

Odontomas: Clinicopathologic study of 104 cases and a case report of compound odontoma associated in an unerupted maxillary central incisor in a child

Pedro Paulo de Andrade Santos,¹ Keila Martha Amorim Barroso,² Lélia Batista de Souza,³ Márcia Cristina da Costa Miguel,³ Éricka Janine Dantas da Silveira³

Abstract

Objectives. Odontomas are benign odontogenic tumors composed of enamel, dentine, cement and pulp tissue. They are usually clinically asymptomatic, but often associated with tooth eruption disturbances. In exceptional cases the odontoma erupts into the mouth. Odontomas can be subdivided into compound and complex types. **Study design.** This paper describes a series of 104 cases of odontoma from the files of the Oral Pathology Service, Dentistry Department, Federal University of Rio Grande do Norte (UFRN), Natal-RN, Brazil, from January 1976 to March 2012. We reviewed for epidemiologic data, clinical presentation, radiographic, histopathologic characteristics and a rare case of compound odontoma associated with focal areas of ameloblastic fibroma in an unerupted maxillary central incisor. The relevant literature is reviewed. **Results.** Of the 104 cases of odontoma, 62.5% were compound and 37.5% complex. Gender distribution was 68 cases female, 35 male and 1 case this information was not available, in compound odontomas 45 cases were female and 20 male and in complex odontomas 23 cases were female and 15 male. The median age was 22 years, in complex odontoma and in compound odontoma the median was 19 years. The compound and complex odontoma arose mainly in young people. Compound odontoma occurred more frequently in the maxilla, the mandible was more frequent in complex odontoma. Most compound odontoma occurred in anterior region of maxilla and complex odontoma occurred in posterior region of mandible. **Conclusion.** Odontomas compound and complex may present as hamartomas rather than neoplasms, because they have limited growth potential, and if the odontomas which interfere with tooth eruption were extirpated early, the impacted teeth would probably erupt normally.

Key-words. Odontomas, Compound odontomas, Complex odontoma, Odontogenic tumors.

Introduction

Odontomas are considered to be developmental anomalies resulting from the growth of differentiated epithelial and mesenchymal cells.¹ Odontomas are the most common odontogenic tumors, in which all structures that form dental tissues are represented. Histologically, two types of

odontomas are recognized: complex and compound odontoma.^{2,3} Compound odontomas consist of tooth-like structures that radiographically appear as opacities. Complex odontomas comprise a mixture of odontogenic tissues without dental organization.^{2,4} Clinically, three types of odontomas are recognized in the literature: central (intraosseous) odontoma, peripheral (extraosseous or soft tissue) odontoma, and erupted odontoma.^{2,5} Odontomas are often associated with an unerupted or impacted tooth and the failure of a permanent tooth to erupt is the most common clinical manifestation.⁶ Odontomas generally appear as small, solitary or multiple radiopaque lesions detected during routine radiographic examination that may cause disturbances in tooth eruption, such as impaction, delayed eruption or retention of primary teeth.⁷ This paper describes the clinicopathologic characteristics of 104 odontomas and a rare case of compound odontoma associated with focal areas of ameloblastic fibroma in an unerupted maxillary central incisor.

¹. DDS, PhD, Oral Pathology, General Pathology, Histology and Embriology Professor of Federal University of Campina Grande (UFCG), Healthy Center and Rural Tecnology (CSTR), Estrada Patos-Teixeira Km03, Santa Cecília, CEP 58700-970, Patos-PB, Brazil.

². DDS, MSc, Oral Pathology Post-Graduate Student of Federal University of Rio Grande do Norte (UFRN). Av. Senador Salgado Filho, n. 1787, Lagoa Nova, CEP 59056-000, Natal-RN, Brazil.

³. DDS, PhD, Oral Pathology Post-Graduate Program Professor of Federal University of Rio Grande do Norte (UFRN). Av. Senador Salgado Filho, n. 1787, Lagoa Nova, CEP 59056-000, Natal-RN, Brazil.

Corresponding author:

Profa. Dra. Éricka Janine Dantas da Silveira.

