

Multiple tooth anomalies in a nonsyndromic patient with class II division 2 malocclusions: A case report and a literature review

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Abstract

Reports in the literature about the craniofacial characteristics of patients with class II division 2 malocclusions show a lot of different patterns accompanied by palatally displaced upper incisors, congenital missing teeth, polydiastema, fusion, germination, tooth impaction, peg-shaped lateral incisors, persistent teeth, hypodontia, persistent deciduous teeth, transpositions, and supernumerary teeth. The following case report focuses on the description of the clinical characteristics observed on a patient with a very unusual conjunction of dental and skeletal anomalies mentioned above, as well as a literature review on the related issues. Extra-intra-oral examinations, radiographic evaluations, orthodontic consultation, and reviewing the literature concluded that this nonsyndromic patient that refused to receive all dental treatment approaches is special with its uniqueness.

Key words: Angle class II, fused teeth, malocclusion, overbite, tooth abnormalities

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Introduction

Class II division 2 malocclusions may occur in a variety of dentoalveolar malocclusion structures. Genetic inclination may play an important role among these differentiations. Dental anomalies in number, size, position, or structure are also important causes that can lead to malocclusions. Genetic or environmental factors, such as drug embryopathy, fetal alcohol exposure, or hyper/hypovitaminosis in a pregnant mother, are thought to be the etiology of the anomalies. It may be easy to say that the etiology of the two conditions can be considered to be a nested phenomenon. Basdra *et al.* reported that class II division 2 malocclusions are closely associated with some types of congenital tooth anomalies, such as missing teeth, peg-shaped laterals, transpositions, or impactions.^[1] These malocclusions, which could be seen either on their own or with various accompanying disorders, may actually be polygenic and additive in nature through combined expression of genetically assigned

anatomical components. However, previous studies have not examined how many dental anomalies could be seen in one patient.^[1] For this reason, this case study presents a very unusual combination of multiple dental anomalies, including germination, peg-shaped teeth, congenitally missing teeth, a geminated impacted canine, persistent teeth, and polydiastema with class II division 2 skeletal anomaly in a 15-year-old boy.

Case Report

A healthy 15-year-old young boy was referred to our clinic due to a toothache in the lower right canine region. Informed consent given by the institution was signed by the patient's parents. The patient had no systemic disorders, and there was no syndrome history in his medical or

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family background. During the extra-oral and intra-oral examinations [Figure 1a-h], some skeletal and dental anomalies such as deep-bite, polydiastema, missing teeth, class II molar and canine relationships, peg-shaped laterals, and a fusion/germination phenomenon were observed. After a detailed radiographic examination including panoramic, cephalometric, postero-anterior and periapical radiographs, various dental anomalies, such as hypodontia, gemination, peg-shaped teeth, persistent deciduous teeth, and impacted geminated teeth were noticed [Figure 2a-c].

During clinical observation, it was noticed that the patient had a tendency for class II division 2 malocclusion. The basic features of this malformation include the class II molar relationship of the dental arches combined with retroclination of the upper incisors and excessive incisal overbite [Figure 1d and f]. The gonial angle seemed to be low, resulting in a rather square facial profile [Figure 1c]. The labiomental groove was pronounced beneath the lower lip, evoking and strengthening the diagnosis of this class II division 2 malocclusion. For this reason, consultations with clinicians from the orthodontics department were done and after a proper orthodontic evaluation including cephalometric analysis [Figure 3], it was found that the patient had a normal sagittal maxillary position ($SNA = 86.9$) and a retruded mandibular position ($SNB = 78.0$). According to the vertical evaluation, the patient had a low angle pattern ($Sn-GoGn = 26.4$) as well as lower facial height ($ANS-Me = 57.1$). These outcomes confirmed that these dental anomalies were

associated with skeletal class II division 2 malocclusion with a reduced lower facial height [Figure 3]. The racial status of the patient was “white-caucasian.” For that reason, the European cephalometric norms were subjected for the patient.^[2]

Radiographic examination revealed that the mandibular permanent central and lateral incisors were bilaterally congenitally missing [Figure 4a]. In addition, the right and left mandibular third molars and the left maxillary third molar were also found to be congenitally missing [Figure 4b and c]. Because seven congenitally missing teeth were determined in one patient, this condition could be termed as being “oligodontia,” according to the literature.^[3] The right maxillary permanent canine and the second premolar teeth were observed as impacted radiographically [Figure 5].

