

## CASE REPORT



# Compound odontoma causing impaction of permanent maxillary incisors with associated ghost cell keratinization: A case report

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## Abstract

Many times compound odontomas are found associated with unerupted teeth. The canines, followed by maxillary central incisors and third molars are the major teeth which are impacted by odontomas. Herewith, this paper reports a case of compound odontoma associated with impacted permanent right maxillary central and lateral incisors. An 11-year-old girl reported with a chief complaint of retained primary tooth. Intraoral examination revealed that primary maxillary right central and lateral incisors were retained while permanent maxillary left central and lateral incisors were erupted and well accommodated. A non-specific swelling was there on labial aspect of S1 and S2 tooth region, which was measuring about 0.5 cm × 1 cm, enlarging mesiodistally from distal of permanent left central incisor tooth to distal aspect of over retained right primary lateral incisor. Digital orthopantomograph showed calcified masses in the form of tooth-like structures present above the root apex and distal aspect of S1 and S2, causing root resorption. On the source of the past, clinical and radiographic assessment a final opinion of compound odontoma was made.

## Introduction

Odontoma is defined as calcifying benign odontogenic tumor composed of various tooth tissues such as enamel, dentin, pulp, and cementum and representing the second most common odontogenic tumor of the jaw bones.<sup>[1]</sup> These are also considered to be developmental anomalies consequential from the growth of entirely differentiated epithelial and mesenchymal cells that confers to ameloblasts and odontoblasts.<sup>[2]</sup>

The etiology is not obvious, even though local trauma, genetic factors, and infection have been recommended as possible causes.<sup>[3]</sup> The pathogenesis of odontomas is thought to be associated with trauma during development of primary dentition, hereditary anomalies (Gardner's syndrome, Hermann's syndrome, basal cell nevus syndrome), odontoblastic hyperactivity or alterations of the genetic mechanisms accountable for scheming dental growth.<sup>[4,5]</sup>

Hitchin recommended that odontomas are moreover inherited or owing to an altered gene or intervention with genetic control of tooth growth postnatally. Heredity is a probable issue, and constant lamina might be the hidden hereditary developmental anomaly. Further, it was reported that the

investigational production of such type of lesion in rat caused by traumatic damage.<sup>[6]</sup>

According to WHO's histological classification of odontogenic tumors of the oral cavity, there are two recognized histological types of odontomes: Compound odontoma and complex odontoma. Compound odontoma appears as numerous, sometimes hundreds of small teeth while complex odontoma appears as a single amorphous mass enclosed in a fibrous capsule.<sup>[7]</sup> Compound odontomas might be diagnosed at some age however they are frequently detected for the duration of the first two decades of life. Many times compound odontomas are found associated with unerupted teeth. The canines, followed by maxillary central incisors and third molars are the most periodic teeth impacted with odontomas.<sup>[8]</sup> Herewith, this paper reports a case of compound odontoma associated with impacted permanent right maxillary central and lateral incisors.

## Case Report

An 11-year-old girl reported at Department of Pedodontics, Navodaya Dental College and Hospital, Raichur with a chief

complaint of retained primary tooth. On extraoral examination, no noticeable disfigurement of the face was noted. Intraoral examination revealed that primary maxillary right central and lateral incisors were retained while permanent maxillary left central and lateral incisors were erupted and well accommodated [Figure 1a]. An ill-defined swelling was there lying on labial part of S1 and S2, measuring about 0.5 cm × 1 cm, extending mesiodistally as of distal side to permanent left central incisor to distal of over retained right primary lateral incisor. Superior-inferiorly it extended roughly 1 mm over the marginal gingiva in relation to S1 to the vestibular depth. However, the overlying mucosa was somewhat blanched. The patient was well and in good health medically.

On palpation, swelling was bony firm to hard in consistency, not tender, ill-defined, which was fixed to the underlying tissue, and not mobile. Further palatal aspect in relation to S1 and S2 also showed a mild ill-defined swelling. Patient's family history was non-contributory. Clinical differential diagnoses that can be included were ameloblastic fibro-odontoma, adenomatoid odontogenic tumor, calcifying odontogenic cyst.

Periapical radiographic evaluation of the upper anterior segment revealed the presence of multiple irregular masses of calcified tooth-like structures present near the roots of the maxillary right primary central and lateral incisors [Figure 2a]. Digital orthopantomograph showed calcified masses in the form of tooth-like structures present above the root apex and distal aspect of S1 and S2, leading to root resorption. Root of these tooth-like structures was not clearly visible [Figure 2b]. The radiographic differential diagnoses included are odontoma, ameloblastic fibro-odontoma, and ameloblastic fibro-dentinoma.

On the source of the past, clinical and radiographic assessment a final opinion of compound odontoma was made.