Oral Pathology Post-Graduate Program Professor of Federal University of Rio Grande do Norte (UFRN). Av. Senador Salgado Filho, n. 1787, Lagoa Nova, CEP 59056-000, Natal-RN, Brazil.

Table 1. Distribution of frequency, gender and localization of odontomas.

Type	Number of cases		Gender			Localization		
	n	%	Female n %	Male n %	NA n %	Maxilla n %	Mandible n %	NA n %
Compound Odontoma	65	63	45 66	20 57	0 0	41 69	18 49	6 75
Complex Odontoma	39	38	23 34	15 43	1 100	18 31	19 51	2 25
Total	104	100	68 100	35 100	1 100	59 100	37 100	8 100

NA: Indicates not available

Materials and Methods

One hundred four cases of odontomas were retrieved from the files of the Oral Pathology Service, Dentistry Department, Federal University of Rio Grande do Norte (UFRN), Natal-RN, Brazil, from January 1976 to March 2012. This retrospective search case notes, radiographs and radiology reports, and paraffin blocks for histopathology review. The odontomas were further classified into compound and complex types according to classic definitions. Data for age, gender, location of the lesion, presence of unerupted teeth, treatment, and recurrence were obtained from information submitted with the biopsy request and from review of the dental charts and radiographs. The location of the lesion in the maxilla or mandible was classified as the anterior (incisor to canine) or posterior (premolar and molar regions). In case of larger lesions involving anterior and posterior regions we use the term anterior-posterior. The study was approved by the Ethics Committee of UFRN (protocol no. 027/10).

Results

Of the 104 cases of odontoma, 65 cases (62.5%) were compound and 39 (37.5%) complex. Gender distribution was 68 cases female, 35 male and 1 case this information was not available, in compound odontomas 45 cases were female and 20 male and in complex odontomas 23 cases were female and 15 male (Table 1). The age of the patients varied widely (range 4 to 82 years) median age of 22 years, in complex odontoma the age varied from 4 to 82 (median of 22 years) and in compound odontoma the age varied from 7 to 70 (median of 19 years). The compound odontoma arose mainly in young people between the ages of 4 to 14 years, and complex odontoma was more frequent between ages of 15 to 25 years (Table 2).

Compound odontoma occurred more frequently in the maxilla (69.5%) than in the mandible (48,6%), the mandible was more frequent in complex odontoma (51.4%) and in maxilla (30,5%). Most compound odontoma occurred in anterior region of maxilla (55%) and complex odontoma

Table 2. Age distribution of cases.

Age (years)	Compound Odontoma		Complex Odontoma		Odontomas (Total)	
	n	%	n	%	n	%
4-14	25	38	6	15	31	30
15-25	24	37	19	49	43	41
26-36	12	18	5	13	17	16
37-47	2	3	3	8	5	5
48-58	0	0	3	8	3	3
59-69	0	0	0	0	0	0
70-80	1	2	1	3	2	2
>80	0	0	1	3	1	1
NA	1	2	1	3	2	2
Total	65	100	39	100	104	100

NA: Indicates not available

Table 3. Site of distribution of odontomas.

Type	Maxilla						Mandible									
	Anterior		Premolar		Molar		Anterior		Premolar	Molar	NA	Total				
	n	%	n	%	n	%	n	%	n	%	n	%				
Compound Odontoma	36	55	0	0	5	8	9	14	0	0	9	14	65	100		
Complex Odontoma	8	21	0	0	10	26	1	3	1	3	17	44	2	5	39	100

NA: Indicates not available

Table 4. Duration distribution of cases.

Duration (months)	Compound Odontoma		Complex Odontoma		Odontomas (Total)	
	n	%	n	%	n	%
< 6	1	2	3	8	4	4
7 – 12	7	11	1	3	8	8
13 – 24	3	5	1	3	4	4
25 >	3	5	3	8	6	6
NA	51	78	31	79	82	79
Total	65	100	39	100	104	100

NA: Indicates not available

occurred in posterior region of mandible (44%), (Tables 1 and 3).

The duration of the odontomas varied widely (range 6 to 84 months), unfortunately this information wasn't available in 62% of compound odontomas and 38% of complex odontomas (Table 4).

