



ISSN: 0975-833X

Available online at <http://www.ijournalcra.com>

International Journal of Current Research
Vol. 15, Issue, 01, pp.23485-23489, January, 2023
DOI: <https://doi.org/10.24941/ijcr.44725.01.2023>

INTERNATIONAL JOURNAL
OF CURRENT RESEARCH

RESEARCH ARTICLE

COMPLEX ODONTOMA AN INCIDENTAL FINDING: A CASE REPORT

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ARTICLE INFO

Article History:

Received 14th October, 2022

Received in revised form

17th November, 2022

Accepted 19th December, 2022

Published online 30th January, 2023

Key words:

Complex Odontoma, Intraoral
Enucleation, Odontogenic Tumour,
Odontoma.

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Citation: Dr. Sayantan Nag, Dr. Krishna Kishor, Dr. Anu Singh, Dr. Abhishek Kumar Singh and Dr. Shakti Kumar Sonu. 2023. "Complex odontoma an incidental finding: a case report". *International Journal of Current Research*, 15, (01), 23485-23489.

ABSTRACT

Odontomas are the most common odontogenic tumors. They are broadly classified into Compound Odontoma and Complex odontoma. Complex odontoma is a conglomerate of all the dental tissues that is characterised by normal histodifferentiation but abnormal morphodifferentiation producing little or no resemblance to normal tooth form. They are usually asymptomatic so incidental findings are common. We report an interesting case of complex odontoma that was associated with a small bony swelling of the mandible. The tumour was treated by surgical enucleation under local anaesthesia.

INTRODUCTION

The term 'odontoma' was first used in 1866 by Broca who defined it as a tumor formed by the overgrowth of complete dental tissue. Odontomas, which are considered developmental anomalies (hamartomas) rather than true neoplasms are the most common benign odontogenic tumors and composed of enamel, dentin, cementum, and pulp. Various theories have been suggested for etiology of odontomas: local trauma in primary dentition duration, infectious and/or inflammatory processes, hereditary abnormalities, and any alteration in genetic structures that can affect the tooth development². Hitchin, suggested that odontomas are inherited through a mutant gene or interference, possibly postnatal, with genetic control of tooth development. In humans, there is a tendency for the lamina between the tooth germs to disintegrate into clumps of cells. The persistence of a portion of lamina may be an important factor in the etiology of complex or compound odontomas and either of these may occur instead of a tooth. In either case, a mutation in the epithelial cells of the persistent lamina or of the tooth germ itself may change the inherent capacity of the odontogenic epithelium to go through the cap and bell stages necessary for tooth formation and still retain its ability to stimulate mesenchymal differentiation necessary for dentin formation and to form functional ameloblasts and odontoblasts leading to a composite³.

On the basis of gross, radiographic, and microscopic features, two types of odontoma are recognized: the compound and the complex. The compound odontome has all the dental tissues represented in a more orderly fashion, so that the lesion consists of many small tooth-like structures each having enamel, dentin, cementum and pulp arranged as in a normal tooth. The odontoma, complex type (ODCx) is a hamartomatous lesion in which all the dental tissues are represented, individual hard tissues being mainly well formed but occurring in a more or less disorderly pattern⁴. The ODCxs are slow growing, expanding lesions that are usually detected in the second decade of life. The lesions are invariably asymptomatic and are usually discovered on routine radiographic examinations. Most of the odontomas are associated with pathologic changes such as malformation, impaction, delayed eruption, malpositioning, cyst formation or displacement and resorption of adjacent teeth, but only rarely are they seen to be associated with the absence of one or more contiguous teeth⁴. WHO defines complex odontoma as malformation in which all of the dental tissues are represented, and individual tissues mainly are well formed but occur in disorderly pattern. Compound odontoma is twice as common when compared to complex odontoma. 60% of complex odontomas occur in women. Complex odontomas occur in mandibular first and second molar region with slight or marked bony expansion. The treatment of choice is surgical excision of the lesion followed by histopathological study to confirm the diagnosis⁵.

CASE REPORT

A 27-year-old female patient presented with a chief complaint of pain in right lower back side of jaw for past 2 months. The pain was intermittent and dull aching in nature.



