

Multiple Compound Odontomas in the Jaw: Case Report and Analysis of the Literature

Jayme Bordini, Jr, DDS, PhD,* Cintia Milani Contar, DDS, MS,†
João Rodrigo Sarot, DDS, MS,‡ Ângela Fernandes, DDS, PhD,§
Maria Ângela Naval Machado, DDS, PhD¶

Odontomas are odontogenic tumors, considered to be developmental anomalies resulting from the growth of differentiated epithelial and mesenchymal cells. These tumors are formed of enamel and dentin, and can also have variable amounts of cement and pulp tissue. A compound odontoma forms an agglomeration of small structures resembling teeth, whereas a complex odontoma forms an irregular mass in a disorderly pattern. A literature review disclosed 5 cases of extensive and multiple compound odontoma previously published. An additional rare case involving jaws and erupting into the oral cavity of a 17-year-old Brazilian male is described and the clinical, radiographic, and histopathologic aspects, gender, age, location, and treatment are discussed.

Report of a Case

A 17-year-old male was referred to the Department of Pathology at the Federal University of Paraná, Brazil, by his general dentist due to a facial swelling with masticatory dysfunction and a 4-year history of bleeding gums. Past family and medical history were unremarkable. General

physical examination showed a healthy-looking male with a diffuse, hard, bone swelling on the right posterior region of the maxilla and on the right side of the chin, covered with normal skin. Intraoral examination showed some unerupted posterior teeth, and bilateral bone expansion in maxilla and mandible. In some areas there were tooth-like structures penetrating the oral mucosa (Figs 1, 2).

Radiographic examination showed multiple and diffuse tooth-like opacities occupying both jaws and maxillary sinus (Fig 3). The posterior teeth were impacted and unerupted owing to these calcified lesions. Computed tomography showed the right maxillary sinus filled almost completely with the tumor masses and minor involvement of the left maxillary sinus (Fig 4). The masses extended from both ascending rami and body of the mandible, with a major bone expansion on the right side (Fig 5).

A routine blood examination showed nothing abnormal. The serum calcium, phosphorus, and alkaline phosphatase levels also were within normal limits.

On the basis of clinical and radiologic aspects, the initial diagnosis was multiple compound odontomas and surgical treatment was indicated.

Surgical removal of the masses was accomplished under general anesthesia. Bucco-palatal mucoperiosteal flaps extending from tuberosity to tuberosity in the maxilla and bucco-lingual flaps extending from the right ramus to the

*Adjunct Professor, Department of Stomatology, Faculty of Dentistry, Federal University of Paraná, Curitiba, Brazil.

†Postgraduate Student, Department of Stomatology, School of Dentistry, Pontifical Catholic University of Paraná, Curitiba, Brazil.

‡Postgraduate Student, Department of Implantology, Faculty of Dentistry, Federal University of Santa Catarina, Curitiba, Brazil.

§Adjunct Professor, Department of Stomatology, Faculty of Dentistry, Federal University of Paraná, Curitiba, Brazil.

¶Chairman, Department of Oral Pathology, School of Dentistry, Pontifical Catholic University of Paraná, Curitiba, Brazil.

Address correspondence and reprint requests to Dr Contar: School of Dentistry, Pontifical Catholic University of Paraná, Rua Imaculada Conceição 1155, 80215901, Curitiba, PR, Brazil; e-mail: cintiacontar@hotmail.com

© 2008 American Association of Oral and Maxillofacial Surgeons

0278-2391/08/6612-0028\$34.00/0

doi:10.1016/j.joms.2007.08.027



FIGURE 1. Intraoral occlusal view, tooth-like structures penetrating the oral mucosa in maxilla.

Bordini et al. Multiple Compound Odontomas. *J Oral Maxillofac Surg* 2008.



FIGURE 2. Intraoral occlusal view, tooth-like structures penetrating the oral mucosa in mandible.

Bordini et al. Multiple Compound Odontomas. J Oral Maxillofac Surg 2008.

left ramus in the mandible were raised. All tooth-like tumor masses were excised and the impacted teeth were extracted with the enucleation of the tumor (Fig 6). The wounds were closed and the healing was uneventful.

