



A Case Report of Symptomatic Compound Composite Odontome Associated with Unerupted Premolar: A Brief Literature Review

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Received: April 21, 2018; **Published:** June 09, 2018

Abstract

Odontomas are nonaggressive, hamartomatous developmental malformations composed of mature tooth substances and may be compound or complex depending on the extent of morphodifferentiation or on their resemblance to normal teeth. The majority of odontomas are asymptomatic and discovered incidentally; however, may be accompanied with pain and swelling as secondary complaints in some cases. Hence, such type of lesion may cause disturbance in adjacent teeth eruption pattern, like impaction or delayed eruption or retained deciduous dentition. For this reason, a conservative surgical excision is needed as a treatment plan. Based on clinical, radiographic, histopathological features, a case report of compound composite odontoma is being discussed in this article.

Keywords: Odontoma; Hamartoma; Tooth Like Elements; Odontogenic Tumors

Introduction

The most common odontogenic tumors of the jaws are odontomas which constitute 22% of all odontogenic tumors [1]. In second edition of WHO histologic typing, odontomas are classified under the heading of tumors containing odontogenic epithelium with odontogenic ectomesenchyme, with or without dental hard tissue formation [2]. Hard tissue formation is mainly made up of enamel and dentin, but they also have different amount of cementum and pulpal tissue [3]. Like teeth, once fully calcified they do not develop further [4].

In 1867, the first person to give the term odontome was Paul Broca. He later defined odontome as tumors formed by the overgrowth of transitory or complete dental tissue [2].

By the definition, odontoma is referred as any tumor of odontogenic in origin [1]. Due to the growth of completely differentiated epithelial and mesenchymal cells the odontomas are supposed to be a developmental anomaly [1]. This differentiation gives rise to ameloblasts and odontoblasts functions which in turn form different amount of enamel, dentin and pulpal tissue [5,7]. This enamel and dentin formed were in an abnormal pattern because the odontogenic cells failed to reach the normal stage of tooth development i.e. morphodifferentiation [5]. Hence, due to this reason they are regarded as a developmental anomaly rather than a true neoplasm.

Early in 1974, Shafer, Hine and Levy illustrated odontomas to be odontogenic tumors but recently they supported and accepted it to be a hamartoma [8].

Compound and complex odontomas are the two variety of odontomas [6]. In 2005 World Health Organization (WHO) defined compound odontomas as a "tumor like malformation with varying numbers of tooth-like structures" [1]. Since odontomas are regarded as mixed odontogenic tumors, the cells and tissues can appear either normal or insufficient in structure which may vary in the dental tissues formation. Thus, in compound odontoma minute tooth like structures are formed and in complex odontoma dental hard tissues are haphazardly arranged [2,3]. In the present article we have reported a case of a compound odontoma in a 13-year-old patient, which is associated with unerupted permanent premolar.

Case Report

A female patient around 13 years old came with a chief complaint of pain in mandibular right posterior region to the Department of Pedodontics. She complained of intermittent pain which gradually increased in intensity since last 3 days especially on chewing food. The medical and family history of the patient was inconclusive. Intraoral examination revealed proximal caries on both mesial and distal side of 75 which was associated with pain on percussion and no history of trauma has been reported. No abnormality was noted on extra oral examination.

IOPA of 75 was taken which showed radiopacity in association with mesial root & crown of unerupted 2nd premolar. Distinct radiopacity was seen. Radiopacity is surrounded by radiolucent rim with sclerotic border.