An irregular bilateral morphology of the mandibular permanent canines, with greater mesiodistal crown width developmental occluso-gingival slots on the buccal and lingual surfaces, was found to be geminated [Figures 4a and 6a-c]. A vitality test was performed on the geminated teeth, and the right mandibular canine was found to be nonvital, the other teeth were found to be vital, although the teeth were diagnosed as being caries-free. Gingivitis related to all the teeth was detected, especially around the geminated teeth. The maxillary permanent right canine was also found to be geminated. The geminated tooth on the upper jaw was found to be impacted,

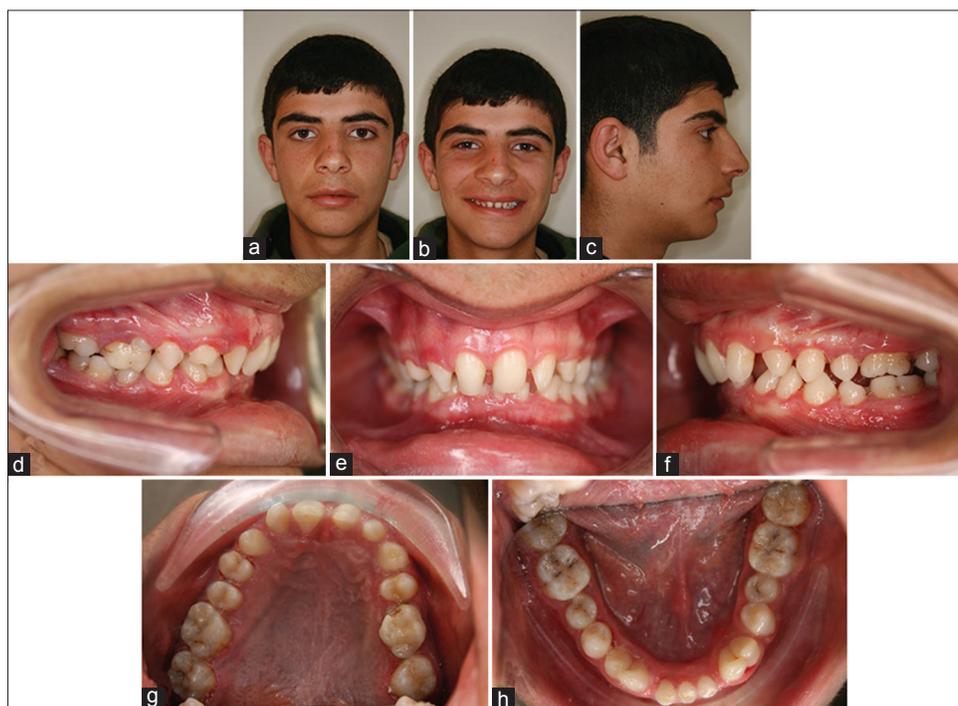


Figure 1: Extra-oral photographs of the patient. (a) Anterior facial view. (b) Anterior smiling view. (c) Profile view. (d) Right dental view (note that the impacted tooth is visible on the gingival margin.) (e) Frontal dental view. (f) Left dental view. (g) Occlusal view of the upper dentition (note the polydiastema). (h) Occlusal view of the lower dentition

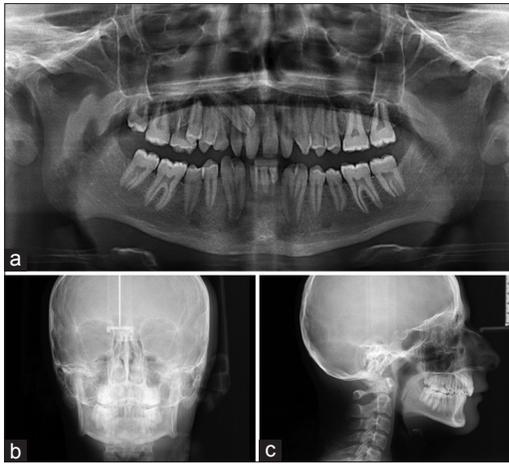


Figure 2: (a) Panoramic radiograph of the patient. (b) Antero-posterior radiograph of the patient. (c) Lateral cephalometric radiograph of the patient

