



## **A Hamartoma or a Neoplasm? A Case Report of an Odontoma Associated with a Dentigerous Cyst.**

Dr. Zahida Parveen<sup>\*1</sup>, Dr. Anand Krishnan<sup>2</sup>, Dr. Vinay Marla<sup>3</sup>, Dr. Abin Varghese<sup>4</sup>, Dr. Kamraj Loganathan<sup>5</sup>, Dr. Ajay Telang<sup>6</sup>

1. Assistant Professor, Faculty of Dentistry, Manipal University College Malaysia.
2. Senior Lecturer, Faculty of Dentistry, Lincoln University College, Petaling Jaya, Malaysia.
3. Associate Professor, Faculty of Dentistry, Lincoln University College, Petaling Jaya, Malaysia.
4. Assistant Professor, Penang International Dental College, Penang, Malaysia.
5. Professor, Penang International Dental College, Penang, Malaysia.
6. Associate Professor, Penang International Dental College, Penang, Malaysia.

**Corresponding Author: Dr. Zahida Parveen**, Assistant Professor, Faculty of Dentistry, Manipal University College Malaysia.

**Copy Right:** © 2022 Dr. Zahida Parveen, This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

**Received Date: May 16, 2022**

**Published Date: June 01, 2022**

**Abstract:**

*Abstract: Odontomas are benign Odontogenic tumors composed of dental tissues which are thought to be the most common Odontogenic tumors. Odontomas are morphologically classified into the 'Compound-type' and the 'Complex-type' based on their resemblance to a normal tooth. The case report describes the occurrence of an odontoma in association with a dentigerous cyst in a paediatric patient and highlights the prompt management of these lesions to avoid any complications in the future.*

**Keywords:** *Odontoma, dentigerous cyst, Hamartoma, impacted tooth, Retained deciduous teeth*

**Introduction**

Odontomas are benign Odontogenic tumors composed of dental tissues which are thought to be the most common Odontogenic tumors. The term "odontoma" was first coined by Paul Brocain in 1867 who suggested that these tumors are formed as a result of disproportionate transitory or full growth of dental tissues.[1] There are two schools of thought with regard to Odontoma. One considers it to be a hamartoma whereas the other suggests it to be an Odontogenic tumor. According to the latest WHO classification, Odontoma is considered to be a mixed epithelial and ectomesenchymal tumor composed of dental hard and soft tissues. [2] However, the ambiguity of an odontoma to be a hamartoma or a neoplasm still exists.

Odontomas are morphologically classified into the 'Compound-type' and the 'Complex-type' based on their resemblance to a normal tooth.[3] Compound odontomas show distinguishable tooth forming structures, namely enamel, dentin, and cementum. Morphologically, these types of odontomas may resemble a tooth but without normal anatomical features and dimensions. Complex odontomas on the other hand show indistinguishable tooth-like structures arranged haphazardly and morphologically present as unrecognisable mineralized structures.[4] Both these forms of odontomas present as radio-opaque structures and show a propensity to occur in certain areas. Compound odontomas are common in the maxillary anterior region whereas complex odontomas are commonly found in the mandibular posterior region. [3,4]

Odontomas are characterised by slow and painless growth and may be associated with the retention of primary tooth and/or delayed eruption of a permanent tooth.[5] Various theories have been suggested

Citation: Dr. Zahida Parveen "A Hamartoma or a Neoplasm? A Case Report of an Odontoma Associated with a Dentigerous

Cyst." MAR Dental Scinces.5.5

[www.medicalandresearch.com](http://www.medicalandresearch.com) (pg. 2)

for the development of an odontoma. These lesions occur as a result of continued budding or abnormal proliferation of the cells of the tooth germ. Some causes considered are trauma to tooth germ in the primary dentition period, hereditary anomalies like Gardner's syndrome, hyperactivity of odontoblasts, or alteration in the genes related to tooth development.[6] In addition to morphological variations in the presentation of an odontoma, these can be associated with the simultaneous presence of other pathologies. There are various incidences of occurrences of odontoma with an associated Odontogenic cyst or a tumor.[7] This case report describes the occurrence of an odontoma in association with a dentigerous cyst in a paediatric patient and highlights the prompt management of these lesions to avoid any complications in the future.

