






# Simultaneous occurrence of dentigerous cyst and compound-complex odontoma causing dental impaction

Marco Tullio Brazao-Silva<sup>1</sup> , Douglas Magno Guimarães<sup>2</sup> , Anamaria de Lima Laranjeira<sup>1</sup> , Valdir Cabral Andrade<sup>3</sup> , Danillo Costa Rodrigues<sup>1</sup> 

## ABSTRACT

The present work illustrates and discusses this disease that represents the most common odontogenic tumor in clinical practice, the Odontoma. However, an unusual variation of the same is included here in the literature, characterized by the exams as a form of compound-complex odontoma additionally causing dentigerous cyst formation and dental impaction. The unusual case occurred in a 21-year-old woman, complaining of missing the tooth 23. The details of the exams allowed the visualization of compound and complex areas in the lesion, cystic formation and dental impaction. The histopathological findings confirmed the diagnosis of the Odontoma, and the interpretation with the set of other exams led to the final diagnosis of Compound-complex odontoma with dentigerous cyst. The present case highlights the need for early removal of Odontoma due to the possibility of developing cysts, compromising adjacent structures, in addition to its growth potential already reported in the literature.

**Keywords:** Compound-complex odontoma, Dentigerous cyst, Differential diagnosis, Odontogenic tumors.

## INTRODUCTION

Odontomas can be classified as the most common benign odontogenic tumors, accounting for between 39-51% of all these tumors<sup>1</sup>. The disease involves deregulation in cells of odontogenic potential of both ectodermal and ectomesenchymal origin, thus creating dental tissue in their composition, such as enamel, dentin and cementum. In this sense, when dental structures are identified within the tumor mass, it is called a compound odontoma, while in situations where the tissues are randomly organized without the formation of denticles, even if they are anomalous, the reserved term is complex odontoma. Compound odontoma is the most common subtype, radiographically shown as a well-delimited radiolucent area filled by radiopaque, tooth-like bodies of varying size and shape<sup>2</sup>. The complex variant appears as an irregular mass with radiodensities varying according to the region of dental structures present in the mass<sup>3</sup>. A radiolucent margin around the odontomas represents tissue similar to the dental follicle.

Odontomas are often asymptomatic, so they are almost always discovered in routine radiographic examinations, often associated with dental impaction, mispositioning and anodontia<sup>3,4</sup>. Total enucleation of the lesion must be performed to avoid complications such as the appearance of cysts associated with the odontoma, in addition to being the only way to guarantee a conclusive diagnosis, and it may also allow the eruption of a nearby impacted tooth, if any<sup>4</sup>. However there are situations in which the removal of the lesion can generate bone structural fragility, or in cases of larger lesions, so that more advanced surgical approaches and complex planning may be necessary<sup>5</sup>.

The present work aims to present a typical case of odontoma, illustrating a conservative approach, and opportunely reviewing the practice of diagnosis and therapeutic management for this lesion, which is not uncommon in clinical dental practice. The case was analyzed and approved by the Research Ethics Committee of the State University of Montes Claros (Unimontes), under register number 4,416,065.

<sup>1</sup> Universidade Estadual de Montes Claros. Departamento de Odontologia, Montes Claros, (MG), Brasil

<sup>2</sup> Centro universitário do Pará, Belém, (PA), Brasil

<sup>3</sup> Universidade Federal de Juiz de Fora. Departamento de Odontologia, Governador Valadares, (MG), Brasil



## CASE REPORT

A 21-year-old male patient, brown, complained of aesthetic dissatisfaction due to the absence of the tooth 23. On physical examination, the absence of the tooth 23 and a small increase in the volume of hard consistency in the region of teeth 22 and 24 were observed. A radiopaque mass surrounded by a radiolucent halo, with small structures of density similar to dental material extending from the mesial of 22 to the distal of 24, in addition to the presence of the included tooth 23 close to the lesion (Figure 1). For a better assessment of the lesion extension and surgical planning, a cone beam computed tomography with 1mm slices and 3D reconstruction was requested (Figure 2), which was possible to observe an increase in the pericoronal follicle associated with tooth 23, suggestive of a dentigerous cyst (Figure 2). 3). Based on clinical and imaging data, the diagnostic hypothesis was composite odontoma in the left anterior region of the maxilla and dentigerous cyst associated with tooth 23. The treatment plan was surgical removal of the lesions and tooth 23 under local anesthesia.

Through the Neumann flap, the bone under the lesion was removed to facilitate access, then the tumor was completely separated from the bone and removed together with tooth 23 and the associated cyst, maintaining bone continuity (Figure 4). During removal, denticles were found that characterize the composite odontoma and a larger amorphous mass, characteristic of complex odontoma, which after an anatomopathological evaluation showed the presence of all dental tissues important to conclude the diagnosis of odontoma, including immature enamel matrix.

