

An Unusual Odontome – The Silent Tormentor – A Physical Obstacle to Central Incisor Eruption – Report of a Rarest Case

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Abstract: Various developmental dental malformations can occur in the oral cavity leading to plethora of complications. Dental anomalies pertaining to tooth number, size, shape, structure and composition can form when environmental or genetic insult occurs during tooth organogenesis. Dental anomaly like supernumerary tooth or odontome are type of dental malformations when present can cause plethora of complications in children decreasing self-esteem of the patient. The aim of this research paper is to present a case showing an unusual occurrence of odontome associated with another dental anomaly causing impaction of the maxillary central incisor in an 11-year-old female Indian patient. Therefore, the present paper is first of its kind showing an uncommon development of odontome not reported so far in the dental literature representing a rarest dental entity.

Keywords: Cone-beam computed tomography scan, Impacted maxillary central incisor, Odontome, Radiographic examination, Supernumerary tooth, Tooth eruption disturbance.

Introduction

Odontomas are rare dental developmental anomalies pertaining to structure and composition of teeth observed in an individual. They are considered as tumor-like malformations or hamartomas composed of mixed dental tissues such as epithelial and mesenchymal cells. It is also explained that they are the most common odontogenic tumors, characterized by benign and slow growing tumors comprised of all dental tissues in different proportions with varying degrees of development [1]. The word 'odontome' was first framed by an author Pierre Paul Broca in 1867 [2]. He defined the term as 'tumors formed by the overgrowth or transitory of complete dental tissue consisting of enamel, dentin, cementum and pulp tissue. Odontomas are asymptomatic lesions, located within bone for many years without clinical manifestations or sometimes associated with missing permanent teeth or retained primary teeth [1]. However, other symptoms like numbness in the lower lip and swelling in the affected area are also reported. The exact etiology for development of odontomas is not known. Literature postulates possible interaction of genetic and environmental factors like infection, genetic mutation, inheritance and trauma as causative agents [3]. They are usually developed by an improper growth of completely differentiated epithelial and mesenchymal cells of odontogenic origin. Odontomas can develop in any age group and most commonly documented in second decade of life with no significant gender predilection. Presence of odontomas results in clinical problems such as delayed eruption or impaction of permanent teeth, displacement of roots or teeth, root resorption, congenital agenesis of teeth, delayed exfoliation of primary teeth and widening of follicular space [4]. In 2005, World Health Organization (WHO) classified odontomas into two types as compound and complex based on the radiographic and microscopic characteristics such as presence of odontogenic tissue [5] (Table 1).

As odontomas are usually asymptomatic, they are detected on conventional radiographic examination including either intraoral periapical or panoramic radiographs as an incidental finding [3]. Recently an advanced imaging techniques like cone-beam computed tomography has revolutionized in the arena of pediatric dentistry for diagnosis of unusual dental anomalies including odontomas to analyse their exact location, structure, relation with adjacent vital structures which helps in proper treatment planning [6-8]. The aim of the present research paper is to illustrate occurrence of an odontome causing impaction of the maxillary central incisor in an 11 year-old Indian male patient.

Table 1: WHO Classification of Odontomas [5]

Odontome Type	Description
Compound	Multiple irregular, small teeth like structures or denticles seen arranged in a more orderly pattern.
	Formed by many little tooth-like structures attached to each other and held together by a more or less complete connective tissue capsule.
	They are easily identifiable.
	Are usually small but large lesions containing up to 100 denticles is also present.
	Mostly occur in anterior region of jaw (61%).
	Occur mostly in the incisor, canine region of the maxilla.
	Are arranged in a radiating pattern around a central core of dental pulp.
Complex	Contains all dental tissues but occurs in a less organized pattern.
	Formed by a single amorphous mass of mature odontogenic tissues without any structural organization with tissues.
	Occurs in posterior region of dental arch (34%).
	Occurs mostly in the premolar and molar region of the mandible.

