CASE REPORT

# Syndontia with talon cusp

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## ABSTRACT

Teeth are specialized structural components of the craniofacial skeleton. Developmental defects occur either alone or in combination with other birth defects. Macrodontia of anterior teeth may occur as an isolated condition or as a result of fusion or gemination and can occur in the primary or permanent dentition. Fusion is more commonly seen in the anterior maxillary region. This case presentation reports a case of fusion of a supplemental tooth to one in the normal series in conjunction with a talon cusp. This condition is extremely rare and has been reported at fourth occasion in the literature. The etiology, prevalence, clinical features, and management of the aforementioned anomalies have been reviewed in detail. Early diagnosis of this condition is important because it may cause clinical problems, such as esthetic concerns and tooth crowding.

***Key words:*** Fusion, gemination, macrodontia, talon cusp

## INTRODUCTION

The growing tooth is the biologic recorder providing precise and permanent record of variations and fluctuations in the tooth development.[1] Double tooth,[2] double formations, joined teeth, fused teeth, or dental twinning[3,4] are the terms frequently used to describe the anomaly of conjoined teeth.[2] Other terms, such as fusion Germination, Connation, linking tooth, syndontia and schizodontia have also been suggested.[2] Because the etiology and ontogeny are uncertain the term "double tooth" is probably the most appropriate term.[5]

Macrodontia, macrodontism, megalodontia, megalodontism, gigantism, megadontium are terms that have been introduced to describe teeth that are abnormally large.[6] Macrodontia of anterior teeth may occur as an isolated condition or as a result of fusion or gemination and can occur in the primary or permanent dentition. Fusion is defined as the joining of two teeth by pulp and dentin with the presence of two canals. Gemination is defined as an attempt by a single tooth germ to undergo division to form two teeth.[7] Fusion may be unilateral or bilateral and is more commonly seen in the anterior maxillary regions of permanent teeth. In addition to affecting the normal teeth, there might be cases of fusion between a normal tooth and a supernumerary tooth.[8]

Talon cusp is an uncommon odontogenic anomaly comprising an accessory cusp-like structure. This was first described by Mitchell in 1892 and thereafter named talon cusp by Mellor and Ripa due to its resemblance to an eagle's talon.[8] The diverse clinical manifestations of the anomaly have led the talon cusp to be described in many different ways: exaggerated cingula, cusp-like hyperplasia, accessory cusp, supernumerary cusp, interstitial cusp,[4] and palatal accessory cusp.[5] A large talon cusp may project with connection to the incisal edge of the tooth to give the crown a "T" or "Y" shape.[5]

Although macrodontia is a rare trait, individual cases slowly make their way into the literature and add to our knowledge and understanding of this condition.[9] The aim of this article is to highlight facts about this anomaly and present a rare case demonstrating macrodontia in the form of fusion along with talon cusp in maxillary central incisor.

## CASE REPORT

A 12-year-old male patient visited the outpatient department with chief complaint of an oversized protruded tooth in the maxillary right anterior teeth region. The patient reported that no other individual in the family had this type of anomaly and there were no anomalies found in the deciduous dentition or any history of trauma. General examination of the patient did not reveal any other associated abnormalities. Intraoral