Fig 1. Frontal View



Fig 2. Intraoral View



Fig 3: OPG Before tooth extraction



Fig 4. OPG 1 month post extraction

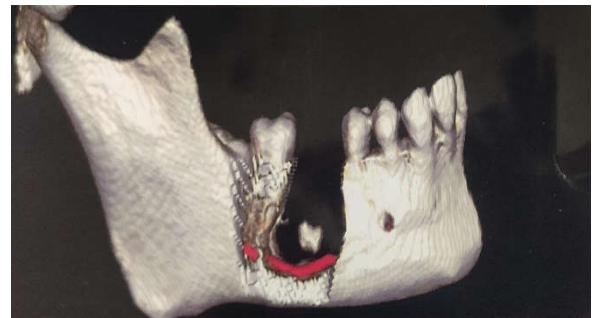


Fig 5. CBCT 3D Reconstruction (Sagittal cut)

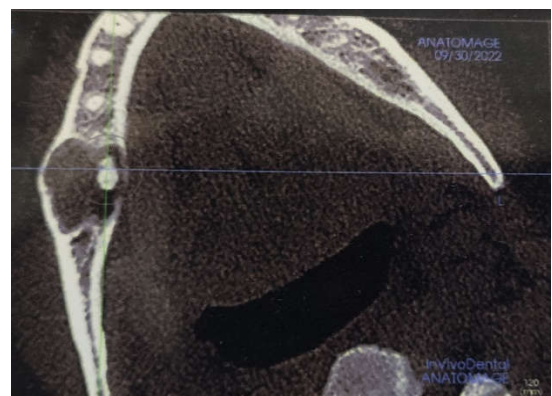


Fig 6. CBCT Axial cut

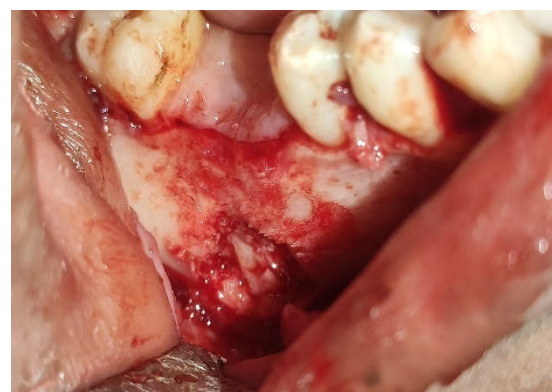


Fig 7: Exposure of the lesion

The patient had a history of tooth extraction in the same area before 1 month elsewhere. The past medical and family history was unremarkable. Intraoral examination revealed missing mandibular right first molar tooth. There was mild swelling of dimension 1x1 centimetre in the vestibule with respect to right lower mandibular first molar.

On palpation the swelling was bony hard in consistency and there was vestibular tenderness in relation to right mandibular first molar tooth. Orthopantomograph (OPG) were previously done by the patient when she got her tooth extracted which revealed a radio-opaque mass surrounded by a thin radiolucent halo. The overlying tooth (mandibular first molar) had root resorption. 1 month post extraction OPG shows healing socket with the same radio-opaque mass in-situ.

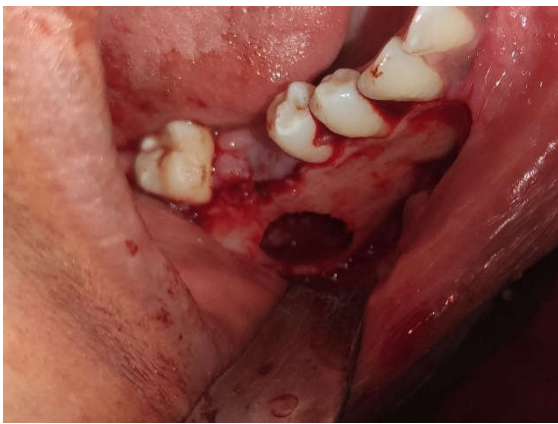


Fig 8. Cavity after enucleation



Fig 12. 3 months post operative intra oral healing



Fig 9. Specimen



Fig 10. Intraoral view after closure



Fig 11. 3 months Post operative OPG

As the buccolingual dimension of the lesion was not assessed by the OPG, a cone beam computed tomography (CBCT) was advised. CBCT revealed a single unilocular radiolucency with respect to the periapical space of 46 region, measuring 15.38mm x 14.24mm x 16.12mm with greatest dimensions. A radio-opaque single tooth like structure seems to be impacted and associated with unilocular radiolucency involving perforation of buccal and lingual cortical bone. Lesion caused inferior displacement of right inferior dental canal towards the lower border of mandible. A provisional diagnosis of complex odontoma was made. Ameloblastic fibro odontoma and ameloblastic odontoma were considered in differential diagnosis. Taking the size of the lesion into consideration, excision of the lesion was planned under local anaesthesia.

An intraoral approach was taken where after taking all septic precautions and administration of 2% lignocaine with 1:80000 adrenalin, a crevicular incision was placed from distal to 43 to distal to 47. A full thickness mucoperiosteal flap was raised. A bony window was created using a bur and osteotome to expose the lesion. The lesion consisted of a small amorphous calcified mass which was then enucleated in small pieces. The removal of the lesion resulted in a small locular cavity with inferior alveolar nerve intact at its lower border. The soft tissue was closed primarily without packing or grafting the cavity with 3-0 silk sutures and the specimen was sent for histopathological examination (HPE). HPE confirmed the diagnosis of complex odontoma. The patient was under observation for 3 months period. Postoperative OPG revealed good bony healing without any recurrence. Intraoral healing was satisfactory without any defects. There was no paraesthesia or anaesthesia postoperatively noted of the areas supplied by inferior alveolar nerve on the right side.