Case Report

An 11-year-old female patient with Indian ethnicity reported to a private dental clinic complaining of missing tooth in the upper front region of the oral cavity. Patient gave a history of self-exfoliation of milk tooth almost 3 years back and waited for normal eruption of the permanent tooth. Adjacent contralateral tooth was erupted at normal time following primary tooth exfoliation. Patient was moderately built, well nourished, normal behaviour and did not show any signs and symptoms of metabolic, systemic or syndromic disorders. There was no history of trauma to the anterior region. On intraoral examination mixed dentition was observed with clinical missing of 21. Maxillary right central incisor was erupted in proper position. A soft tissue bulge was observed in 21 region (Figure 1). To confirm the presence of 21, patient was subjected to conventional intraoral periapical radiographic examination which showed an impacted 21 located high above (almost 14 -15 mm) from the incisal edge of 11 (Figure 2). In place of 21, irregular masses of different radio-density surrounding a small tooth like structure located in horizontal direction were observed. Small tooth like structure exhibited crown and root portion containing enamel, dentin and pulp. To locate exact location of impacted irregular masses, CBCT scan was performed (Figure 3 and 4). CBCT imaging revealed horizontally located 21 in labio-lingual direction with crown towards labial direction and root towards lingual direction. Two-third portion of 21 root was completed with apical one-third root found with an open apex. An agglomerate masses with small tooth like structure was observed surround by 6 to 7 denticles which appeared as 'cluster of grapes' with different sizes (Figure 3 and 4). Considering intraoral features, radiographic findings and literature search, the case was diagnosed as 'horizontally impacted supernumerary mesiodens associated with compound odontome with multiple denticles.' Patient was explained about existing dental malformation and surgical removal of both odontome and mesiodens was advised and carried out under local anesthesia. Regarding treatment of impacted 21, as its root was still not completed; 'wait and watch' protocol was advised for 3 months for its self- eruption. Following 3 months if the tooth found to be not erupting, then orthodontic pulling of impacted 21 was advised. Therefore, patient is still under observation.



Figure 1: Intra-oral photograph showing soft tissue bulge (yellow arrow) in relation to clinically missing 21

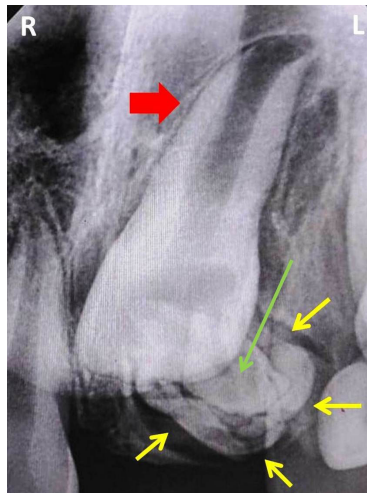


Figure 2: Intra-oral periapical radiograph showing impacted 21 (red arrow), horizontally impacted mesiodens with small crown and root (green arrow) and compound odontome surrounding the mesiodens (Yellow arrows).



Figure 3: CBCT image showing impacted 21 in labio-lingual direction (yellow arrow), horizontally impacted supernumerary mesiodens (red arrow) surrounded by 6 to 7 denticles of compound odontome (white arrow).

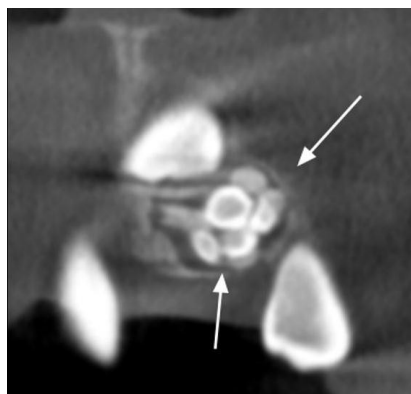


Figure 4: Occlusal view of CBCT image exhibiting appearance of denticles as 'cluster of grapes' pattern (white arrows).