### Case Presentation

An eleven-year-old healthy girl reported to the dental clinic for a missing tooth in the mandibular anterior region. Intra-oral examination revealed mixed dentition with no decayed teeth. It was found that her primary mandibular left central incisor (71) was present, whereas her permanent mandibular left central incisor (31) was missing. Intraoral periapical (IOPA) and orthopantomographic (OPG) views were advised. Intra-oral radiograph revealed an irregular radio-opaque mass with multiple tooth-like structures within it, apical to 71. The root apex of 71 appeared to be resorbed. The location of impacted 31 was identified as inferior to the radio-opaque lesion. The crown of the impacted 31 was circumscribed by an irregular radiolucency suggestive of a cyst (FIGURE 1). Based on these findings a provisional diagnosis of compound odontoma along with dentigerous cyst in impacted 31 was made and an excision biopsy was advised.

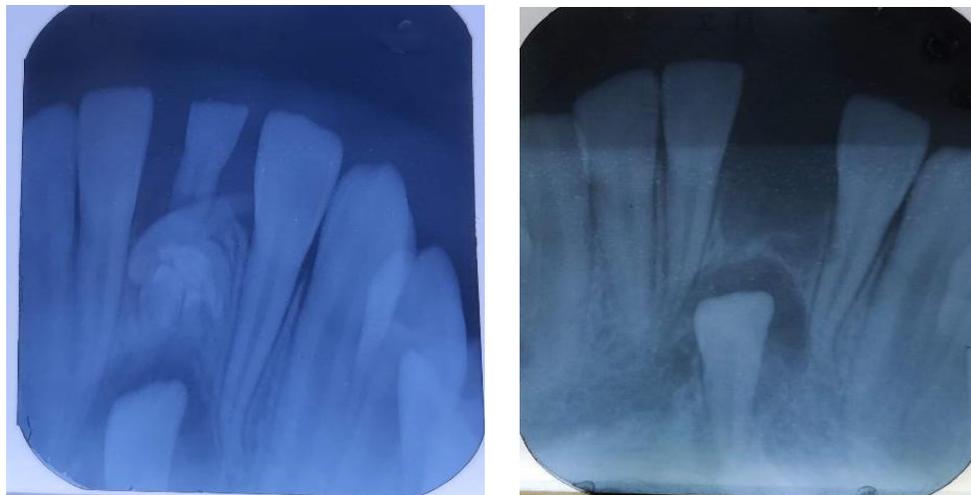


Figure 1

As the child was cooperative, surgical enucleation of the odontoma was planned under local anaesthesia. A mucoperiosteal flap was raised in regions 31, 32, and 41 to expose the bone. A window was made in the bone close to the apex of 71 using a straight slow-speed handpiece bearing a round tungsten carbide bur under normal saline irrigation. Denticles were exposed, and a total of six denticles of various sizes and shapes were removed along with the lining tissue which was sent for histopathological examination to confirm the diagnosis and rule out any malignant changes (FIGURE 2). Microscopic examination of the soft tissue showed cystic lumen lined by non-keratinized stratified squamous epithelium composed of cuboidal cells, and 2 to 6 layers thick (FIGURE 3).

The lining epithelium also shows ciliated columnar epithelial cells in areas. The underlying connective tissue stroma was fibrous and devoid of inflammatory cells. The denticles were not processed since these had the morphological characteristics resembling a tooth. After the extraction of 71, the position of 31 was determined. After thorough irrigation of the enucleated site, the flap was repositioned and sutured with 3-0 Vicryl.

The patient was followed up regularly for the next 3 months and showed uneventful healing. But since the impacted tooth (31) did not erupt in the next 3 months, a CBCT was taken (FIGURE 4). A well-defined radiolucency measuring approximately 1x1cm in size was observed in relation to the crown of 31, which was surgically removed. Based on these findings a final diagnosis of Compound Odontoma along with Dentigerous Cyst in 31 was given.