Table 1: Published cases of odontomas since 2010 by J. C. Park et al¹¹

Authors	Size	Position	Age	Sex	Type
Bageswadi et al. ¹	4×3.5 cm	Rt. Mn. Posterior	22	M	Complex
Spini et al. ⁴	6 cm	Mn. Anterior	9	M	Complex
Perumal et al. ³	5.5×4×2.5 cm	Rt. Mn. Posterior	24	F	Complex
Lee et al. ⁷	2.7×1.0×1.8 cm	Rt. Mn. Anterior	41	F	Compound
	2.3×2.0×1.9 cm	Rt. Mn. Posterior	16	F	Compound
Chrcanovic et al. ⁸	4×3×2.75 cm	Rt. Mn. Posterior	21	F	Complex
Utumi et al. ²	4 cm	Rt. Mn. Anterior	10	F	Complex
Akerzoufi et al. ¹²	6×6 cm	Lt. Mn. Posterior	35	F	Compound
Lehman et al. ¹⁷	8×4 cm	Rt. Mn. Posterior	7	F	Compound
Bisicic et al. ¹⁹	5×3 cm	Rt. Mn. Posterior	10	F	Complex
Reddy et al. ²²	2.9×1.6 cm	Rt. Mn. Posterior	13	M	Complex

Rt.: right, Lt.: left, Mn.: maxilla, Mn.: mandible

DISCUSSION

Odontomas account for a large percentage of all odontogenic tumors. A number of studies have examined large series of these tumors, and odontomas have been identified as the most frequent lesions. Buchner et al. in 2006 examined a sample of 1088 odontogenic tumors, in which odontomas accounted for 75.9% of the total. Previously, in 2002, Ochsnius et al. analyzed a sample of 362 odontogenic tumors in which odontomas represented 44.7%. In the year 1997, Mosqueda et al. evaluated a series of 349 odontogenic tumors, of which 34.6%

corresponded to odontomas. Odontomas have been the subject of many studies, some involving a large number of cases, such as the group analyzed by Philipsen et al. in 1997 (with 134 cases), or the study published by Hisatomi et al. in 2002 (with 107 cases). Since odontomas represent a large proportion of these lesions, adequate knowledge of their characteristics is necessary in order to establish correct diagnosis and treatment¹³. The term odontoma was coined by Paul Broca in 1867. He defined the term as tumors formed by the overgrowth of transitory of complete dental tissues¹⁴. The odontoma is thought to arise from remnants of the dental lamina, the cell rests of Serres, they are the most commonly responsible for odontogenic tumours. The lesions are slow growing and non-aggressive in behavior and are regarded by some authors as hamartomatous malformations which lack the diagnostic features of persistent and uncoordinated growth that characterize tumours and hence have a limited growth potential¹⁶. H.M. Worth in 1937 classified odontomas as: a) epithelial odontomas arising from dental epithelium, e.g., dentigerous cyst, adamantinoma. b) composite odontomas arising from the dental epithelium and dental mesoblastic tissues, e.g., complex, compound, geminated and dilated. Thoma and Goldman in 1946, gave a classification as: a) Geminated composite odontomas: nearly well-developed fused teeth. b) Compound composite odontomas: made up rudimentary teeth. c) Complex composite odontomas: calcified structures not resembling normal anatomical arrangement of dental tissues. d) Dilated odontomas: enlarged crown or root portion of tooth. e) Cystic odontomas: odontoma encapsulated by fibrous connective tissue in a cyst or in the wall of a cyst. Odontomas are also classified as intraosseous and extraosseous odontomas. The intraosseous odontomas occur inside the bone and may erupt into the oral cavity (erupted odontome). The extraosseous or peripheral odontomas are odontomas occurring in the soft tissue covering the tooth-bearing portions of the jaws and having a tendency to exfoliate. World Health Organization (WHO) classified odontomas into three groups: a) Complex odontoma; when the calcified dental tissues are simply arranged in an irregular mass bearing no morphological similarity to rudimentary teeth. b) Compound odontoma: composed of all odontogenic tissues in an orderly pattern, which result in many teeth-like structures, but without morphological resemblance to normal teeth. c) Ameloblastic fibro-odontome: consists of varying amounts of calcified dental tissue and dental papilla-like tissue, the later component resembling an ameloblastic fibroma. The ameloblastic fibro-odontome is considered as an immature precursor of complex odontoma. A new variant called as hybrid odontomas are quoted in few published literatures⁷.