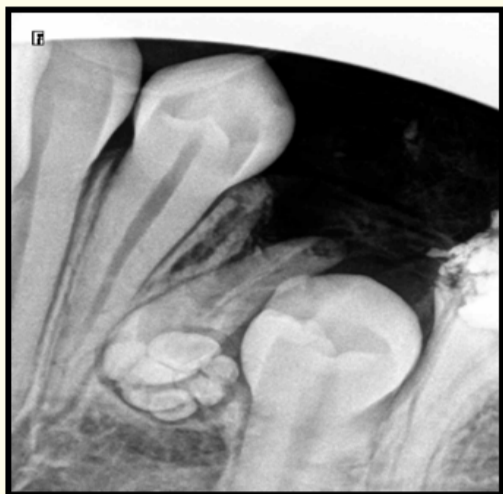


Figure 1: IOPA showing radiopacity in association with mesial root and crown of unerupted 2nd premolar. Distinct radiopacity was seen. Radiopacity is surrounded by radiolucent rim with sclerotic border.

Based on histopathological examination provisional diagnosis of compound odontome was confirmed which showed transverse and longitudinally cut dentinal tubules along with amorphous basophilic masses resembling cementum. Numerous trabeculae with osteocytes in lacunae were noted in few sections.

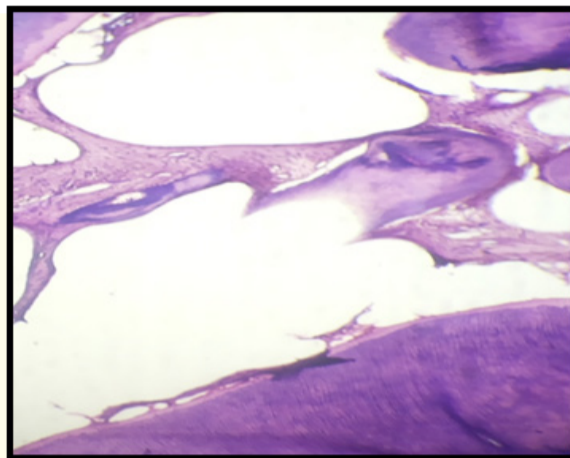


Figure 3: H and E stain section under 40x objective shows irregularly arranged dental hard tissues with pulp-like tissues. Longitudinal section of dentinal tubules is noted.

A provisional diagnosis of odontoma was made based on clinical and radiological findings and then surgical excision was planned for the treatment. By infiltrating the incisive foramen, anaesthesia to nasopalatine nerve was given. In order to expose the periapical lesion, the buccal full thickness triangular mucoperiosteal flap was raised. The tooth underneath was left in place undisturbed for further eruption into the oral cavity. Postoperatively after one-week sutures were removed.

The excised specimen was sent to histopathological department containing tooth no. 75 (FDI) with mesiolingual root and multiple bits measuring all app. 1.5 x 1 x 0.1 cm with hard consistency, whitish in color, irregular surface and border.



Figure 2: Multiple bits of containing tooth no. 75 (FDI) with mesiolingual root and multiple bits measuring all app. 1.5x1x0.1cm with hard consistency, whitish color, irregular surface and border.

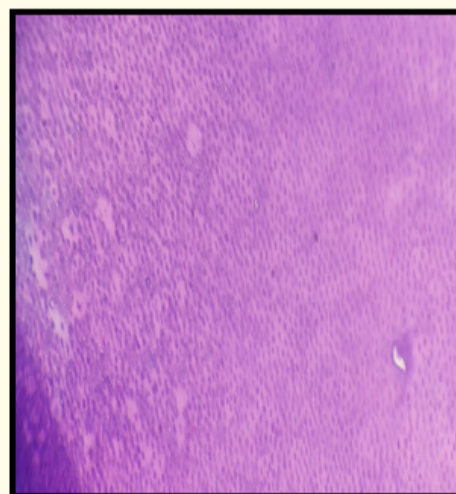


Figure 4: H and E stain section under 40x objective shows transverse section of dentinal tubules.