**Figure 2**

**Figure 3**

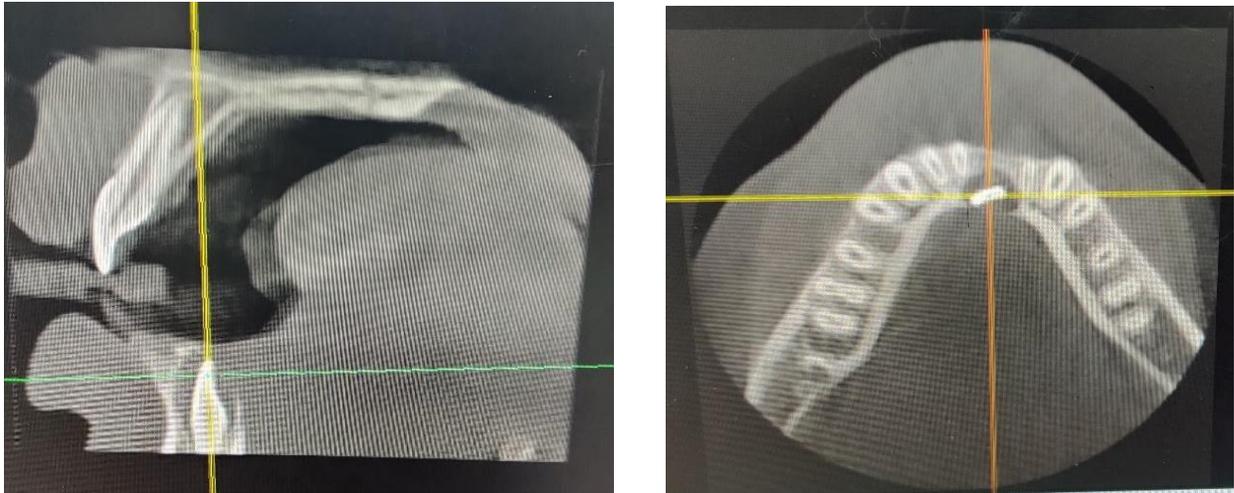
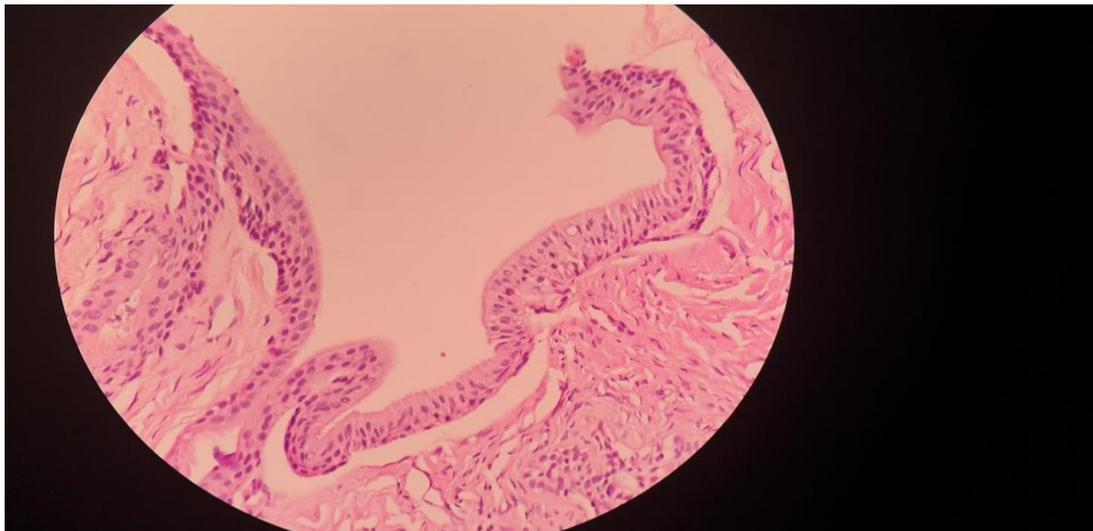


Figure 4



## Discussion

Odontoma is the most common odontogenic lesion of the oral cavity. These are generally considered as tumor-like malformations or hamartomas due to their non-aggressive nature and slow growth potential but are designated as a “mixed’ (epithelial & ectomesenchymal) Odontogenic tumor according to the WHO classification 2017.[1,2] The incidence of these lesions is said to be in the range of 20-67% as reported by different studies on Odontogenic neoplasms.[8] Two types of odontomas have been described, the compound odontoma and the complex odontoma and variations have been found between these two suggesting that these two types may be pathogenetically different.[8,9] The variations are noted generally in terms of its appearance but also in terms of age, gender, and location of occurrence. Compound odontomas are frequently encountered in younger males in the maxillary anterior region

Citation: Dr. Zahida Parveen “A Hamartoma or a Neoplasm? A Case Report of an Odontoma Associated with a Dentigerous Cyst.” MAR Dental Sciences.5.5