The origin of complex odontoma is unknown; some suggest trauma or infection to be the cause. In a study conducted by Lopez-Areal et al., they found that a child developed multiple odontomas after experiencing trauma with intrusion of incisor teeth at the age of 10 months. Hitchin has said that odontomas are inherited or developed as a result of genetic mutation. An increased number of odontomas were found in people with Gardner's syndrome which is a heritable syndrome. Recurrence of complex odontomas is very rare⁵. R.M.Kodali et al cited that although it is agreed that ODCx tend to occur in the posterior regions of the mandible, differences exist in the literature regarding their prevalence. With a slight preponderance to females, the relative frequency of occurrence varies between 5 and 30%. They may be discovered at any age, while the majority of cases (84%) occur before the age of 30. With a peak in the second decade of life, less than 10% are only found in the patients over 40 years of age⁴. Although they are commonly asymptomatic, clinical indicators of odontoma may include retention of deciduous teeth, noneruption of permanent teeth, pain, expansion of the cortical bone and tooth displacement. Other symptoms include anesthesia in the lower lip and swelling in the affected area³. Radiologically odontoma presents as a well-defined radiopacity situated in bone, but with a density that is greater than bone and equal to or greater than that of a tooth. It contains foci of variable density. A radiolucent halo, typically surrounded by a thin sclerotic line, surrounds the radiopacity. - e radiolucent zone is the connective tissue capsule of a normal tooth follicle. - e thin sclerotic line resembles the corticated border seen in a normal tooth crypt. - e developmental stages can be identified based

on radiologic features and the degree of calcification of the lesion at the time of diagnosis. The first stage is characterized by radiolucency due to the absence of dental tissue calcification, the second or intermediate stage shows partial calcification and the third or classically radiopaque stage exhibits predominant tissue calcification with the surrounding radiolucent halo described above³. Based on the radiolucent rim around the radiopaque mass on the radiograph, the differential diagnoses considered included osteoma and cemento-osseous dysplasia (COD). Other mixed odontogenic tumors such as ameloblastic fibro-odontoma or ontoameloblastoma, which grow aggressively, were also considered. Radiographically, osteomas are well-demarcated, ovoid, radiopaque lesions¹⁸ that can be distinguished from complex odontomas by the lack of a radiolucent rim around the lesion. It is easy for CODs in the end stage to be confused with complex odontomas because they appear as dense radiopaque masses with radiolucent rims on radiographs.

However, CODs occur in persons over 30 years of age and are related to the root, whereas complex odontomas occur mainly in adolescents and are located in the alveolar bone of the jaw. Ameloblastic fibro-odontomas present as well-demarcated radiolucencies containing radiopaque material and usually occur in patients younger than 20 years of age. Ontoameloblastomas, in contrast, are characterized by multilocular radiolucent lesions with radiopaque areas resembling dental tissue, and also predominantly affect patients younger than 20 years of age.²¹ The main difference between complex odontomas and mixed odontogenic tumors is their growth potential, as exemplified by the fact that even an ontoameloblastoma can cause root resorption of the adjacent teeth¹¹.

The treatment of choice according to most consulted authors is surgical extraction, since the odontoma may interfere with eruption of the permanent tooth, displace the adjacent teeth, or give rise to a dentigerous cyst. Such treatment is carried out under local or general anesthesia. However, local anesthesia with an intrabuccal approach is clearly the predominant option. The resected odontoma should be submitted for histological study, to confirm the diagnosis¹³. Similarly in our case as the associated tooth was already extracted 1 month back elsewhere, we performed an intraoral approach for enucleation of the lesion and subsequently subjecting the specimen for histopathological examination which further revealed the lesion to be a complex odontoma. As the bony defect after enucleation was more or less small in dimension, we did opt for autologous grafting taking the donor site morbidity into consideration. The 6 months post operative healing and bone formation was satisfactory considering the age and good growth potential of the patient.

Table 1: Published cases of odontomas since 2010 by J. C. Park et al¹¹

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Akerzoul et al. ¹²	6×6 cm	Lt. Mn. Posterior	35	F	Compound
Lehman et al. ¹⁷	8×4 cm	Rt. Mn. Posterior	7	F	Compound
Biocic et al. ¹⁹	5×3 cm	Rt. Mn. Posterior	10	F	Complex
Reddy et al. ²²	2.9×1.6 cm	Rt. Mn. Posterior	13	M	Complex

Rt.: right, Lt.: left, Mn.: maxilla, Mn.: mandible

CONCLUSION

This report has helped us to better understand the clinical and radiological features of complex odontomas. Moreover, the cases of complex odontomas can be differentiated from other conditions mostly by age, position of the lesion, its sex predilection and radiological features which includes differences in opacity of the radio-opaque mass and features of the well-defined radiolucent rim. Its appropriate diagnosis and conservative surgery is the most accurate treatment.

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