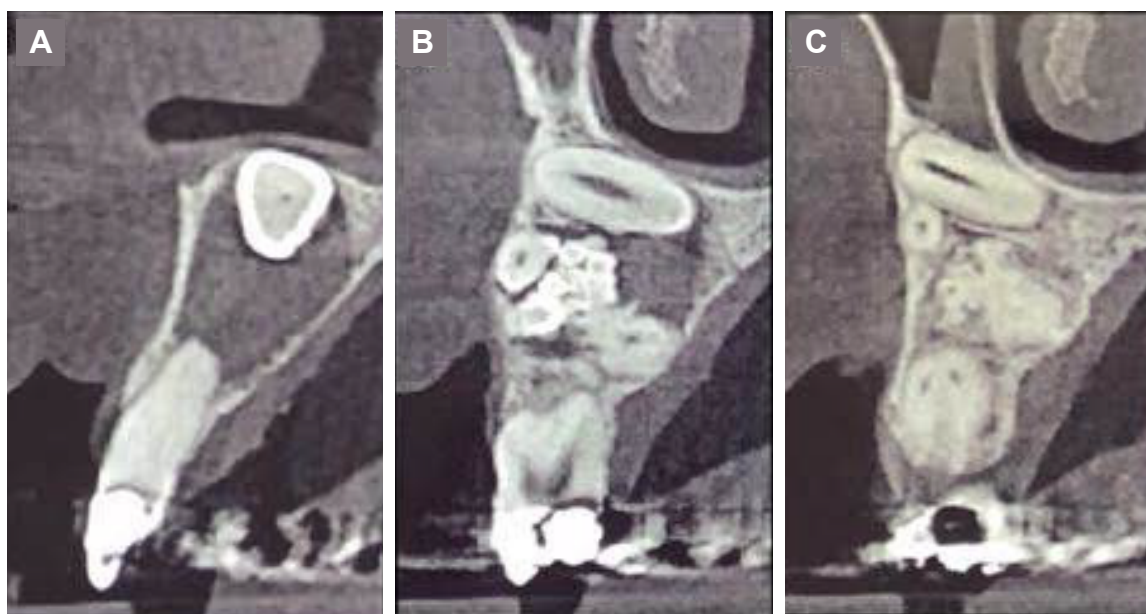
The correlation between imaging findings showing denticle-shaped structures and adjacent amorphous structure, ample hypodense space involving mineralized tissues (Figure 3A) confirmed by the trans surgical cystic appearance (Figure 4B), and complementary histopathological examination confirming that it was only an odontoma with the usual capsular tissues corresponding to pricorony hood, led to the conclusion that the case is a compound-complex odontoma with an associated dentigerous cyst. The patient was followed up at the Stomatology clinic with satisfactory evolution and no recurrences after 1 year.



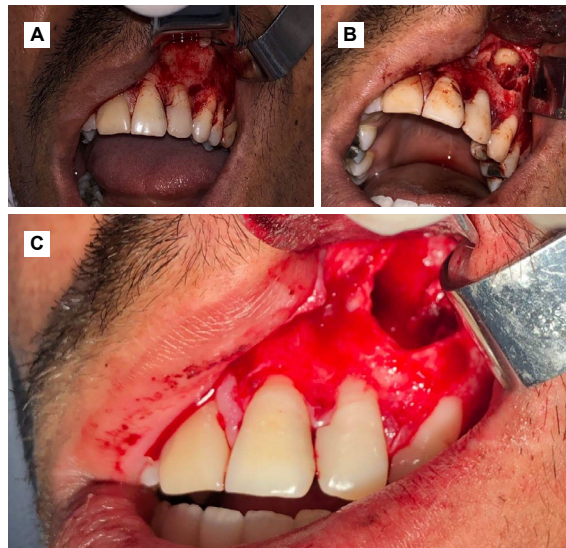
**Figure 1:** Panoramic radiography, showing mixed lesion with structures similar to denticles and radiopaque microfragments, surrounded by a radiolucent line, causing impaction of tooth 23 and deviation of the erupted roots of 22 and 24.