Radiographically, compound odontoma appeared as a collection of several to numerous toothlike radiopaque structures, and complex odontoma as an amorphous, solitary mass of calcified material. All 104 cases of odontoma were treated by conservative surgical enucleation, no recurrence of the lesion was observed during a follow-up of 1 to 10 years.



Figure 1: The right maxillary primary central incisor is absent.

Case Report

An 11-year-old boy was referred to the Dentistry Department of the Federal University of Rio Grande do Norte for evaluation of delayed eruption of the permanent maxillary left central incisor (Figure 1). The boy was physically healthy and had no family history of unerupted teeth or hypodontia and no history of orofacial trauma. No signs or symptoms of temporomandibular joint disorder were noted during initial examination. Extraoral clinical examination showed no asymmetries. No oral or palatal swelling was palpable upon intraoral examination. A panoramic radiograph was obtained that revealed an irregularly shaped radiopaque mass surrounded by a thin radiolucent zone, superimposed

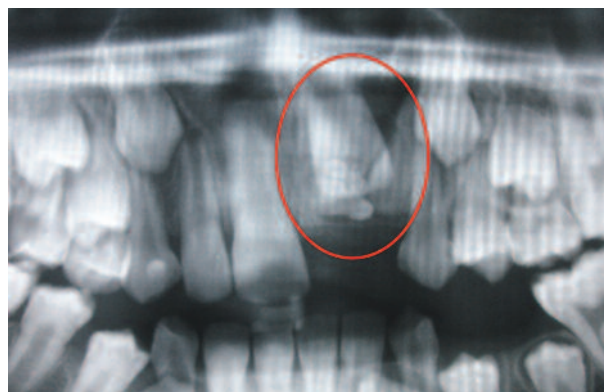


Figure 2: Panoramic radiograph taken one week after the operation; Impaction of the upper left central incisor.



Figure 3: The specimen removed surgically was composed of a few small, calcified structures encapsulated with fibrous connective tissue.

on to the crown of the left unerupted incisor [Figure 2). The provisional diagnosis of an odontoma impairing eruption of the permanent left central incisor was made, and the patient was scheduled for surgical removal of the lesion. A labial mucoperiosteal flap was raised in the region of the upper incisor under local anesthesia. Bone was removed with a bur and the lesion was excised. Macroscopic examination showed few small and calcified tooth-like structures encapsulated by fibrous connective tissue, measuring 1.5 x 1.1 x 0.4 cm in major diameter (Figure 3). Histopathologic analysis showed the presence of tubular dentin adjacent to loose pulp connective tissue (Figure 4) and an odontoblastic layer and pre-dentin at the periphery (Figure 5). Focal areas of hypercellular connective tissue showing mesenchymal

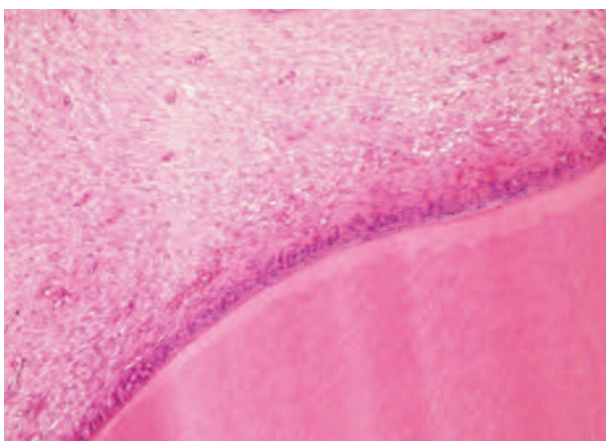


Figure 5: The inner soft and reticular connective tissue was covered by stratified epithelium resembling odontoblasts (400x)

