Triplication in permanent teeth: A rare case report

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Abstract Fusion is an anomaly manifested in both deciduous and permanent dentitions. Triple tooth refers to the union of three separate tooth entities. It can involve the normal dentition or supernumerary teeth. Triplication is rarely encountered in deciduous and permanent dentition with an incidence of 0.02%. The case presented herein describes a rare case of triplication in permanent maxillary incisors and supernumerary teeth in a 15-year-old female.

Keywords: Concrescent triplet, double tooth, fused teeth, odontoplasia, supernumerary tooth, tooth abnormalities

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INTRODUCTION

Odontogenesis is a complex process, wherein more than 200 genes are known to play a significant role in tooth development. An imbalance can lead to an abnormality in the number, size, shape or structure of the developing tooth/teeth.^[1] Malformations in primary dentition can lead to relatively transient esthetic and functional problems in addition to retard or alter the development of permanent teeth. However, when such malformations occur in permanent dentition, the treatment planning should be executed with taking care in mind that the teeth are immature permanent teeth.

Dental anomalies such as fusion, gemination, supernumerary teeth and concrescence are not rare. Conjoined, double or triple teeth are other terms used to designate such abnormalities.^[2] Whereas the fusion of three teeth is termed triplication defect. Although "double teeth"

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(result of fusion or gemination) are relatively common with a reported prevalence ranging from 0.1% to 1.55%, occurrence of union of three teeth that is reported as "triple tooth" is very rare.^[2,3] The prevalence of triplication in the deciduous dentition has been reported to be 0.02% and is more commonly seen in maxillary arch than the mandibular arch and the condition is more prevalent in males.^[4] In literature search, we did not encounter any report with triplication in permanent dentition. Herein, we present a rare case of triple teeth in permanent dentition in a 15-year-old female with associated periapical pathology.

CASE REPORT

A 15-year-old female [Figure 1a] reported to the Department of Pedodontics and Preventive Dentistry, with a chief complaint of unesthetic upper front tooth with an associated swelling [Figure 1b]. There was a history of painless swelling for 8 months. Swelling has slowly progressed to the present size. Past history and

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family history were not relevant. Extraoral examination revealed hard bony swelling, which was nontender in nature, skin over the swelling was normal, but there was associated facial asymmetry on the left side. On intraoral examination, swelling extended from the upper left central incisor to canine region, involving the vestibule. There was presence of an abnormal maxillary central incisor which gave an appearance of either talons cusp or dens in dente. In addition, there was trauma from occlusion in respect to the abnormal tooth. The patient was advised for an intraoral periapical radiograph, orthopantomogram (OPG) [Figure 1c] and cone beam computed tomography (CBCT) imaging. OPG revealed missing upper right and left lateral incisors and an abnormal tooth structure. The CBCT in three-dimensional reconstruction [Figure 2a], axial view [Figure 2b] and the panoramic view [Figure 2c], showed an abnormal tooth structure in maxillary left anterior region confirming a provisional diagnosis of fusion of teeth.

After routine blood investigations, the patient was prepared for the surgical exploration of the site. Surgical exploration revealed the presence of three roots. After the surgical exposure of the site, three roots were visible which confirmed the diagnosis of fusion of three teeth, known as triplication of teeth [Figure 3a]. After granulation tissue curettage, the area was irrigated with normal saline, and a fragment of tooth was removed to maintain the bony contour [Figure 3b]. The cavity was packed with a hydroxyapatite bone graft and following that a plasma-rich fibrin membrane was placed [Figure 3c and d] which acted as scaffold. The hard tissue and soft tissue were sent for histopathological examination. Histopathological examination revealed



Figure 1: The extraoral (a) and intraoral (b) picture showing mild swelling on the left maxillary anterior region. The orthopantomogram revealing a fusion of the teeth (c)

chronic nonspecific inflammation. The patient was kept on follow-up. On follow-up visits, since the graft did not take up, extraction of the tooth was carried out. The extracted tooth was then sent for histopathological analysis by decalcification [Figure 4a and b] and ground sectioning [Figure 4c and d].