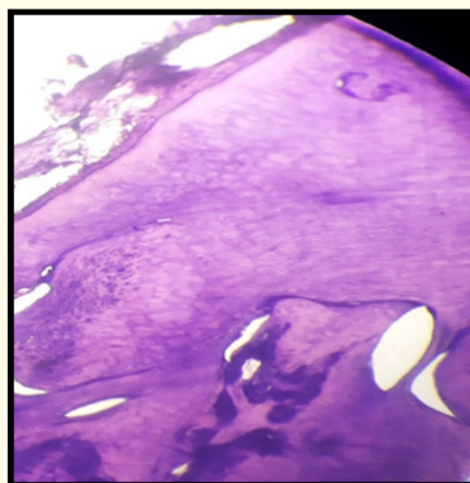


Figure 5: H and E stain section under 40x objective shows area of amorphous basophilic masses resembling cementum.

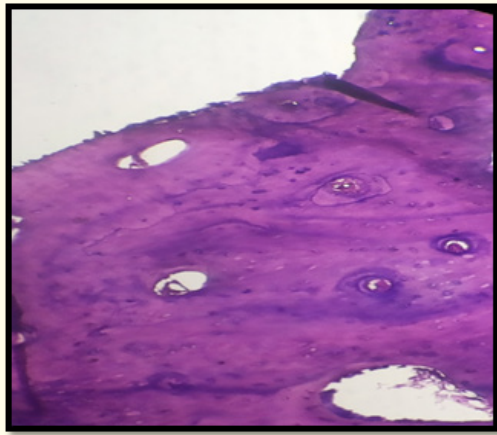


Figure 6: H and E stain section under 40x objective shows numerous trabeculae with osteocytes in lacunae.

Based on clinicopathologic correlation, a final diagnosis of “Compound Composite Odontoma” was confirmed

Discussion

There have been many reports of odontomas. The larger amount of odontogenic tumors in which all the structures that form dental tissues reported are the odontomas [12]. The common clinical presentation for an odontoma is the alliance with impacted or unerupted or retained primary teeth [13]. The case presentation described above was in compliance with the finding.

In 1866 after Brocas classified odontomas in proportion to the stages of tooth development and also numerous classifications were given based on the structural tissues from which the tumor had arisen [14].

World Health Organization (WHO) gave the most common classification. The four lesions that contain normal appearance of enamel and dentine are described in classification given by WHO [15]. They are as follows:

Ameloblastic fibro-odontoma, Odonto-ameloblastoma, Complex odontoma, Compound odontoma.

An additional classification for odontomas given by H. M. Worth was derived from three layers which include, ectoderm termed as Enameloma, mesoderm termed as dentinoma / cementoma and from both ectoderm and mesoderm termed as compound or composite odontome / germinated / dilated odontoma [16].

Two types of odontoma are documented based on gross, radiologic and microscopic features, which includes compound and complex [17-20].

In 1914, Gabell, James and Payne grouped odontome into three types based on their development like epithelial, epithelial and mesodermal and connective tissue [21,22].

According to their position within the jaws: (a) Intraosseous (erupted odontoma), (b) Extraosseous or peripheral odontomas [23,24].

Thomas and Goldman (1946) classification of odontomes: Germinated composite odontomes - (two or more, more or less well-developed teeth fused together), Compound composite odontomes-(made up of more or less rudimentary teeth), Complex composite odontomes-(calcified structure, which bears no great resemblance to the normal anatomical arrangement of dental tissues), Dilated odontomes-(the crown or root part of tooth shows marked enlargement), Cystic odontomes- (normally encapsulated by fibrous connective tissue) [25,26].

The term composite was eliminated by Z Gorlin, et al. and he classified odontomas as only compound or complex. But still Complex composite and Compound composite odontome are the two fundamental types of odontoma [20,22].

Vibha singh, et al. (2010) reported a new type of odontoma known as hybrid odontoma [1].

In 1952 Robinson restricted the term odontome for those tumors which aroused from epithelial and mesenchymal dental forming tissues [27].