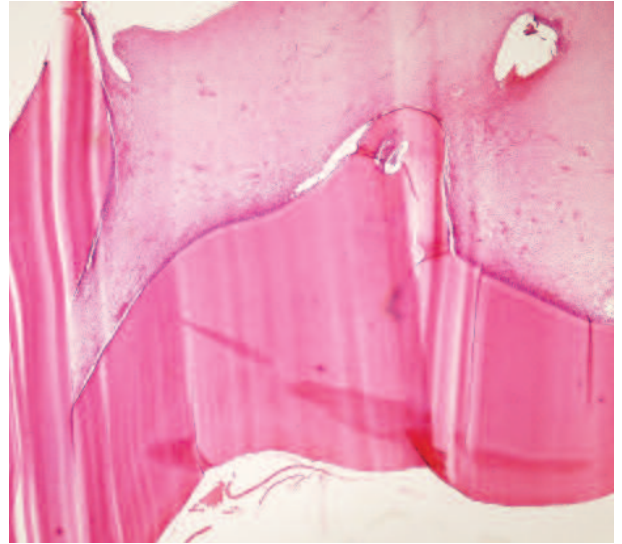


Figure 4: Microscopic examination showed a structure consisting of dentine and connective tissue resembling a pulp tissue (40x).

proliferation, islands and cords of odontogenic epithelium, and scarce stroma were also identified (Figure 6). The histopathologic diagnosis was compound odontoma. The patient presented no evidence of recurrence or complications after follow-up for one year.

Discussion

Odontogenic tumors are rare jaw lesions that affect less than 1% of the population. In children, these tumors mainly result in eruption disturbances of the permanent teeth, whereas disorders associated with the primary dentition are relatively rare.⁸ Here we investigate 104 cases of odontomas and report a rare case of compound odontoma that caused impaction of the permanent maxillary incisor in a child.

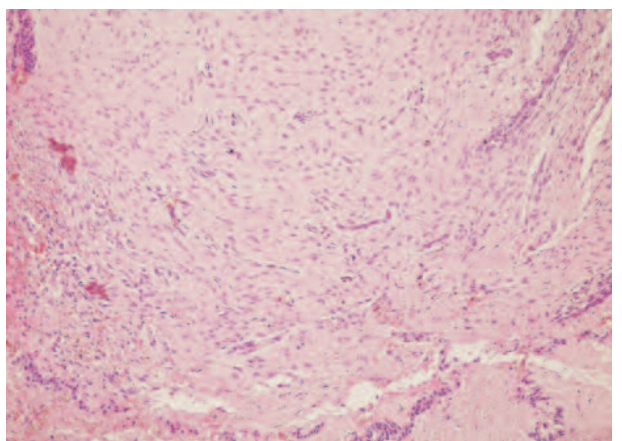


Figure 6: Histological features of ameloblastic fibroma show cords and islands of odontogenic epithelium in a primitive connective tissue stroma resembling dental papilla (200x).

Odontomas are the most common odontogenic developmental anomalies found in the jaws.⁹ The term odontoma has evolved since its introduction in 1866 and was initially applied to several different odontogenic tumors.¹⁰ According to Gyulai-Gaál et al.⁸ odontomas are benign calcifying hamartomas and are the most common type of odontogenic jaw tumors among patients younger than 20 years. In our study we observed that odontomas affected more patients younger than 25 years. And female were more affected than male by compound or complex odontomas.

With respect to location, we found that odontomas were more frequent in maxilla (59 cases) than mandible (37 cases), the area most affected by compound odontomas was anterior region of maxilla (55%) and complex odontomas in posterior region of mandible (44%), our results agree with the literature and the incidence of odontomas in the maxilla has been reported to range from 50.9 to 59.3%. In the studies of Regezi et al¹¹ and Kaugars et al¹² the most common site affected was the anterior region (incisors and canines) of the maxilla, followed by the anterior region of the mandible. Kaugars et al¹² reported that the incidence of odontomas in the molar region gradually increases with each successive decade of life.

In the study of Hidalgo-Sánchez et al¹³ the most frequent clinical manifestations were the retention of permanent teeth observed in 55.4% of the patients (more frequent in the anterosuperior region), swelling (14%), persistence of temporary teeth in the mouth (12.7%, more frequent in the anterosuperior region), agenesis of permanent teeth (7.2%), pain (4%), infection or inflammation (3.3%), malpositioned teeth (1.1%), and other unspecified manifestations (2.3%). In agreement with these authors, in the present case the odontoma was located in the region of the permanent maxillary incisor and caused impaction and delayed eruption of the tooth.