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examination revealed the presence of mixed dentition stage. The tooth number 11 had a wide crown and showed the presence of a notch on the incisal edge along with prominent mamelons [Figure 1]. The macrodontic incisor measured 15 mm mesiodistally at the midcoronal level and 8.5 mm cervicoincisally. Lingual examination revealed an accessory cusp-like structure, resembling a talon cusp extending from cervical margin of 11 towards its incisal edge measuring 5.5 mm cervicoincisally. Non carious developmental grooves were present at the junction of the talon cusp and the palatal surface of the tooth [Figure 2]. The maxillary central incisor on the contralateral side appeared normal in dimensions but was in crossbite with mandibular left central incisor [Figure 1]. The maxillary lateral incisor on the affected side had erupted palatally [Figure 2]. The number of teeth remained unaffected in the segment. Mandibular anterior segment also revealed marked crowding. The affected tooth responded normally to pulp vitality test. Intraoral periapical radiograph revealed the presence of a large anomalous tooth, which was superimposed by lateral incisor palatally [Figure 3]. The maxillary lateral incisor on the affected side was extracted as part of the treatment plan. The intraoral periapical radiograph then revealed two roots with individual pulp canals. The Orthopantomograph also revealed an impacted supplemental tooth on the contralateral side [Figure 4]. Therefore, based on the clinical and radiographic findings, the case was diagnosed as talon cusp associated with fusion (syndontia) between right maxillary central incisor and a supplemental tooth.

After extraction of maxillary right lateral incisor on affected side an orthodontic appliance for the correction of crossbite on contralateral side, was given [Figure 5]. This was followed by endodontic therapy of pulp chamber and both the pulp canals with selective grinding of talon cusp [Figure 6]. Finally hemisection of the anomalous tooth was performed [Figure 7] for esthetic reasons with continuation of orthodontic treatment. Evaluation of the patient after 6 months revealed marked improvement in esthetics.

## DISCUSSION

Development of tooth is a continuous process in which a number of physiologic growth processes and various morphologic stages interplay to achieve the tooth's final form and structure.[8] When only one tooth is affected then it has to be considered as an anomaly. However, when the characteristic occurs bilaterally it may represent an inheritable trait. Generalized macrodontia may present as characteristic of a syndrome.[5]

Dental morphology is one of several factors that may be involved in the etiology of dental crowding or spacing.[10] Several mechanisms have been proposed to explain the etiology of fusion, including the influence of pressure or physical forces producing close contact between two developing teeth.[4,8,11] as localized pressure on a tooth germ during morphodifferentiation might cause buckling, with either outfolding or infolding of the dental lamina.[12] Of great interest is the fact that once any two or more cusps become united by

**Figure 1:** Tooth number 11 presented with wide crown and incisal notch. Maxillary central incisor on the contralateral side was in crossbite with mandibular left central incisor