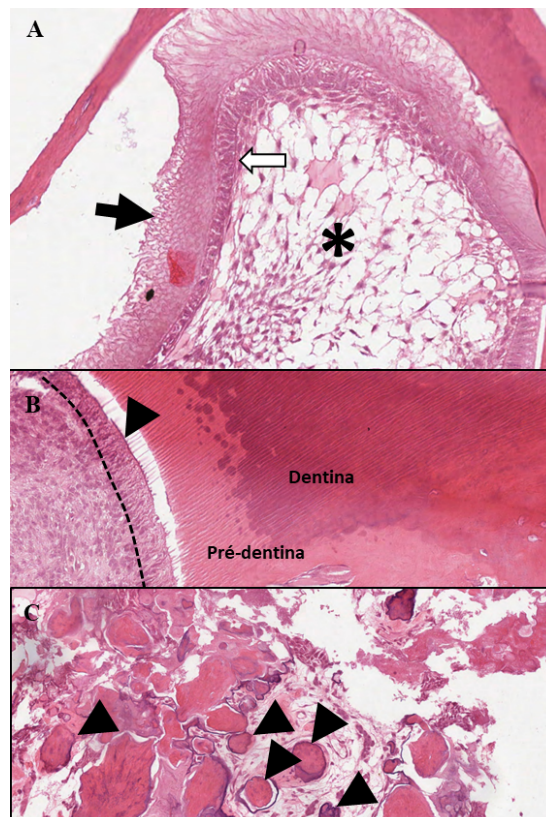
**Figure 2:** Three-dimensional reconstruction of the lesion by computed tomography, showing a solid mass with a density similar to that of dental enamel, with an irregular surface, in proximity to the cystic area and impacted tooth.



**Figure 3:** Sagittal sections by computed tomography. A) shows a wide hypodense area and coronal part of the impacted tooth 23. B) The presence of root part of 23 and hyperdense content inside the hypodense region, some in the form of microteeth in cross section, with a hypodense central area mimicking dental pulp, next to an amorphous mass with regions of density similar to enamel. C) a hyperdense mass with a density similar to the dentinal tissue, amorphous and surrounded by a thin hypodense line, the set involving almost the entire thickness of the maxillary bone ridge.



**Figure 4:** A. Trans surgical view showing bone swelling. B. Removal of the denticles and exposure of the crown of the impacted canine. C. Appearance of the bone cavity after surgical extraction of the odontoma and impacted canine.



**Figure 5:** Histological sections of the odontoma stained with Hematoxylin-Eosin. A. Acellular matrix resembling fish scales indicating tooth enamel (black arrow) and a row of columnar cells bordering this matrix similar to ameloblasts (white arrow), followed by flattened cells that abruptly move away maintaining intercellular bridges, loosely arranged, indicating the stellate reticulum of an enamel organ as in odontogenesis. B. Odontoblast cells (arrowhead and outlined by black dots) bordering an artefactual area of a histological cleft that meets a tubular eosinophilic matrix indicating immature dentin. C. area showing basophilic mineralized material that forms spherical structures called cementicles (arrowhead), characterizing a formation of disorganized cementoid tissue.



## DISCUSSION

The etiopathogenesis of odontoma, like so many other oral diseases, is unknown, with debate on theories involving traumatic possibility, heredity (Gardner syndrome, Hermmman syndrome, basal nevoid syndrome) and mutations<sup>2,6</sup>. It is known that in this condition there is great activation of genes, such as those of the Wnt-1 and  $\beta$ -catenin pathways, which are related to the initiation and development of tooth germs<sup>6</sup>. However, odontoma is currently conceptualized as a hamartoma rather than a true neoplasm, although it remains classified within the group of benign odontogenic neoplasms, according to the most recent classification of head and neck tumors by the World Health Organization<sup>7</sup>.

Clinically, these lesions can go unnoticed and usually do not cause an increase in volume<sup>8</sup>. As demonstrated in the case reported, the tumor did not show significant clinical changes. The odontoma can grow slowly and cause no symptoms, making it difficult to be perceived by the patient. Thus, it is commonly diagnosed in routine radiographic exams or commonly to evaluate missing teeth, causing dental impaction<sup>2,3</sup>. However, there are cases of odontomas that cause spontaneous gingival eruption or even an increase in bone volume, which is a warning sign for the patient when looking for a diagnosis<sup>9,10</sup>. Finally, there are cases of more aggressive, atypical odontomas, especially the complex odontoma variant, which require a higher risk surgical approach due to the extent of the removal surgery and the need for stabilization of peripheral anatomical structures<sup>10</sup>.