Discussion

Presence of maxillary incisors especially central incisor is essential for facial esthetics, phonetics and mastication. Absence or disturbance in eruption of central incisor leads to decreased self-image and psychological burden on child and parents as well. Maxillary central incisor normally erupts at the age of 7 to 8 years. Failure in its eruption by this age raises 'queries' on congenital agenesis, obstruction from a physical barrier for eruption, presence of hidden dental anomalies, disturbance in eruption pattern or associated cysts or tumors [8,9]. Therefore when a clinician comes across with such scenario in children should always rule out those queries and comprehensive examination has to be performed. Moreover, parents of children must be aware of occurrence of such conditions and consult their respective dentist for immediate treatment. Various factors have been listed in articles for the delayed eruption or missing of maxillary central incisor [3,10,11]. The most common dental condition responsible for delayed eruption of central/lateral incisor is the development of dental malformations in the place of incisors [2]. Extensive review of dental anomalies' literature revealed numerous case reports on occurrence of mesiodens and odontomas in the maxillary anterior region with most common being the mesiodens [12-14]. Nagaveni et al reported ample of case reports, case series and survey on development of mesiodens or supernumerary tooth in the maxillary anterior region obstructing eruption of the central incisor or associated with cysts among Indian population [12,18]. All cases were successfully managed by surgically extracting the culprit mesiodens, enucleation of cyst followed by orthodontic management of impacted central incisors.

Publications showing prevalence of odontomas in different population around the globe are scarce. There are only few research papers representing case series documented with 50 to 70 cases [3,19,20]. A retrospective study [19] from Taiwan identified 81 odontomas in 81 patients with mean age of 18 years (45 females and 36 males) using previous medical records. There were 62 compound and 19 complex odontomas. There was increased frequency of occurrence in the maxilla (70%) affecting the anterior maxilla (62%). Sixty four (79%) of the 81 odontomas were associated with 80 impacted teeth consisting of 2 primary teeth, 7 supernumerary teeth and 71 permanent teeth [19]. Out of 71 impacted teeth, the maxillary central incisor (27%) was most commonly affected followed by the maxillary canine (26%) and mandibular canine (24%). Following histologic examination, it was found that presence of dentin in 100%, enamel matrix in 90% of cases, pulp tissue in 96%, fibrous capsule in 93%, cementum in 88%, reduced enamel epithelium in 86%, ghost cells in 83% cases and nests of odontogenic epithelium in 58% of cases. Dentigerous cyst was associated with 9% of odontomas [19]. Another case series published by Nagaveni et al [3] reported three cases of odontome formation in Indian patients. In this report, age of the patient ranged from 9 to 26 years. In two patients the clinical finding observed was delay in eruption of the upper central and lateral incisors. Among three odontomas, two cases were complex type and one was of compound odontome. In two cases, surgical removal of odontome was carried out [3].

Articles published on the domain 'odontome' illustrated agglomerated masses of small radiopaque structure diagnosed as either composite or compound odontome [5]. The present case is unique as it was associated with another anomaly like supernumerary tooth called 'mesiodens' associated with compound odontome consisting of six to seven denticles. This pattern of odontome appearance is not reported so far in the literature as per best of authors' current knowledge. However, there are reports showing occurrence of dens in dente/dens invaginatus within a mesiodens or mesiodens with talon cusp or mesiodens with multiple lobe or mesiodens associated with cyst [14-18].