The patient was arranged under local anesthesia, i.e. 2% lignocaine - 1:80,000 adrenaline for surgical excision. A crevicular incision was organized in relative to labial part of S1, S2, and S3, further by two diagonal relieving incisions. To make possible correct way into the lesion, S1 and S2 be extracted. The mucoperiosteal flap was elevated towards the side aspect of maxilla, which exposed the occurrence of merged bunch of denticles [Figure 1b]. Elimination of the calcified collection was made without injuring adjacent unerupted permanent teeth. The extraction socket was normal. The region was curetted smoothly

and pointed bony borders were smoothed off, the region was cleaned with normal saline solution, and next the flap positioned back and held in place with the aid of 3-0 resorbable suture.

Macroscopically, the specimen consisted of numerous pieces of asymmetrical denticles [Figure 3]. The recovery was unremarkable. The patient was recalled for removal of sutures after 1 week. Histological sections showed tooth-like collection of dental hard and soft tissue together with cementum, dentine, enamel matrix and pulpal tissue in fibrous connective tissue stroma [Figure 4a and b]. Only some areas of enamel matrix by way of ghost cell keratinization were also seen.

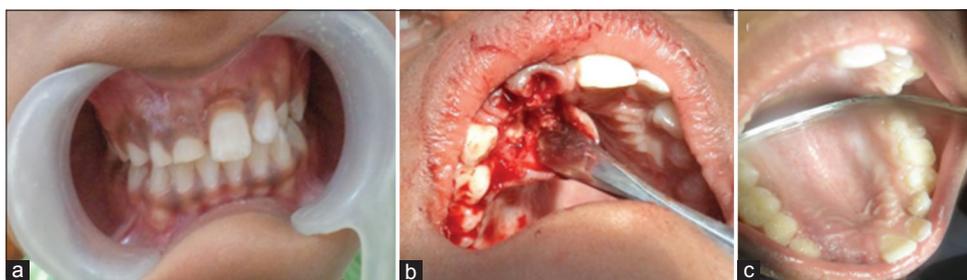
The patient's parents were advised assessment follow-up meeting once in 3 months for 12 months was completed in order to admit the eruption of unerupted permanent incisors and to observe the recurrence of the odontoma. To date patient was stable, and no recurrence was noticed [Figures 1c and 2c].

## Discussion

Odontomas are typically seen in children of school-age, and thus the average age at the instance of diagnosis was 14 years. Further, the majority of compound odontoma cases (74.3%) were diagnosed before the age of 20 years, during routine radiographic examination, occurring frequently in the anterior maxillary area, devoid of some gender predisposition.<sup>[9]</sup> The present case was also seen in 11-year-old girl affecting right maxillary anterior region.

Clinically, odontomas are asymptomatic; frequently stay small, hardly ever exceed the diameter of the tooth. Infrequently they do turn into large and might create growth of bone with resultant facial asymmetry. Signs and symptoms connected consist of retained deciduous teeth, impacted teeth, swelling and occurrence of infection. The major location for impacted teeth connected with odontomes is the anterior maxilla. Usually, compound odontoma (intraosseous) is situated at the periapical area of primary tooth and permanent tooth crown avoiding the eruption of the last.<sup>[10]</sup> Similarly, the present case was asymptomatic, present in maxillary anterior region, associated with retention of primary teeth and impaction of permanent teeth.

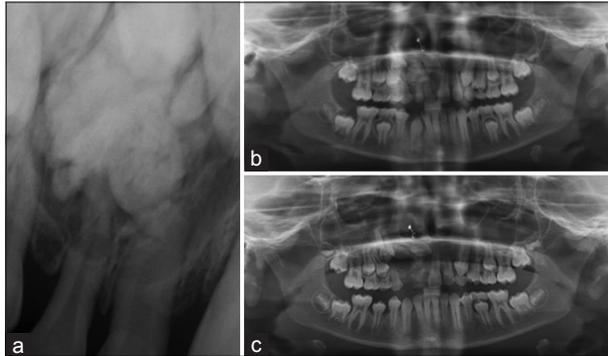
Compound odontomas are mostly of three patterns: (1) Denticular type - Collected with two or more detached denticles, showing crown with a root or Hertwig epithelial root



**Figure 1:** (a) Clinical photograph showing retention of primary right central and lateral incisors, (b) clinical photograph showing reflected mucoperiosteum and exposed calcified mass, (c) follow-up photograph showing no signs of recurrence

sheath with a allocation of dental hard tissues analogous to that originate in a tooth, (2) particulate type - Collected with two or more split masses of particles bearing no macroscopic similarity to tooth and consisting of firm dental tissues abnormally set, (3) denticulo particulate type – within this category, denticles and conglomerate areas are there side by side.<sup>[7]</sup>