Alves et al.,<sup>11</sup> demonstrated in their retrospective study that the age group most affected by this tumor was 21 to 40 years old, collaborating with the reported case in which the patient was 21 years old. In the same study, a higher prevalence of cases of complex odontoma was observed, however Silva et al.,<sup>12</sup> found a higher prevalence of the composite odontoma in relation to its complex form, more frequently in females, not reporting the presence of this variation of odontoma containing simultaneously areas of composite odontoma and areas of complex odontoma<sup>11,12</sup>. Thus, there is a tendency for

odontomas, mainly those composed by the anterior maxilla, with a predilection for females, while some series show a variation for complex odontomas, which may involve more frequently the posterior mandible, with a slight male predilection<sup>8,11,12</sup>. The case in question represented an uncommon variant of odontoma, which can be called compound-complex odontoma, in the anterior region of the maxilla, as reported by other authors<sup>2,13,14</sup>.

The radiographic examination is essential to assist in the correct diagnosis and elaboration of the treatment plan. The compound odontoma variant appears as a collection of radiodensity structures similar to teeth of various sizes and shapes, whereas the complex odontoma form presents as a radiopaque mass surrounded by a thin radiolucent margin, often mistaken for an osteoma. Other lesions that could be included in the radiographic differential diagnosis, especially in relation to complex odontomas, would be focal sclerosing osteomyelitis, periapical cemental dysplasia, cemento-ossifying fibroma and cementoblastoma<sup>15</sup>. In the case in question, the lesion presented radiographically with characteristics of both variants, raising the diagnostic hypothesis of compound-complex odontoma<sup>7,12,15,16,17</sup>.

Several authors agree that, as well as diagnosis, surgical removal of odontomas is indicated early to prevent further complications for the patient, since these lesions may be associated with other odontogenic tumors and cysts, and may even reach large dimensions<sup>10</sup>. Based on the technique described by Gold et al.,<sup>18</sup> early tumor enucleation was performed and the material was sent for anatomopathological evaluation. When the odontoma impacts a tooth, complete removal of the odontoma can allow normal tooth eruption of that impacted tooth, often making extractions avoidable, especially in younger patients and when orthodontic traction is possible given the positioning and shape of the from impacted tooth<sup>4</sup>. However, there are situations where the odontoma can fuse with the root of the tooth or cause changes that justify its removal, or be associated with a large radiolucent area representing a dentigerous cyst formed in the capsular tissue of the odontoma<sup>19,20</sup>. Cases of odontomas associated with a calcifying cystic odontogenic tumor may

also justify a distinct, expansive clinical behavior. Some authors indicate that computed tomography can help in this distinction due to the formation of irregular masses and contiguous hypodense areas<sup>17</sup>. However, this examination should not be sufficient to distinguish a compound-complex odontoma, so CT is useful in interpreting the boundaries of a lesion or combined lesions, but the histopathological aspect will define the diagnostic conclusion.

In histological analyses, the odontoma may exhibit a typical organization of teeth, but always showing areas of immature enamel matrix, tubular mineralized connective tissue compatible with dentin. In addition to areas of basophilic mineralization corresponding to dental cementum and loose connective tissue that can show an area of odontoblasts bordering dentin, such as islets of odontogenic epithelium and structures in the beginning of formation that resemble the enamel organ<sup>1,2,7,11</sup>. In the present case, there were areas contributing to the imaging view showing the organization of a tooth-like structure, due to the sequential arrangement of enamel matrix, dentin and connective tissue containing odontoblast cells, but containing many areas with random mineralized tissues, mainly cementoid. There were no findings that could classify the disease as any other odontogenic tumor, similar to the case of compound-complex odontoma reported by Merat et al.,<sup>13</sup>. Thus, the present case adds to the literature reports another case of compound-complex odontoma, which does not offer any change in terms of clinical behavior and therapeutic approach. But, in this case, a cystic dilation of the odontoma follicle was interpreted as a dentigerous cyst due to its dimensions and content.

In conclusion, the key point of clinical learning is highlighted, which is the fact that odontoma is the most common benign tumor of the gnathic bones and has two commonly described variants, compound and complex. The present work showed the possibility of the compound-complex combination, which can generate the combination of radiographic aspects of the two variants, but does not lead to any other clinical or therapeutic peculiarity. After the removal of the lesion, histopathological analysis is mandatory

to conclude the diagnosis of odontoma or even to determine the existence of hybrid diseases, as seen by the possibility of association between odontoma and calcifying odontogenic cyst, dentigerous cyst, among others that could alter clinical management and follow-up.