[www.medicalandresearch.com](http://www.medicalandresearch.com) (pg. 5)

whereas the complex forms occur commonly in older females in the mandibular posterior teeth.[2,9] A complex odontoma is thought to be a terminal stage lesion and hence considered as a hamartoma.[10] Both these forms of odontomas have a similar rate of incidence as described by Oliveira et al who found that the incidence of compound odontoma ranged between 9% to 37% and those of couples odontomas ranged from 5% to 30%.[11]

Odontomas are identified incidentally during routine dental evaluation and rarely forms the chief complaint of the patient. Overall, most of the cases of odontomas are diagnosed during the first and second decades of life and only less than 10% after the age of 40.[12] In our case, the patient reported with the chief complaint of missing permanent tooth and was asymptomatic. The time of diagnosis of odontoma in our patient who is eleven years old corresponds with the existing literature.

The presence of odontoma was ascertained during an attempt to determine the presence of the missing permanent tooth within the mandible. Compound odontomas are commonly thought to be seen in the anterior maxilla.[2] However, in our case, it was found in the anterior mandible which is an uncommon location. In the absence of symptoms, other presentations should be considered to evaluate the presence of an odontoma. A tooth may be missing congenitally or it may be impacted due to some cause. The most common causes of impaction are due to lack of adequate jaw space, an improper path of eruption, or the presence of any pathology preventing the eruption of that tooth. Odontomas may cause such obstruction to eruption or may actually develop from the tooth germ of the missing tooth altogether.[11]

The odontomas are harmless lesions and as such are considered as a hamartoma and not a true neoplasm.[1] However, these seem to cause some complications. Most commonly reported local complications include disturbances in eruption and malposition of teeth. Resorption of the root of an adjacent tooth due to an odontoma has also been reported. It is recommended that an odontoma should be surgically removed to allow the eruption of the permanent tooth. Care should be taken not to injure the adjacent structures. The impacted tooth should not be extracted and followed up so as to anticipate normal tooth eruption following the removal of the odontoma. [5] Various articles have said removal of the odontoma led to spontaneous eruption of the impacted permanent tooth. One article has suggested that failure to identify an odontoma early resulted in impaction of a tooth with complete root formation.[11] Bengston et al and Oliver & Hodges suggested that such teeth be brought to proper position within the dental arch using orthodontic traction. [13,14] In our case, the impacted 31 failed to erupt due to the presence of a dentigerous cyst associated with it and hence had to be extracted to remove the pathology.

The occurrence of odontomas does not always occur in isolation but there are instances wherein it is seen along with another pathology mostly Odontogenic in nature. This may have varied implications when considering odontoma as a mere hamartoma or as a precursor to a more pathologic entity. Various reports have described the concurrent occurrence of an odontoma along with dentigerous cysts,

Citation: Dr. Zahida Parveen "A Hamartoma or a Neoplasm? A Case Report of an Odontoma Associated with a Dentigerous Cyst." MAR Dental Scinces.5.5

[www.medicalandresearch.com](http://www.medicalandresearch.com) (pg. 6)

calcifying odontogenic cyst, or even ameloblastoma. [15,16] This highlights the importance of early identification and removal of odontoma to avoid complications and lengthy treatment in the future. It has been suggested that any evidence of delayed eruption of permanent tooth or displacement of the deciduous tooth should be followed up by radiographic imaging in paediatric patients so as to provide a less complex and inexpensive treatment and also to offer a better prognosis.[6] In our case, the presence of an unerupted tooth led to the identification of odontoma which in turn also led to the diagnosis of a dentigerous cyst. All these pathological entities were treated so as to avoid any neoplastic alteration in the future. One year follow-up of the patient has been uneventful without any complications detected. According to a study done by Kaugars et al. in which 351 cases of odontomas were evaluated, it was found that 27.6% were associated with a dentigerous cyst.[17] These findings suggest that the discovery of an odontoma leads to a possibility of another co-existing Odontogenic lesion.

The presence of an odontoma may be the cause of the development of cystic transformation in a nearby unerupted tooth which in turn lead to further complications.[18] Another study conducted by Zhang et al on 2082 cases of dentigerous cysts revealed the co-existence of other pathologies. To conclude, this highlights the careful evaluation of cases presenting with any of the Odontogenic lesions and underlies the importance of early identification for obtaining a good prognosis.