Microscopic examination showed a structure consisting of dentine and connective tissue resembling a pulp tissue (Figs 7, 8). The inner soft and reticular connective tissue was covered by stratified epithelium resembling odontoblasts (Figs 7, 8). Based on the histopathologic features, a definitive diagnosis of compound odontoma was established.

Postoperatively, there was no evidence of recurrence or complications during a 1-year follow-up. After this period, the patient moved to another city and did not come back for implants and prosthetic reconstruction.

Discussion

In a study of 340 cases, Fernandes et al¹ determined the relative frequency of odontogenic tumors in a Brazilian population. They found 85 cases of odontomas (24.91%), 33 compound (9.7%), and 52 complex (15.3%). In a review of 104 cases of odontomas, Owens et al² identified 67 (64.4%) compound, 32 (31.0%) complex, and 5 (4.6%) diagnoses of both compound and complex odontomas. However, mul-

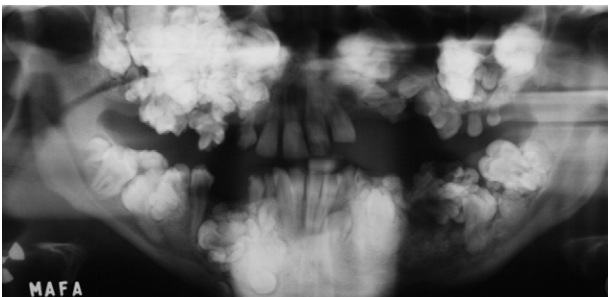


FIGURE 3. Panoramic radiograph, multiple tooth-like opacities in the jaws.

Bordini et al. Multiple Compound Odontomas. J Oral Maxillofac Surg 2008.

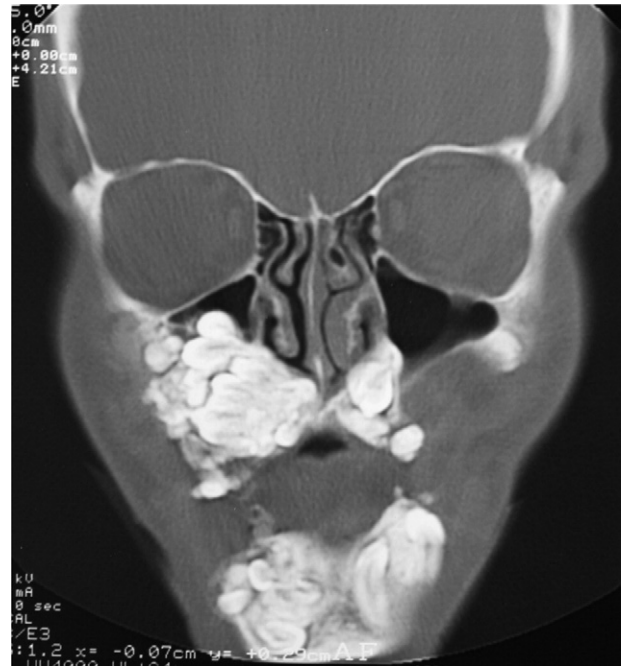


FIGURE 4. Coronal computed tomography, notice the great involvement of the right maxillary sinus.

Bordini et al. Multiple Compound Odontomas. J Oral Maxillofac Surg 2008.

iple odontomas with extensive involvement of jaws are found very rarely in humans³ and their prevalence is unknown.

A review of English literature showed 6 published reports³⁻⁷ of extensive multiple compound odonto-



FIGURE 5. Axial computed tomography, bone expansion caused by the lesions in the mandible.

Bordini et al. Multiple Compound Odontomas. J Oral Maxillofac Surg 2008.