## REFERENCES

1. Silveira FM, Macedo CCS, Borges CMV, Mauramo M, Vasconcelos ACU, Soares AB, et al. Odontogenic tumors: An 11-year international multicenter study. *Oral Dis*. 2021; 27:320-4.
2. Silva FBM. Odontoma composto complexo em maxila: relato de caso. *Int j sci dent*. 2013; 1:19:9-20.
3. Jaeger F, Alvarenga RL, Lage FO, Reis IG, Leal RM. Odontoma composto - relato de caso clínico. *Rev Port Estomatol Med Dent Cir Maxilofac*. 2012; 53:252-7.
4. Kämmerer PW, Schneider D, Schiegnitz E, Schneider S, Walter C, Frerich B, et al. Clinical parameter of odontoma with special emphasis on treatment of impacted teeth—a retrospective multicentre study and literature review. *Clin Oral Invest*. 2016; 20:1827–35.
5. Sousa-Neto SS, Pereira GB, Lemes WD, Silva VHC, Felga CSF, Martins AFL. odontoma composto em adulto e sua complexidade de tratamento: relato de caso clínico. *Rev Odontol Bras Central*. 2019; 28:266-9.
6. Song, JS, Stefanik D, Damek-Poprawa M, Alawi F, Akintoyé SO. Differentiation and Regenerative Capacities of Human Odontoma-Derived Mesenchymal Cells. *Differentiation*. 2009; 77: 29–37.
7. Vered M, Fowler CB, Neville BW, Soluk TM. Odontoma. In: El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ, editors. *WHO classification of head and neck tumours*. 4th ed. Lyon, IARC, 2017. p. 224-226.
8. Iatrou I, Vardas E, Theologie-Lygidakis N, Leventis M. A retrospective analysis of the characteristics, treatment and follow-up of 26 odontomas in Greek children. *J Oral Sci*. 2010; 52: 439-47.
9. Hanemann JA, Oliveira DT, Garcia NG, Santos MR, Pereira AA. Peripheral compound odontoma erupting in the gingiva. *Head Face Med*. 2013;9:1-4.
10. Bueno NP, Bergamini ML, Elias FM, Braz-Silva PH, Ferraz EP. Unusual giant complex odontoma: A case report. *J Stomatol Oral Maxillofac Surg*. 2020;121;604-7.
11. Alves PM, Santos PPA, Cavalcanti AL, Queiroz LMG, Souza LB. Estudo clínico-histopatológico de 38 odontomas. *Rev Odontol Unesp*. 2008;37:357-361, 2008.
12. Silva DJS, Souza JJ, Almeida RAC, Andrade ESS. Estudo Clínico-Patológico de Odontomas Diagnosticados no Laboratório de Patologia Bucal da Faculdade de Odontologia de Pernambuco-FOP. *Rev Cir Traumatol Buco-Maxillofac*. 2015; 5:31-6.

13. Merat BVT, De Andrade LS, Arantes ER, Leite AFSA, Lourenço SDQC. Diagnóstico e tratamento de um caso de odontoma composto-complexo: relato de caso e revisão da literatura. *Rev Fac Odontol Lins.* 2020; 30:85-93.
14. Torul D, Keskin M, Gun S, Odabasi D. Complex-compound odontoma: A rare clinical presentation. *Odovtos Int J Dental Sci.* 2020; 22:23-8.
15. Soluk MT, Pehlivan S, Olgac V, Aksakalli N, Alatli C. Clinical and histopathological investigation of odontomas: Review of the literature and presentation of 160 cases. *J Oral Maxillofac Surg.* 2012;70:1358-61.
16. Mahl CRW, Lemos VP, Fontanella V, Miguens Jr SAQ. Odontoma Composto diagnosticado como achado radiográfico em exame interproximal: relato de caso. *R Fac Odonto Porto Alegre.* 2005;46:70-3.
17. Marques YMFS, Botelho TDL, Xavier FCDA, Rangel AL, Rege ICC, Mantesso A. Importance of cone beam computed tomography for diagnosis of calcifying cystic odontogenic tumour associated to odontoma: report of a case. *Med Oral Patol Oral Cir Bucal.* 2010; 18: 490-3.
18. Gold L, Upton GW, Marx RE. Standardized Surgical Terminology for the Excision of Lesions in Bone: an argument for accuracy in reporting. *J Oral Maxillofac Surg.* 1991; 49: 1214-7.
19. Talari BH, Ananda D, Prince CN, Annaporna CS. Fusion of complex odontome with permanent mandibular molar. *J Pharm Bioal Sci.* 2012; 4:146.
20. Dagrus K, Purohit S, Manjunatha BS. Dentigerous cyst arising from a complex odontoma: an unusual presentation. *Case Reports.* 2016:bcr2016214936. doi: 10.1136/bcr-2016-214936.

---

Corresponding Author:  
marcotullio@gmail.com  
Marco Tullio Brazão-Silva

Editor:  
Prof. Dr. Felipe Villela Gomes

Received: sep 19, 2021  
Approved: feb 23, 2022

---