An exceptional circumstance is spontaneous eruption of an odontoma into the oral cavity, i.e., exposure of the tumor through the oral mucosa. This situation can cause pain, inflammation of adjacent soft tissues, or infection associated with suppuration.¹⁴ This was not observed in the present case because the patient was asymptomatic and the tumor did not erupt. According to Gyulai-Gaál et al.⁸ most odontomas present clinically as small lesions, cause no signs or symptoms, and are usually discovered during routine radiologic examination. However, odontomas may cause dentition anomalies such as those observed in the present case.

Radiographically, compound odontomas appear as a

collection of several to numerous tooth-like radiopaque structures, whereas complex odontomas present as an amorphous, solitary mass of calcified material. Both types of odontoma are often surrounded by a narrow radiolucent zone.¹⁵ The degree of calcification of odontomas involving the primary dentition is sometimes lower than that seen in permanent teeth, a fact resulting in the observation of only weakly radiopaque features. Careful examination of the radiographs is therefore important.⁷ In the present case, panoramic radiography revealed an irregularly shaped radiopaque mass and some remnants of rudimentary teeth superimposed onto the crown of the permanent maxillary left incisor. These characteristics led to the clinical and radiographic diagnosis of odontoma.

Suspected odontomas, both compound and complex odontomas, need to be examined microscopically to establish a definitive diagnosis. Histologically, compound odontomas often present normal appearing enamel, dentin, cementum, and pulp. Odontogenic epithelium, odontoblasts and mesenchymal pulp tissue may also be present. Complex odontomas mainly consist of mature tubular dentin. The dentin surrounds circular structures of the enamel matrix and is surrounded by cementum and a fibrous capsule as observed in the present case. The haphazard arrangement of the dental tissues is responsible for the indistinguishable clinical appearance of odontomas.¹⁶ With respect to the frequency of the different types of odontomas, we observed in this study more cases of compound than complex odontoma, Hidalgo-Sánchez et al¹³ observed the same, a clear predominance of compound odontomas over complex odontomas. The opposite was reported by Alves et al¹⁷ who identified complex odontomas in 55.3% of a series of 88 odontoma cases.

In our study we found that all 104 cases of odontoma were treated by enucleation, and according to the literature enucleation is the accepted treatment of odontomas. However, whenever the size and location of the odontoma may endanger the inferior mandibular nerve or adjacent teeth, a different approach should be considered to avoid any complications.¹⁸ In the present case related enucleation was performed without difficulties.

Ameloblastic fibroma is a distinct odontogenic tumor. It is a true neoplasm with a potential for both recurrence and malignant transformation.¹⁹ Most cases present as painless swellings or are discovered as a result of disturbances in tooth eruption. Radiographically, ameloblastic fibroma appears as a well-demarcated radiolucency, often associated with a malpositioned tooth.²⁰ Histologically, the epithelial tumor component is characterized by proliferating islands,

cords and strands of odontogenic epithelium. The strands often resemble the dental lamina during early tooth development and consist of a double or triple layer of cuboidal cells. On the other hand, the islands frequently show a peripheral row of tall cuboidal or columnar, ameloblast-like cells. The central portion of the islands may resemble the stellate reticulum of the embryonic enamel organ. The amount and density of the epithelial component may vary markedly from area to area within the same tumor. Cystic degeneration within the epithelium is not a common finding.⁴ In the present case, macroscopic examination showed focal areas of ameloblastic fibroma which, however, were not sufficient for a diagnosis of this tumor since areas of compound odontoma predominated in the specimen.

Conclusion

In this research we found that compound odontomas are more prevalent than complex odontomas, more frequent in female, affecting most anterior region of maxilla and posterior region of mandible, with a wide range of age and more frequent in young people and wide range of duration of these lesions. And in this case report, we describe a compound odontoma and a rare but possible arising ameloblastic fibroma from the fibrous capsule of an odontoma that was the cause of impaction and retention of primary or permanent tooth. Odontoma or other mixed odontogenic tumors might be the cause of tooth impaction or retention and detailed radiographic examination and treatment are necessary. An early diagnosis and management of odontomas are essential to prevent later complications, such as failure of eruption of the primary and permanent teeth.