DISCUSSION

Although the etiology of fusion is still unclear, it is believed that physical force or trauma results in the contact of developing tooth germs, producing necrosis of the epithelial tissue that separates them and leads to fusion. Other possible causes include viral infection during pregnancy and the use of thalidomide. Dominant autosomal heredity has also been proposed as one of the etiologic factors.^[4,5]

Various studies cited the possible etiopathologic factors for triple tooth formation. The extent of fusion is determined by the stage of development when the union occurred. Knapp and McMahon suggested in case where all permanent teeth are present it could be a gemination and a fusion with supernumerary teeth. Trubman and Silberman described it as a combination of fusion and germination.^[6] In the present case, probably, there is triplication of permanent maxillary left central incisor with the congenitally missing maxillary left lateral incisor and a supernumerary tooth. A possible explanation can also be derived as the contralateral side also showed a missing maxillary lateral incisor, indicating a triplication of permanent maxillary left central incisor with two supernumerary teeth.



Figure 2: Cone beam computed tomography in three-dimensional reconstruction (a), axial view (b) and panoramic view (c), shows abnormal tooth structure in maxillary left anterior region



Figure 3: Upon surgical exploration, the three roots depicting triplication of maxillary central incisor (a) are apparent. After curettage of the granulation tissue, a fragment of tooth was removed to maintain the bony contour (b). The cavity was packed with a hydroxyapatite bone graft and following that a plasma-rich fibrin membrane was placed (c) which acted as scaffold (d)

As reported in literature, triplication is more common in deciduous dentition, making our case unique, with the anomaly in permanent teeth.^[12] Age of the children generally ranged from 1 year 11 months to 10 years with a mean age of 4.9 years. Triple tooth is more prevalent in boys compared to girls with a ratio of 2:1 and this finding is in contrast to our case. Maxillary triple teeth are more common than mandibular by location.^[2,7] Furthermore, there is a preponderance of triple tooth on the left side over the right side (4:3) with only Schultz-Weidner reporting bilateral occurrence of maxillary triple tooth.^[8]

As stated in the article of Dhooria and Badhe,^[9] Knapp and McMahon used the word "triple tooth" for the first time describing this anomaly in children. In archeological literature, Bennazi *et al.* have reported recently a case of triple teeth in a 5-year-old child discovered in a late medieval cemetery in Italy.^[10] After the description of the first case by Bennett in the late 1880s, Sprawson in 1931 probably described another case of triple tooth in his book.^[9,11] Since then, there were only five cases reported till 1984 when Knapp and McMahon reported a case and reviewed briefly all the previous cases. Literature review shows the occurrence of triple teeth only in deciduous dentition but not evident in permanent dentition [Table 1]. The present case is first of its kind with triplication apparent in permanent maxillary left incisors.

Apart from being unesthetic, clinical problems such as dental caries, abscesses and fistulae can complicate restorative and endodontic procedures. Thus, early identification of dental conjoining anomalies has been



Figure 4: Photomicrograph of decalcified section and ground section of a tooth showing fusion of teeth in low power (a and c) and high power (b and d)

Table 1: Lit	erature	review	of	triplication	of t	eeth
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Author	Year	Age/sex	Dentition	
Goenka, <i>et al</i> .	2009	7/male	Deciduous	
Mohapatra, et al.	2010	10/male	Deciduous	
Suresh Yadav, et al.	2011	10/male	Deciduous	
Babaji, <i>et al</i> .	2012	6/male	Deciduous	
Urvashi sharma, et al.	2013	7/male	Deciduous	
Mohassel, <i>et al</i> .	2014	5/female	Deciduous	
Srinivas and Shatraj	2015	7.5/female	Deciduous	
Juneja, <i>et al</i> .	2015	9/male	Deciduous	
Deepti Thakkar, et al.	2016	5/male	Deciduous	

recommended. In the present case, two teeth, i.e. 21 and supplementary teeth were visible intraorally but the third teeth were impacted. Due to lack of space, rotated 21 and palatally placed supplemental teeth, there was localized gingivitis. Malocclusion and continuous irritation lead to the formation of chronic noninflammatory lesion which leads to buccal cortical plate expansion. Hence, it is of prime importance to rule out the presence of triplication in permanent dentition too.

Differential diagnosis of the conjoined teeth can be compound odontoma, which consists of discrete small tooth-like structures.^[5] Clinical detection of triple teeth in permanent dentition is often difficult as radiographs usually fail to detect them. It is recommended that radiographs have to be taken at varied angulations with different exposure guidelines. Triple teeth also complicate endodontic, oral surgical procedures and periodontal treatment apart from being unesthetic. Since the longitudinal grooves created by the fusion of three teeth are susceptible to caries, sealant therapy and fluoride application may be necessary.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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