There are three main types of compound odontomas, as classified according to Gravey, et al. Denticular type: Composed of two or more separate denticles, each having a crown and a root or epithelial sheath of Hertwig with a distribution of dental hard tissue comparable to that found in a tooth, Particulate type: Composed of two or more separate masses or particles bearing no macroscopic resemblance to a tooth and consisting of hard dental tissues abnormally arranged, Denticulo particulate type: Denticles and conglomerate masses or particles are present side by side [28].

Most odontomas are detected during the first two decades of life [3,10]. One study analyzed 396 cases and showed that diagnosis usually happens between ages 11 and 15 years [29]. Another study comprising 149 cases concluded that the lesions are detected most often during the second decade of life [21]. Frequently impacted by odontoma are canines, followed by upper central incisors and third molars [29]. This was not in accordance with our case. They develop and mature while the corresponding teeth are forming and cease development when the associated teeth complete development [30,31].

They are painless and slow growing lesions which were opposite to our findings. Garcia-Consuegra, et al. (2000) reported pain and inflammation in association with odontoma in only 4% of Spanish patients [32] which usually varies between 1 and 2 cm in diameter [9] and slightly more common in males (59%) compared to females (41%). Another study shown occurrence of the compound odontoma in the maxilla (67%) and in mandible (33%), with a marked predilection for the anterior maxillary region i.e. (61%) [10,33,34].

Both the odontomas had a frequency of occurring on the right side of jaw than on the left with 62% comprising for compound type and 68% comprising for complex type [6]. In incisor cuspid region of upper jaw the compound composite odontome occurred more commonly and in molar and premolar region of mandible complex odontomas occurred more [14,35,36]. Some authors like

Bland Sutton in 1988 reported incidence of both the types of odontoma in different areas like maxillary sinus [14,37], mandibular ramus [38], subcondylar region [39] or mental foramen [37], mid palate [40] and the middle ear [41]. Hermann (1957) reported a case of compound composite odontoma which consisted 2,000 denticles [14].

Odontomas commonly occur in permanent dentition and are rarely reported in association with primary teeth [42,43]. In 1949 Tratman contemplated that odontomas are not prone to deciduous dentition [44].

The definite cause for formation of odontoma remains still unknown [6]. For controlling the dental development, various pathological conditions like trauma, inflammation or infection, mature ameloblasts, dental lamina remnants, hereditary anomalies like Gardeners syndrome, odontoblastic hyperactivity, any changes in genetic component are responsible [45].

Due to growth pressures and inadequate space the odontomas are liable to occur. The authors like Euler (1939) [46] and Atkinson (1949) [47] have an opinion that these growth pressures may be important for few composite odontomas. Later on, Hitchin and Ferguson [48] thought that the pressure effect might have arisen from a developing lower premolar germ of an inherited large crown which is embraced by the roots of its deciduous predecessor.

Infection coming from deciduous predecessor may also be a causative factor, but a generalized infection may be of significance. Odontoma can also occur if prenatal dentition is infected with treponema palladium, during pregnancy if mothers have been infected with rubella, in acute maxillitis of infancy, in acute pyogenic infection of whole maxilla in 6^{1/2} years old patient [46]. Thus, when associated with any infection odontoma may occur due to division of a tooth germ or it may impede with tooth development which could pathologically affect the genetic control of tooth formation.

Based on this Torreti, *et al.* [49] recommended that these particular cells have the potential of developing tumors with different appearance and content.

Fijerskov [50] established that dental lamina remnants of retained tooth contain epithelial island which undergoes proliferation to form odontoma while others showed degeneration to form cystic cavity that encloses the tooth for which the spur may have a genetic defect in tooth formation.

Later on, when the buds get divided into numerous particles they may develop alone to form tooth like structures and without such division they may form haphazard structures of dental tissues that form a complex odontoma [17,19,51,52]. Hence the changeover from one type to another is associated with different degree of morpho or histodifferentiation or both which is hard to discriminate between both the types [53].