**Figure 2:** Talon cusp on the palatal surface of affected tooth

**Figure 3:** Intraoral periapical radiograph showing a large anomalous tooth superimposed by lateral incisor palatally

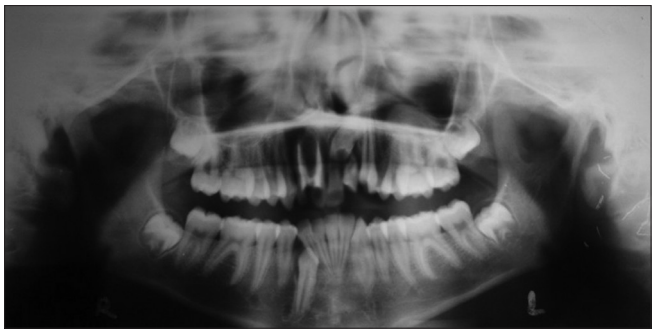

**Figure 4:** Orthopantomograph showing an impacted supplemental tooth on the contralateral side

**Figure 6:** Endodontic therapy of affected tooth with selective grinding of talon cusp

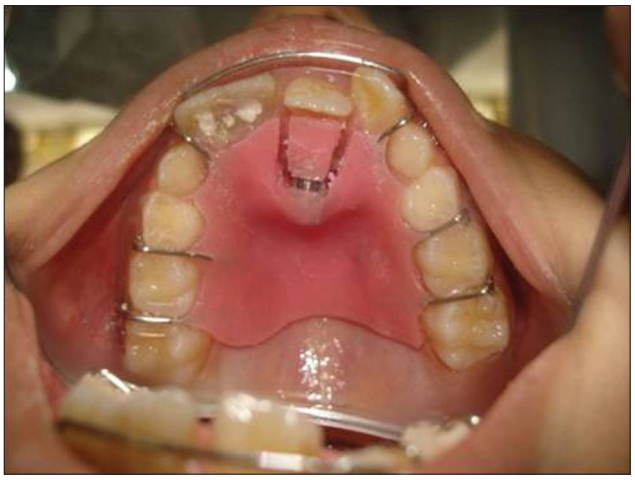

**Figure 5:** Orthodontic appliance for the correction of crossbite on contralateral side and for alignment of teeth

**Figure 7:** Hemisection of the affected tooth

calcification, extension, and fusion of the intervening areas, further separation of the cusps by the continuing multiplication of intervening cells is impossible.[13] Retardation of calcification by chemical or enzyme action would give time for growth and more cell divisions, which would result in a different ultimate morphology or expression.[13] Other causes include necrosis of epithelial tissue between two developing teeth, embryologic persistence of the interdental lamina between two germs, genetic predisposition, and environmental factors, such as thalidomide embryopathy, fetal alcohol exposure, or hypervitaminosis A of the pregnant mother.[4,7] Although the etiology is difficult to access in our case, pressure effect between central incisor and a supplemental tooth appears to be the most significant causative factor.

Isolated macrodontia may be the simple enlargement of all tooth structures, or it may be associated with morphologic anomalies.[14] Dichotomy of the tooth germ is the most common theory to explain the process of gemination or fusion causing macrodontia. The tooth bud splits into two, resulting in two teeth of equal or differently sized parts that may become fused or form one large abnormal tooth.[7]

Central morphologic anomalies are usually the result of developmental defects of dominant autosomal hereditary origin, attributable to trauma or produced by some infectious process or radiologic exposure.[4] It seems likely that these anomalies turn out to be primarily under genetic control, although not strongly heritable.[8,12] Whether the condition is autosomal dominant or recessive is currently unclear; nevertheless, there can be only minor penetrance.[5] Cross inheritance may produce large teeth in small jaws; in these cases, the teeth are crowded and malocclusion usually exists. [15]

As for the presence of talon cusp it has been suggested that this anomalous accessory cusp has a multifactorial etiology combining both genetic and environmental factors.[5] This occurs early in odontogenesis, that is, during the morphodifferentiation stage.[16] The pathogenesis is thought to be the proliferation and evagination of an area of the inner enamel epithelium and subjacent odontogenic mesenchyme into the dental organ.[8,16]

Macrodontia is a poorly researched and documented characteristic because very few data are available for

analysis. The published prevalence of macrodontia ranges from 0.03% to 1.9% while it can be as high as 3.6%.[5] True macrodontia of single tooth is an uncommon event.[17] The prevalence of macrodontia as a result of fusion or gemination in the primary dentition, ranges from 0.5% to 2.5%;[7] in the permanent dentition the prevalence is about 0.2%[7] to 0.3%[8] and may range up to 5%.[18] It is also reported that fusion of central and lateral incisors occurs more than that of lateral incisor and canine. Related to Mader's "two-tooth rule," the anomaly may represent a fusion between a normal tooth and a supernumerary tooth.[8]

Talon cusp is an unusual and relatively rare anomaly, which most frequently affects the maxillary permanent incisor.[8] Reports of its distribution according to gender, race, and location are conflicting in the literature[4] and is supposed to occur with a frequency of 0.04–10%.[16] A few reported cases of macrodontia due to fusion have been illustrated in Table 1.