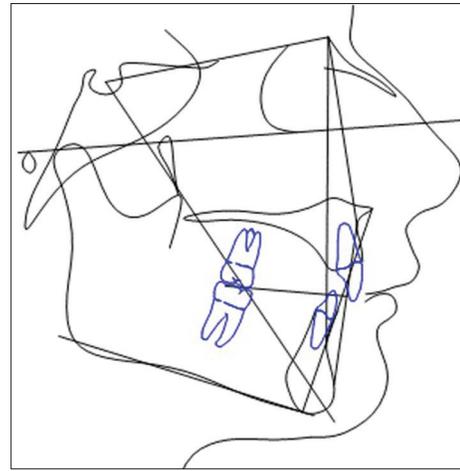


Figure 3: Cephalometric tracing of the patient

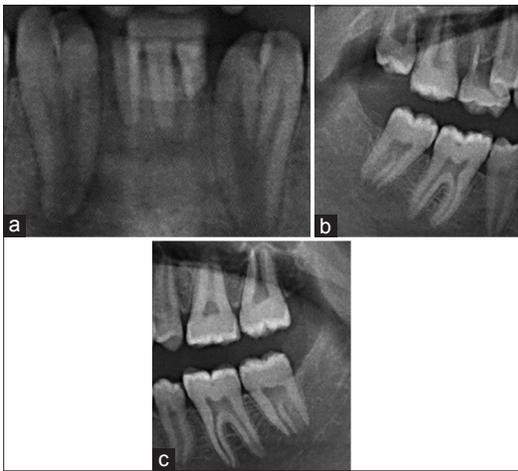


Figure 4: (a) Radiograph of the mandibular anterior area (showing the absence of mandibular central and lateral incisors bilaterally). (b) Radiograph presenting the lackness of the right mandibular third molar. (c) Radiograph exhibiting the lackness of the left mandibular and maxillary third molar



Figure 5: Radiographic view of the impacted maxillary permanent right canine and second premolar



Figure 6: Intra-oral photographs of the patient. (a) Occlusal view. (b) Left intra-oral view. (c) Right intra-oral view



Figure 7: Anterior and lateral intra-oral views of the patient (note the bilaterally peg-shaped maxillary lateral incisors, left maxillary canine and right maxillary central teeth and polydiastema)

radiographically [Figure 5]. The maxillary lateral incisors, bilaterally, the right central incisor, and the left canine were found to be peg-shaped teeth. Polydiastema was noticed related to the peg-shaped teeth, clinically [Figure 7a and b].

The right mandibular primary central and lateral incisors, the left mandibular primary central incisor, and the right maxillary primary canine were persistent [Figures 4a and 5]. The incisal edges of the persistent incisors were eroded because of the deep-bite, although their roots were either intact or minimally resorbed [Figures 6a and 4a].

The diagnosis outcomes were summarized to the patient and all the multi-discipliner treatment plan including departments of periodontology, conservative treatment, surgery, orthodonty, and prosthetic treatment was declared to the parents. However, the family's social security was not suitable for a detailed approach like this and besides they were living far away from the university clinic and traveling several times would cost a lot of money which this family might not afford; therefore, the parents refused to take required treatment. For this reason, the follow-up records and pre-post treatment evaluation could not be performed.

Discussion

There are a number of studies about class II division 2 malocclusions in the literature that show many different forms that are accompanied by anomalies, such as displaced upper incisors, congenital missing teeth, polydiastema, fusion, germination, tooth impaction, peg-shaped lateral incisors, hypodontia, persistent deciduous teeth, transpositions, and supernumerary teeth.^[1] However, the present study aimed to present a more extreme case of class II division 2 malocclusion with a multiple combination of rare dental anomalies, such as bilateral geminated lower canines, an impacted geminated canine, oligodontia, persistent teeth, polydiastema, peg-shaped lateral incisors and canines, and agenesis of the third molars associated with a class II division 2 skeletal pattern. According to our best knowledge, no scientific literature has previously reported on a similar association among these types of dental abnormalities in a single patient.