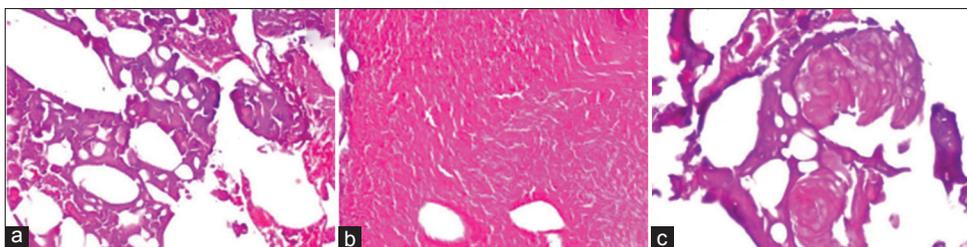
Radiographically, odontomas present as a well-defined, frequently unilocular lesion, which enclose several radiopaque,



**Figure 2:** (a) Intraoral periapical radiograph of maxillary right front region showing tooth-like structures and causing root resorption of 51 and 52, (b) pre-operative panoramic radiograph showing multiple radio-opaque masses in the periapical tooth region of 51 and 52, (c) follow-up post-operative panoramic radiograph showing no signs of recurrence



**Figure 3:** The excised gross specimen showing multiple tooth-like denticles along with extracted 51 and 52 primary teeth



**Figure 4:** (a) Hematoxylin and eosin stained decalcified section showing hematoxyphilic areas resembling enamel matrix ( $\times 200$ ), (b) eosinophilic areas with dentinal tubules resembling dentin ( $\times 200$ ) and (c) hematoxylin and eosin stained decalcified section showing enamel matrix with areas of ghost cell keratinization ( $\times 200$ )

miniature tooth-like structures recognized as denticles located in the bone, except with a density that is superior than bone and equal to or better than that of a tooth. Radiolucent halo, classically bordered by a slight sclerotic line, backdrops the radiopacity. They are discovered on radiographs either incidentally or in search of a cause for a missing tooth.<sup>[8,10]</sup> The present case also showed radio-opaque calcified masses in the form of tooth-like structures present over the root apex and to the distal aspect of 51 and 52, causing root resorption.

Microscopic features of complex odontoma contain all mature elements of dental structure: Dentin, dentinoid, enamel, enamel matrix, pulp tissue, and cementum. These are laid down in an organized fashion, which has a resemblance of normal tooth arrangement. The mass of hard dental structure is surrounded by a free, myxoid connective tissue through odontogenic epithelial cell rests which represents normal dental follicular tissue. Immature odontomas have little hard tissue formation, consisting of weak pulp tissues, dentin, and enamel matrix.<sup>[11]</sup> Similarly, the present case also showed enamel, dentin and pulp in myxoid connective tissue stroma.

Ghost cell keratinization is infrequently seen in the enamel forming cells of a few odontomas. Surrounding hard tissue which gets calcified leads to reduced oxygen supply by walling-off effect which in turn leads to metaplastic transformation of odontogenic epithelium leading to cell death and keratinization.<sup>[8]</sup> Few areas of ghost cell keratinization within the enamel matrix were seen in this present case also.

Odontomas are treated by traditional surgical elimination, and there is small likelihood of recurrence. It has been recommended that all specimens ought to be analyzed by an oral pathologist for histopathologic assessment. Appropriate patient care must include cautious clinical and radiological follow-up. When odontomas are associated with unerupted teeth, orthodontic traction of the impacted tooth almost immediately subsequent to removal of the lesion might be desired.<sup>[12]</sup> Similarly in the present case surgical removal of odontoma was done. Further the possibility of spontaneous eruption of permanent incisors after removal of odontoma cannot be ruled out; so it was decided to keep the patient under periodic observation for next 12 months, before any active orthodontic intervention.

Odontomas are habitually related with complications such as impaction, malpositioning, aplasia and devitalization of

adjoining teeth. As a result of the odontogenic nature, including epithelial and mesenchymal tissue, odontomas can lead to cystic transformation into dentigerous cyst. This cyst results from the cystic degeneration of enamel organ after partial or total development of the crown, cystic transformation of the follicle associated with the unerupted tooth may also occur when its eruption is impeded by the odontoma. Appropriate discovery and surgical enucleation of odontoma followed by curettage is suggested to avoid complications such as tooth loss, cystic changes, bone expansion and delayed eruption of permanent teeth.<sup>[8]</sup>

## Conclusion

Diagnosis of odontomas is apparently associated with age and location. Therefore, lesions from incisor locations are diagnosed and treated at an earlier age than those from the canine or third molar regions. Odontomas are more common associated with impacted teeth and rarely erupt into the oral cavity and can even give rise to inflammation, pain, infection and malocclusion when they erupt into the oral cavity. Therefore, early detection and treatment of odontomas could increase the possibility of preservation of the impacted permanent teeth. Thus, this paper highlights the complications of an odontoma as a probable source of over preservation of a primary tooth, as well as impaction of permanent incisors.

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