Macrodontia out of fusion arise through the union of two normally separated tooth germs.[22] Three forms of macrodontia should be differentiated: true generalized, relative generalized, and macrodontia of single teeth.[6,14] Although true generalized macrodontia associated with hyperpituitarism[6] and suprarenal hyperfunction[15] is rare, relative generalized macrodontia is more common but poorly defined.[6]

*Fusion* (two separate tooth germs fused during formative stage) is the union by enamel and dentin (true fusion); or union by dentin and/or cementum (late fusion); and a late fusion by cementum is called a concrescence.[28] Depending on the stage of development at the time of union, fusion can be either complete or incomplete.[22]

Hattab *et al*[4,8] classified talon cusp as follows.
Type 1: Talon refers to a morphologically well-delineated additional cusp that predominantly projects from the palatal (or facial) surface of a primary or permanent anterior tooth and extends at least half the distance from the cementoenamel junction to the incisal edge.

Type 2: Semi-talon refers to an additional cusp of a millimeter or more extending less than half the distance from the cementoenamel junction to the incisal edge. It may blend with the palatal surface or stand away from the rest of the crown.

Type 3: Trace talon is an enlarged or prominent cingula and their variations, that is, conical, bifid, or tubercle like.

In the anterior region, the anomalous incisors manifest as structures resembling two teeth that have been joined together with a groove on at least the buccal surface and less commonly on the lingual surface and a notch on the incisal edge,[7] which is concomitant with the appearance of the incisal notch in our case. Clinically, the crowns of the teeth appear to be meld together, with a small groove between the mesial and distal sections.[4] Gemination results in a bifid crown, with the coronal halves appearing as mirror images, whereas fusion takes place at an angle causing the tooth to have a crooked appearance.[4] In addition, the geminated tooth may undergo complete gemination and instead manifest itself as two teeth

**Table 1: A few reported cases of macrodontia due to fusion**

| | Findings |
|---|---|
| Conklin[19] | Macrodontia in mandibular left lateral incisor |
| Henry[20] | Left central incisor with two well-formed roots. The crown was also larger than normal as also found in Macrodontia of single teeth. |
| Ekman-Westborg, Julin[13] | Macrodontia combined with postnormal occlusion, bimaxillary crowding, impaction of teeth, and crossbite between right upper and lower first molars. |
| Graubard[21] | Fusion of lower second and third molar and macrodontia of lower first molar in the same patient. |
| Hemmig[11] | Fusion of impacted third and fourth molars. |
| Ruprecht, Singer[17] | Macrodontia of mandibular left first premolar[7] |
| Goldberg, Gross, Rankow[22] | Fusion of mandibular second and third molars. |
| Veno, Mochizuki, Morimoto[23] | Fusion of maxillary central incisors. In another case they reported fusion of mandibular deciduous central incisors. Two secondary central incisor teeth were present.[8] |
| Mann, Dahlberg, Stewart[9] | Macrodontic and crenulated first permanent maxillary and mandibular and second deciduous molars. The adult permanent central incisor crowns were also large.[10] |
| Ballal[24] | An unusual case of double tooth, which was difficult to be diagnosed whether the double tooth was due to Fusion of a supernumerary tooth with the central incisor or was due to the gemination of the central incisor itself. |
| Grammatopoulos[25] | Fusion between a maxillary third molar and a paramolar. The extracted tooth had five separate roots. |
| Prabhakar, Kaur, Nadig[26] | Bilateral fusion of permanent mandibular incisors with talon cusp |
| Nagaveni, Umashanikara, Vidyullatha, Sreedevi, Radhika[27] | An unusual concurrent combination of multiple dental anomalies—talon cusp, dens invaginatus, short root anomaly, and macrodontia affecting both the crown and the root of the permanent mandibular left central incisor |

after undergoing a process called "twinning." When twinning occurs, there will be one more (supernumerary) tooth in the arch, and the two teeth are likely to be mirror images.[7] In cases where there is union of a normal tooth bud to a supernumerary tooth germ, the number of teeth is also normal and differentiation from gemination may be very difficult.[3] The number of teeth in our case remains unaffected as fusion appears to happen between a normal tooth (central incisor) and a supplemental tooth.