Conflict of interest statement

The authors declare no conflict of interest.

References

- Bordini Jr J, Contar CM, Sarot JR, Fernandes A, Machado MAN (2008) Multiple Compound Odontomas in the Jaw: Case Report and Analysis of the Literature, *J Oral Maxillofac Surg* 66: 2617-2620.
- Junquera L, de Vicente JC, Roig P, Olay S, Rodríguez-Recio O (2005) Intraosseous odontoma erupted into the oral cavity: An unusual pathology, *Med Oral Patol Oral Cir Bucal* 10: 248-251.
- Kramer IRH, Pindborg JJ, Shear M (1992) Histological typing of odontogenic tumours. WHO International histological classification of tumours, Berlin: Springer-Verlag.
- Philipsen HP, Reichart PA, Praetorius F (1997) Mixed odontogenic tumours and odontomas. Considerations on interrelationship. Review of the literature and presentation of 134 new cases of odontomas, *Oral Oncol* 33:86-99.
- Daley TD, Wysoki GP, Pringle GA (1994) Relative incidence of odontogenic tumors and oral jaw cyst in a Canadian population, *Oral Surg Oral Med Oral Pathol* 77: 276-280.
- Ibsen OA, Phelan JA (2002) *Oral Pathology for the Dental Hygienist*. 3rd ed. Kuhn SA, ed Philadelphia, WB Saunders Co. pp 626-632.
- Yıldırım-Öz G, Tosun G, Kızıloğlu D, Durmuş E, Şener Y (2007) An Unusual Association of Odontomas with Primary Teeth, *Eur J Dent* 1:45-49.
- Gyulai-Gaál S, Takács D, Szabó G, Suba Z (2007) Mixed Odontogenic Tumors in Children and Adolescents, *J Craniofac Surg* 18(6):1338-1342.
- Mupparapu M, Singer SR, Rinaggio J (2004) Complex odontoma of unusual size involving the maxillary sinus: Report of a case and review of CT and histopathologic features, *Quintessence Int* 35(8):641-644.
- Broca P (1866) *Traite des tumeurs*, v.1. Paris: P Asselin. pp 350.
- Regezi JA, Kerr DA, Courtney R (1978) Odontogenic tumors: analysis of 706 cases, *J Oral Surg* 36:771-778.
- Kaugars GE, Miller ME, Abbey LM (1989) Odontomas, *Oral Surg, Oral Med, Oral Pathol* 67:172-176.
- Hidalgo-Sánchez O, Leco-Berrocal MI, Martínez-González JM (2008) Metaanalysis of the epidemiology and clinical manifestations of odontomas, *Med Oral Patol Oral Cir Bucal* 13(11): E730-734.
- Serra-Serra G, Berini-Aytés L, Gay-Escoda C (2009) Erupted odontomas: A report of three cases and review of the literature, *Med Oral Patol Oral Cir Bucal* 14(6): E299-303.
- da Silva LF, David L, Ribeiro D, Felino A (2009) Odontomas: A clinicopathologic study in a Portuguese population, *Quintessence Int* 40(1): 61-72.
- Lukes SM, Wachter KM (2003) Compound odontoma: A case study, *J Dent Hyg* 77(1):47-49.
- Alves PM, Santos PPA, Cavalcanti AL, Queiroz LMG, Souza LB (2008) Clinical histopathologic study of 38 odontomas. *Rev Odontol UNESP* 37(4):357-361.
- Casap N, Zeltser R, Abu-Tair J, Shteyer A (2006) Removal of a Large Odontoma by Sagittal Split Osteotomy. *J Oral Maxillofac Surg* 64:1833-1836.
- Chen Y, T.J. Li TJ, Gao Y, S.F. Yu (2005) Ameloblastic fibroma and related lesions: a clinicopathologic study with reference to their nature and interrelationship. *J Oral Pathol Med* 34:588-595.
- Barnes L, Eveson JW, Reichart P, Sidransky D (2005) *WHO/OMS, Pathology and Genetics of head and neck tumors*, Lyon, IARC press, pp 308-311.