In the study of Basdra *et al.*,^[1] the author suggested that there is a strong association between class II division 2 malocclusion and congenital agenesis. He also stated that with the exception of the third molars, the absence of all other teeth was seen at least 3 times more often in individuals having class II division 2 malocclusion than in the general public. In another study, it was concluded that there were significant rates of dental abnormalities in orthodontic patients; however, there was no association between orthodontic malocclusion types and dental anomalies, except for impaction.^[4]

Defects in certain genes and etiologic factors in the pre and postnatal periods are thought to be responsible for dental anomalies.^[5] Although genetic background plays an important role in skeletal class II division 2 malocclusion and some dental anomalies such as congenital missing teeth, peg-shaped laterals, transpositions, and impactions, it was found that a minimum number of dental anomalies was reported in skeletal class II division 2 malocclusion according to the results of the study by Uslu *et al.*^[4] However, in this current case report, multiple dental anomalies were presented with skeletal class II division 2 malocclusion.

Familial occurrence of class II/division 2 deep-bite has been attested in several literature publications including twin and triplet reports by Kloeppel,^[6] Markovic^[7] and Litt and Nielsen^[8] and pedigree studies by Korkhaus^[9] and Trauner.^[10] Peck *et al.*^[11] declared that these studies points to indisputable genetic influence, probably of an autosomal dominant type with incomplete penetrance, as a critical element in the formation and expression of Angle's class II/division 2 deep-bite malocclusion. They concluded that inheritance of a complex occlusal variation such as II/2 deep-bite malocclusion may actually be polygenic and additive in nature, through combined expression of genetically determined anatomical components, rather than being the effect of a single controlling gene for entire occlusal malformation. Regarding this case report, the authors thought that some isolated nonsyndromic genetic problems such as a single nucleotide polymorphism or multiple mutations in genes responsible with the formation of teeth or maxilla-mandibular skeletal complex would have been found if the patient had agreed to take the related tests. Future case reports should better focus on the genotypes of the patient.

Hypodontia is the lack of dentition in which one or more (up to five) teeth are congenitally missing, whereas the absence of more than five teeth is known as oligodontia.^[3] The prevalence of hypodontia ranges from 0.3% to 11.3% in the literature; however, oligodontia is rather less apparent, and it is not as common as hypodontia.^[12-15] It can be seen in syndromic patients as well as nonsyndromic individuals. Several studies disclosed that seven genes are currently known to have a potential for causing nonsyndromic oligodontia: PAX9^[16] (paired box gene 9), MSX1^[17] (muscle segment homeobo × 1), EDA^[18] (ectodysplasin A), AXIN2^[16] (axis inhibition protein), NEMO^[19] (NF-kappa-B essential modulator), KRT17^[20] (keratin 17), and EDARADD^[16] (EDAR-associated death domain). These types of abnormal incidents accompanying by class II division 2 malocclusion were known to be infrequent. In this report, seven permanent teeth, including the third molars (3), the central incisors (2), and the lateral incisors (2), were congenitally missing and could be classified as oligodontia. Therefore, the present case is differentiated from the previously published studies related to class II division 2 malocclusion because it included nonsyndromic oligodontia.^[1]

The incidence of gemination occurs when two teeth originate from one tooth bud and consequently, the patient has a bigger tooth, but the number of teeth is normal.^[21,22] This can also cause impaction due to the larger volume of the crown of the geminated tooth.^[23] In addition, it can be said that the gemination phenomenon commonly occurs in the anterior maxillary region.^[24] In the present case, the impacted right maxillary canine was identified as being geminated due to the maxillary location. It did not induce