## **Conclusion**

Odontomas are the most common odontogenic pathology of the oral cavity and pose a diagnostic challenge due to their innocuous presentation. However, these have the potential to cause serious complications if left untreated. Early identification and treatment of this pathology can prevent the development of more aggressive pathologic entities in a patient.

## **References**

1. Satish V, Prabhadevi MC, Sharma R. Odontome: "A Brief Overview". *Int J Clin Pediatr Dent.* 2011;4(3):177-185. doi:10.5005/jp-journals-10005-110.
2. Speight PM, Takata T. "New tumour entities in the 4th edition of the World Health Organization Classification of Head and Neck tumours: odontogenic and maxillofacial bone tumours". *Virchows Arch* 2018, 472, 331–339.
3. Budnick SD. "Compound and complex odontomas". *Oral Surg Oral Med Oral Pathol.* 1976 Oct;42(4):501-6. doi: 10.1016/0030-4220(76)90297-8.
4. Gedik R, Müftüoğlu S. "Compound Odontoma: Differential Diagnosis and Review of the Literature". *West Indian Med J.* 2014;63(7):793-795. doi:10.7727/wimj.2013.272

5. Preoteasa CT, Preoteasa E. "Compound odontoma - morphology, clinical findings and treatment. Case report". Rom J Morphol Embryol. 2018;59(3):997-1000. PMID: 30534846
6. Prabhu N, Issrani R, Patil S, Srinivasan A, Alam MK. "Odontoma- An Unfolding Enigma". J Int Oral Health 2019;11:334-9.
7. Kim KS, Lee HG, Hwang JH, Lee SY. "Incidentally detected odontoma within a dentigerous cyst". Arch Craniofac Surg. 2019;20(1):62-65. doi:10.7181/acfs.2018.02313
8. Uma E. "Compound odontoma in anterior mandible—a case report". Malays J Med Sci. 2017;24(3):92–95.
9. Patekar D, Kheur SM, Gupta AA. "Odontoma - A brief overview". Journal of Oral Disease Markers (2018), 2, 23–25
10. Patricia GA, Harumi I, Yuji M, et al. "Podoplanin expression in odontomas: clinicopathological study and immunohistochemical analysis of 86 cases". J Oral Sci. 2011;53(1):67–75.
11. de Oliveira BH, Campos V, Marçal S. "Compound odontoma--diagnosis and treatment: three case reports". Pediatr Dent. 2001 Mar-Apr;23(2):151-7.
12. Janev E, Redzep E, Kanurkova L, Tosevska S. "COMPOUND ODONTOMA IN CHILDHOOD POPULATION (CASE REPORT)". Journal Of Morphological Sciences 2020, 3(1), 73-78.
13. Bengston AL, Bengston NG, Benassi, LRDC: "Odontomas em pacientes pediátricos". Revista de Odontopediatria 2:25- 33, 1993.
14. Oliver RG, Hodges CGL: "Delayed eruption of a maxillary central incisor associated with an odontome: report of case". ASDC J Dent Child 55:368-71, 1988.
15. Melo RB, de Souza Damasceno YE, da Cunha Junior CAC, Pontes IV. "Compound odontoma associated with dentigerous cyst in the anterior mandible – case report". RSBO. 2015, 12(1):98-102.
16. Jayam C, Bandlapalli A, Patel N, Choudhary RS. "A case of impacted central incisor due to dentigerous cyst associated with impacted compound odontoma". BMJ Case Rep. 2014;2014:bcr2013202447. Published 2014 Mar 20. doi:10.1136/bcr-2013-202447.
17. Kaugars GE, Miller ME, Abbey LM. "Odontomas. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology". 1989(67):172-6.
18. Kalburge JV, Latti B, Kalburge V, Kulkarni M. "Neoplasms associated with dentigerous cyst: An insight into pathogenesis and clinicopathologic features". Arch Med Health Sci 2015;3:309-13
19. Zhang LL, Yang R, Zhang L, Li W, MacDonald- Jankowski D, Poh CF. "Dentigerous cyst: A retrospective clinicopathological analysis of 2082 dentigerous cysts in British Columbia, Canada". Int J Oral Maxillofac Surg 2010;39:878-82.

**Acknowledgement:**

Case was reported in Penang International Dental College, Penang, Malaysia.