Radiographic examination reveals usually two separate canals in case of fusion,[4,5] as also seen in our case, whereas in germination there is usually one large conjoint root canal.[4,5] It is also reported that there is no essential variation from the histology seen in a tooth of normal size.[15] The histologic evaluation in this presented case could not be done as the anomalous tooth was retained.

Macrodontia may sometimes be found in primary teeth in Down syndrome and may be present in other syndromes, such as Langer–Giedion syndrome, KBG syndrome, and hemi-hypertrophy.[7] Macrodontic first permanent molars occur in Casamassimo syndrome, Down syndrome, otodental syndrome, globodontia, and taurodontia.[9] In addition, macrodontia as a result of fusion or gemination is seen more frequently in patients with cleft lip and/or palate and those affected by certain congenital cerebral disorders.[7] Macrodontia of anterior teeth, caused by fusion or gemination, may be associated with other dental anomalies, such as dens-in-dente, hypodontia, and supernumerary teeth, and nondental anomalies, such as syndactyly and nail disorders.[7] Trisomy 21, orodigitofacial syndrome, ectodermal dysplasia, Pierre–Robin syndrome, and certain situations involving labiopalatal fissures can be associated to dental fusion.[18] It may also occur with several syndromes, such as achondrodysplasia, chondroectodermal dysplasia, focal dermal hypoplasia, and osteopetrosis.[4] Talon cusp may be associated with Rubenstein–Taybi, Sturge–Weber, or Mohr syndrome,[8] incontinentia pigmenti achromians,[16] cleft lip and palate, hypomelanosis of Ito, Ellisvan–Creveld syndrome, and Alagille' s syndrome.[4] The medical history in our case was however noncontributory.

Clinical management of fused macrodontic incisors require a collaborative multidisciplinary treatment approach.[7] The traditional surgical treatment of macrodontia, or fused teeth involve either extractions of the affected dentition or resection or partial coronectomy. Usually included is subsequent root canal therapy before the placement of fixed prosthodontic coverage.[29] The extracted macrodontic incisor may also be replaced by autogenous transplantation of the supernumerary incisor (if it has better morphology and esthetics).[7] However, asymptomatic anterior fusion should be left alone unless problems arise with esthetics, spacing, and dental caries. Simple composite restorations can be used to camouflage and prevent caries developing in the fissures.[7,8]

David *et al*[29] described a nonendodontic coronal resection technique for fused and geminated vital teeth. On the contrary, hemisection was performed in our case after endodontic treatment. Some authors prefer orthodontic treatment before restoration of the treated tooth, as also was performed in our case in order to align the teeth in maxillary arch.[18]

The treatment of talon cusp requires careful clinical judgment and is dependent on its size and shape. Small talon cusps are usually asymptomatic and need no treatment. Large talon cusps may cause clinical problems[8] and require sequential grinding,[26] pit and fissure sealing, pulp therapy, restorative treatment, full crown coverage, and extraction of the affected tooth.[4] At the last appointment, to avoid postoperative sensitivity, this area should be covered with resin composite.[20] In our case, the talon cusp was sequentially ground and did not require any endodontic therapy as there was no pulp extension radiographically.

## CONCLUSIONS

Macrodontia appears to be more a textbook heading than a well-documented condition.[13] To the best of our knowledge, cases of fusion with talon cusp has been published in the English literature at three occasions, hence making this the fourth published case.

Macrodontia and visibly fused-like crowns of the permanent dentition may be esthetically unacceptable.[29] Hence, treatment planning of macrodontic incisors may be quite complex and timely management while seeking intervention from a multidisciplinary team is not only essential, but mandatory to prevent future complications.[7] There are different treatment approaches, including extraction with approximation and conversion crowning of suitable adjacent teeth, surgical division of the double teeth, selective grinding, replacement of the extracted teeth with an interim removable partial denture until they can be replaced with a fixed bridge or an implant.[30]

As a result, significantly improved facial appearance with a satisfactory smile, vastly improved occlusion, and increased self-esteem can be achieved.[31]

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