Previous history of trauma is also a factor for developing odontoma. Andreasen (1994) describes an odontoma, due to intrusive luxation or avulsion of the primary tooth. Thus, in primary tooth this type of defective formation is very rare. According to this theory, ameloblasts development gets affected in the morphogenetic stage during early life of tooth formation [54].

Rushton (1957) [46] has described a large protuberance of enamel rising subsequent to trauma of tooth germ before the completion of the enamel cap. In contradiction to the study by Hitchin, Levy [55] showed that the pathological process is initiated only after amelogenesis has occurred and whether trauma would produce hypoplasia, supernumerary or odontoma is depended on stage of development of the cells that are traumatized.

The supposition for cause of hard tissue odontoma are inherited or mutant or interference which will have a genetic control for tooth formation postnatally. Odontoma can occur in one or more of three ways:

1. When genes manage the tooth formation, any interference in the mechanism can lead to odontoma.
2. When any mutation occurs in genes can lead to odontoma.
3. Any inheritance of abnormal genes can also lead to odontoma.

Any mutation in the epithelial cells of tooth germ will change the inherent capacity of odontogenic epithelium to pass the cap and bell stages of tooth development. This in turn will stimulate the mesenchymal cells to differentiate to form functional ameloblasts and odontoblasts which ultimately leads to odontoma [47].

The segregation of normal and tumor odontogenic cells is done by idiom of certain molecules which was noted by Papagerakis, *et al.* [56] The mesenchymal cells containing genes were also noted in odontogenic epithelium. This information in turn is related to greater appearance of tumor-specific genes that are transcribed at an undetectable level or at epithelial mesenchymal transition during normal root formation. Thus, during normal odontogenesis, odontogenic tumor epithelial cells are recapitulating the genetic programs, but tumors cells verified abnormal appearance of these genes.

The Continuum concept given by Cahn and Slums assumed that Ameloblastic Fibroma (AF) will mature as time passes and finally forms odontoma. However, this concept is not accepted as recurrent cases of AF have not noted the steps of maturation into hard tissues which forms tumor of more advanced stage [57].

Radiologic features of odontoma usually relay on stage of formation and degree of organic and inorganic content formation. The first stage is noted as lack of calcification with radiolucent area. The second and third stage shows partial calcification and a radiopaque mass surrounded by radiolucent areas in relation to connective tissue histologically [35,42]. Complex odontoma appear radiographically as opaque mass situated in bone which has density greater than bone and equal or greater than tooth while compound odontoma shows two different radiologic features like, cluster of small shapeless dense mass which does not resemble to shape of tooth and presence of two or more tooth like masses having conical enamel like capped crowns with fusion of radiographic areas [7].

The differential diagnosis includes hypercementosis, Condensing osteitis, periapical osteosclerosis for odontoma.

Odontoma should be removed as it grows very little which contains various tooth structures that can lead to cystic changes [29]. The removal of odontoma is simple as it is a capsulated tumor, but chances of relapse can occur if total removal is avoided. Thus, for all pediatric patients who give a history of delayed eruption, missing tooth, tooth displacement, with or without trauma history, a proper and clear visual, manual and radiographic examination should be done. If odontoma is diagnosed at early stage it helps to adopt less complex and expensive treatment, gives good prognosis and also avoids the chance of recurrence and devitalization of neighboring tooth.

Conclusion

Since odontomas do not show any symptoms, they are detected accidentally or on routine radiographic examinations. A case of painful compound composite odontoma with unerupted premolar has been reported in this article. But to give a definite diagnosis, compound and complex odontomas must be examined properly under the microscope. To avoid any craniofacial complications in later stage of life, early diagnosis and throughout surgical intervention is needed to odontoma.

Acknowledgement

We would like to thank the Department of Pedodontics and Head of Department of Oral and Maxillofacial Pathology for helping us to complete the case report.

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Volume 2 Issue 5 May 2018

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