an increase in the number of teeth in the arch. Moreover, it had a larger tooth appearance, and it was found to be impacted. In contrast to gemination, fusion arises through the union of two normally separated tooth germs when the patient appears to have a missing tooth. However, when a normal tooth bud unites to a supernumerary tooth germ, it could also be described as “fusion.”^[21,22] Fusion is more frequently seen in the anterior mandibular region.^[24] In the present case, bilateral mandibular permanent geminated or fused teeth were found clinically and radiographically. This condition can be termed as bilateral fusion since the anterior mandible location of the teeth caused a decrease in the number of teeth in the arch. On the other hand, the missing teeth seen in the mandible is certainly not an evidence of the fusion phenomenon. Among these types of patients, the number of teeth is also normal; thus, distinguishing between gemination and fusion might be rather strenuous or un-achievable.^[25] In order to overcome the difficulty in the diagnosis and terminology of these types of cases, the term “double or twinning tooth” could be introduced.^[24,26,27] These conditions, gemination or fusion, are very unusual cases in a permanent dentition. Moreover, the fact of being bilateral or multiple makes them relatively rare.^[26,27] Throughout the literature, no studies were noticed regarding the genetic background of tooth fusion as well as gemination. It is obvious that future studies should be concentrating on the genetic basis of tooth problems especially fusion and gemination.

According to our most recent knowledge, no scientific or well-ordered paper has been formerly publicized in the literature about an equivalent connection of multiple dental abnormalities in a single patient. This kind of impaction, tooth agenesis, and peg-shaped teeth are thought to be responsible for the polydiastema seen in the patient in the present case. This condition can cause esthetic, orthodontic, and periodontal problems for practitioners.^[28] When teeth dimensions are out of their normal range and smaller than usual, the term “microdontia” is used to define the case. Until now, three types of microdontia have been characterized as follows: True generalized microdontia, relative generalized microdontia, and microdontia involving a single tooth. Maxillary lateral incisors, which are affected by microdontia and which are commonly called “peg-shaped laterals,” are one of the most well-known forms of this phenotype. A peg-shaped incisor has a remarkable decrease in diameter, extending from the cervical area to the incisal border.^[1,2] However, microdontia of the maxillary and mandibular central incisors is also a comparatively rare condition.^[29] In the current case, the patient had bilateral microdontic peg-shaped maxillary lateral incisors along with a unilateral maxillary peg-shaped central incisor as an unusual condition. This uncommon peg-shaped tooth anomaly is found commonly on the maxillary lateral incisor; other tooth types are rarely influenced by an anomaly such as a mandibular central incisor condition.^[29]

Although the impacted teeth, especially the maxillary canine teeth, were found to be closely associated with class II division 2 malocclusion cases in the literature,^[1] some of the authors demonstrated no relationship between impaction and malocclusion.^[4] In the present case, the right maxillary canine tooth was found to be impacted, and the impaction was located on the apexes of the right maxillary lateral and central incisors, radiographically. The impacted canine had a large pulp chamber and a mesiodistal diameter, and it appeared to be a geminated tooth.

In addition, the patient had right mandibular primary central and lateral incisors, a left mandibular primary central incisor, and a right maxillary primary as persistent teeth. Persistent teeth seen on mandibular primary central and lateral incisors were accepted as being relatively rare in the literature.^[30] Moreover, previous studies showed that the most common reason for the persistence of primary teeth was the congenital absence of a permanent successor teeth, followed by impaction, abnormal position, and late eruption of successor teeth. In accordance with the literature, the current case showed a congenital absence of permanent successor teeth and no or minimal root resorption related to the successor teeth.^[30-32]

Conclusion

The combination of dental abnormalities as seen in this case presumably points to unknown genetic factors. This case presents a rare combination of multiple dental anomalies in a nonsyndromic patient, which is different from formerly reported cases. In addition, the case highlights an association between class II division 2 malocclusion and various dental anomalies such as palatally displaced upper incisors, congenital missing teeth, polydiastema, fusion, germination, tooth impaction, peg-shaped lateral incisors, persistent teeth, hypodontia, persistent deciduous teeth, transpositions, and supernumerary teeth. Clinically, such cases are important and aid in our ability to advance our understanding of the different types of anomalies that might be seen in clinical practice. Dentists infrequently encounter such multiple dental abnormalities cases, and a clearer understanding of the abnormalities may be useful for a true diagnose and better clinical management. Cases of nonsyndromic patients with multiple dental anomalies are intriguing, and new research studies are needed to enlighten the etiology. The authors declare that there is no conflict of interests regarding the publication of this paper.

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