the maxilla and the mandible, respectively. Figure 1 shows the study models, clearly illustrating the dental arch shape and the dental occlusion together with the crowding of the teeth within the dental arches.

Radiographic examination of the panoramic radiograph taken [Figure 2a] revealed a mesially rotated lower right permanent canine (43), with a prominent mesial root dilacerations, enlarged pulp chamber, and root canal and radiculomegaly of the lower right permanent canine (43) extending close to the lower border of the mandible. Further, there was relatively increased root length of the upper permanent canines (13 and 23) and absence of the lower second premolars (45 and 35). The intraoral periapical radiograph [Figure 2b] that had also been taken of the lower right permanent canine (43) showed an elongated root with a crown to root ratio of more than 1:4, increased size of root canal of lower right permanent canine (43), dilaceration, and radiculomegaly. Furthermore, noted was the notching of the mesial part of the root apex of the lower right premolar (44).

A diagnosis was made of a young lady with bilateral Class III molar relationship and Class II canine relationship, moderately severe crowding in both the maxillary and

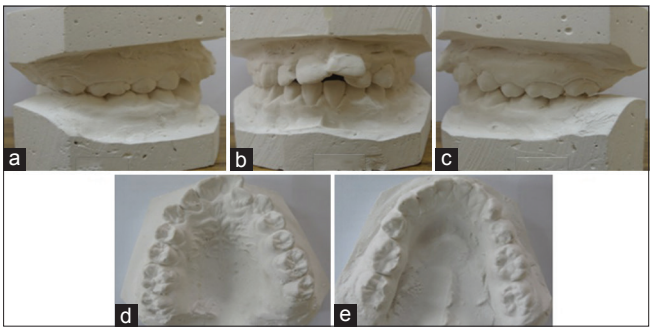

Figure 1: (a-e) The study models of the patient showing different views of the dental arches, dental occlusion, and different positions and rotations of the teeth within the dental arches

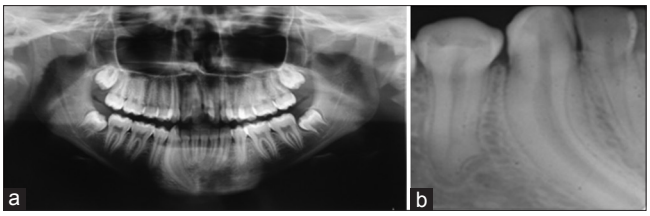

Figure 2: The panoramic radiograph (a) showing radiculomegaly of 43 with root dilacerations mesially, agenesis of second mandibular premolars, increased root length of maxillary canines, and impacted 38 and 48. The intraoral periapical radiograph 43 (b) shows the radiculomegaly of the 43, increased root curvature mesially, increased size of the root canal, and the notching of the root apex of 44 mesially

mandibular arches, increased overjet, multiple teeth rotations, agenesia of the mandibular second premolars, nonsyndromic radiculomegaly of the mandibular right canine, and increased root length of maxillary right and left canine. The objectives of the treatment were to correct the crowding, reduce the overjet, and restore masticatory function and harmony. This treatment was discussed with the parents and agreed upon, and included oral health education and oral hygiene, correction of the crowding to improve the esthetics, reduce the overjet, reduce the maxillary incisor proclination, achieve a harmonious Angle's Class I molar relationship and Class I canine relationship, and regular recalls for reinforcement of oral hygiene measures. However, the patient, unfortunately, had to move away from the city and was hence unable to return for continuation of the treatment. Although to date the case remains untreated, efforts to get the patient has not yielded any fruit yet.

Discussion

Tooth agenesis, the congenital absence of one or more primary or permanent teeth, is one of the most frequently observed dental anomalies in children.[5] Previous studies have reported the prevalence of dental agenesis as to vary from 2.2% to 10.1% (most of them ranging between 6% and 8%).[5] Higher frequencies of tooth agenesis have been reported in females than in males.[5,16] This condition has been shown to be related to other dental anomalies such as microdontia or peg-shaped incisors, taurodontism, transposition, supernumerary tooth, ectopic eruption, retained primary tooth, and ectopic eruption.[17,18] The teeth that were missing in the present case were lower second premolars.

Given dental agenesis is well documented, the most common pattern in the lower arch involves agenesis of the mandibular second premolars, followed the agenesis of the incisors, canine, and the second molar within the same arch. A recent study Kim *et al*.[19] has showed that tooth agenesis could be a symmetrical phenomenon with a prevalence of 40.2% in the lower arch and 52.2% in the upper arch. The relatively diverse arch tooth agenesis could be suggestive of different mechanisms being responsible for tooth agenesis in the upper and lower arches. In the present study, symmetry (left versus right) of agenesis patterns involving the second premolars has been demonstrated. A recent study on genetic defects responsible for tooth agenesis has made some stride in identifying the genes involved.[20] Some of the past studies have reported that MSX1 mutations account mainly for the premolar agenesis and that PAX9 mutations account mainly for molar agenesis.[21,22]

The most expressive variations of a tooth have usually been within the internal and external part of the root. In the case of the canine, variations such as single root with a single canal form, single-rooted canine with two[1,23] or three root canals, and canine teeth with two different roots[1,24] have been reported. Further, other commonly seen in these teeth are descriptive variation elements such as grooves, pits, and fissures; slope of the sides of the cusps; shape of the incisal surface; ratio among enamel, dentin, and cementum; dimensions of pulp chambers; and the quantity, size, and morphology of the number of roots and root canals.[1] Some of these variations have been described for the canine in the present case. While the average tooth lengths of the normal mandibular canine is usually about 24 mm, the lengths of the abnormal one scan sometimes be as extensive as 50 mm or more. The estimated length of the present case was 43 mm.

The diagnosis of radiculomegaly as an abnormality has generally been confirmed between 15 and 20 years of age, when the radiculomegaly becomes evident since during this period, it is expected the root formation of the canines would have been completed. Histological study of abnormal canines has previously indicated disorder in the dentine formation and usually with accompanying thin enamel formation.[25] Although no histological examination had been indicated in the present 17-year-old girl, radiographical examination did not show any definite abnormality within the canine. Again in the present case, the tooth has not caused any discomfort to the patient, but the exception would be that the tooth cannot be orthodontically moved or derotated, due to its abnormality and position within the arch.

The OFCD syndrome characteristically is accompanied with the presence of canine teeth with extremely large roots (radiculomegaly),[26] and the persons suffering from the syndrome would have facial features that include a long narrow face, high nasal bridge, and a bifid nasal tip. The present case did not demonstrate any of these features, and neither were there any cardiac defect nor any evidence of the occasional syndactyly of the second and third toes, hammer-type flexion of the second and fourth toes, radiculnarsynostosis, and vertebral and rib anomalies that would be found in cases of OFCD.[27] In the present case, other than the absence of lower second premolars (44 and 34), the patient did not have the other typical

features of OFCDS,[13] and neither were there any evidence of the general features of the canine crown variations. From a clinical point of view, the anatomical variations that go with the lower canine with radiculomegally can complicate treatment planning relating to endodontic, orthodontic, and surgical procedures if not properly diagnosed. It is important for the surgeon to have an idea on the size and shape of the crown and roots and the number of roots associated with a tooth, its position in the dental arch;[28] hence, the importance of appropriate radiographs before any treatment is commenced. It is also still important for the consideration of elective tooth extraction, which in such case will have to involve making of a flap, osteotomy, and tooth sectioning.

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Conflicts of interest

There are no conflicts of interest.

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