In the present case, main clinical manifestation observed was clinically missing or delayed eruption of the maxillary left central incisor coinciding with previous reports. The primary maxillary central incisor was exfoliated 2 years back. The adjacent contralateral central incisor was erupted almost 3 years back. In the present case a compound odontome associated with centrally located mesiodens supernumerary tooth occurred in the anterior part of the maxilla in the central incisor position. Although literature shows most common occurrence of compound odontome in the right side of the arch, but in the current case it occurred on the left side. The number of denticles present in the compound odontoma varies in number either as single or multiple. In present case, odontome consisted of 6-7 denticles surrounding central located, horizontally impacted mesiodens. An Indian case report showed occurrence of odontome with 70 denticles and impacted lateral incisor [10]. All 70 denticles were surgically removed and the impacted lateral incisor was orthodontically moved to its position in the oral cavity [10]. Another case report illustrated a giant compound odontome comprised of more than 100 denticles in the right mandibular canine region [11]. Sharma et al found 37 denticles [13] and Sharifi Rayeni et al [19] observed 62 denticles of compound odontome from the right maxillary region.

The degree of morpho-differentiation found in odontomas varies from cases to cases. Some cases were found with predominant calcified matrix and some with islands of pulp tissue in association with cords and buds [20,21]. Pippi did an extensive research to find out whether similarities supporting a unified explanation exist for odontomas and supernumerary teeth [22].

The obtained data showed some interesting similarities between supernumerary teeth and odontomas regarding their topographic distribution and pathologic manifestations. It is also evident that there is common origin for odontomas and supernumerary teeth having the expression of same odontogenic hyper-productive process with different gradients of morpho-differentiation in relation to a time and site related signalling pathway of molecules and factors, genetically, non-genetically and epigenetically determined and modulated factors [22]. This same phenomenon can be applied in the present case regarding the development of both a supernumerary tooth and compound odontome.

Diagnosis of odontomas is usually made by conventional radiographs using either intraoral periapical or panoramic radiographs [3]. Recently cone beam computed tomography has played a significant role in the diagnosis and identification of odontomas thereby helping in treatment planning [6,7]. Additionally, modern high-resolution micro-computed tomography (micro-CT) is increasingly employed in dental research. This new technology enables three-dimensional analysis with better spatial resolution compared with CBCT [23]. Radiographically odontomas appear as well-circumscribed radiopaque masses and is particular for each type of odontomas. They typically appear as radiopaque masses with a radiolucent rim on intraoral periapical radiographs. They have a lobulated appearance with small radiolucent spaces within the radiopaque mass giving them a 'cluster of grapes' appearance [23,24]. The small, tooth-like structures within the compound odontome can also be seen as separate entities in the surrounding radiolucent zone. The similar 'cluster of grapes' appearance was observed in the present case following CBCT imaging (Figure 3). Mesiodens was placed in horizontal orientation and exhibited small sized crown and root with enamel, dentine and pulp tissue.

The suggested treatment protocol for odontome always includes surgical removal although they are asymptomatic and do not grow/expand in size [24,25]. This is because they are considered as silent tormentors as they act as physical barrier thereby obstructing the path of tooth eruption. Literature shows no recurrence of odontomas after surgical extraction following 1 to 15 years of postoperative period. In the present case, presence of compound odontome associated with impacted mesiodens totally hindered the eruption of permanent left central incisor leading to its impaction. Therefore surgical removal of both odontome and a supernumerary mesiodens tooth was carried out under local anesthesia to facilitate faster eruption of the central incisor. Following extraction procedure patient was kept under observation for complete eruption of the central incisor for a period of three months. After three months if the central incisor appears not erupting, then a treatment plan consisting of orthodontic movement of the tooth to its position in the oral cavity was decided. Hence patient is still under observation at this point of paper publication.

Conclusion

Odontomas and impacted supernumerary teeth are asymptomatic unless associated with clinical symptoms. They are always detected on routine radiographic examination as an incidental finding. Therefore, early detection of odontomas and supernumerary tooth along with their proper treatment avoids management of future complications. Moreover, both parents and clinicians must be aware of occurrence of dental anomalies like odontome or supernumerary mesiodens as they cause delay or impaction of the permanent tooth resulting esthetic impairment in children. Additionally, academic researcher should always make an effort to identify, record and diagnose unusual dental variations occurring in human being to frame new diagnostic, therapeutic, preventive and classification guidelines to provide optimal dental care.

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