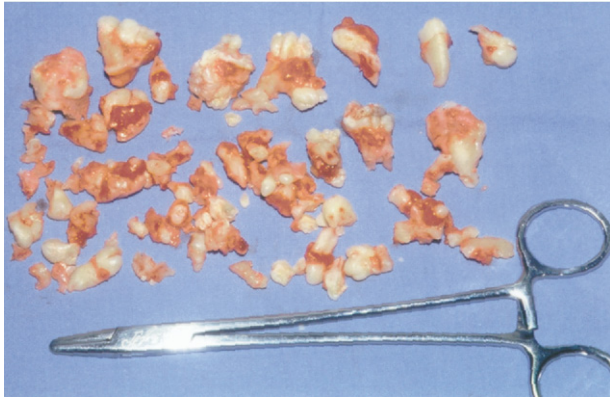


FIGURE 6. Surgical specimens removed from the mandible.

Bordini et al. Multiple Compound Odontomas. J Oral Maxillofac Surg 2008.

mas including the present case (Table 1). The cases presented by Iwamoto et al,⁸ Melnick,⁹ and Lamberg et al¹⁰ were not included in this analysis due to a minor involvement of the jaws or lack of many tooth-like structures.

The exact etiology of odontomas remains unknown, although local trauma, infection, and genetic factors have been suggested.^{2,8,11,12} Systemic syndromes such as cleidocranial dysostosis or Gardner's syndrome could be related to multiple compound odontomas.⁸ Other malformations like esophageal, pulmonary, and aortic stenosis, pneumonia, hepatopathy, and bronchiectasis were described by Bader³ and Schmidser.⁷ However, no systemic symptoms were evident in the cases reported by Ajike and Adekeye,³ Malik and Khalid,⁵ Mani,⁶ and in the present case.

Odontomas may be found at any age but are found usually in the second decade of life.^{11,3,14} Odontomas

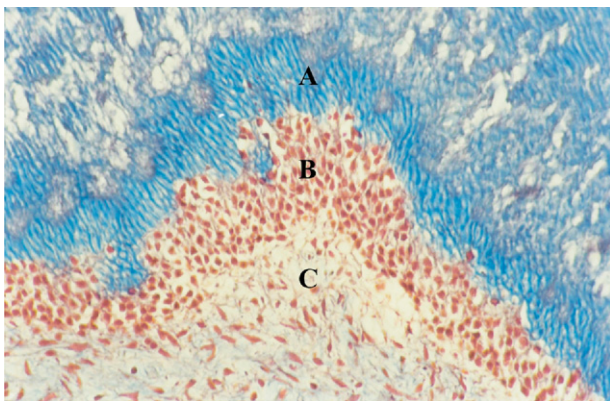


FIGURE 7. Photomicrograph showing dentine (A), odontoblasts (B) with a soft connective tissue-pulp (C). (Tricomic of Masson, original magnification 20x).

Bordini et al. Multiple Compound Odontomas. J Oral Maxillofac Surg 2008.

FIGURE 8. Photomicrograph showing loose fibrous matrix tissue, covered by stratified odontogenic epithelium resembling odontoblasts (arrow). (Tricomic of Masson, original magnification 40x).

Bordini et al. Multiple Compound Odontomas. J Oral Maxillofac Surg 2008.

occur commonly in the permanent dentition and are reported rarely in association with primary teeth.¹³ A very rare case in the primary dentition with extensive involvement of both jaws was reported by Malik and Khalid⁵ in a 7-year-old Libyan female. In 396 cases of odontoma, Katz¹⁵ found only 5 cases of compound and complex odontomas in association with unerupted primary teeth.

Most cases of odontomas occur in an intraosseous location; extraosseous odontomas are very uncommon.¹⁶ There is no apparent site predilection; however, the majority of odontomas that are located in the anterior region of maxilla are compound, whereas the great majority of odontomas located in the posterior areas, especially in the mandible, are complex odontomas.^{3,12}

Odontomas are usually asymptomatic lesions that are discovered incidentally during routine radiography.^{11,12} They are often collocated with impacted permanent teeth, with or without persistence of the primary tooth.¹⁵ However, multiple compound odontomas reported in literature show facial swelling, bone expansion, and delayed eruption of the permanent teeth.^{2,5-7} The present case is similar to the Ajike and Adekeye³ study in 2 aspects: a large extension of tumor widespread in the facial bones, and tooth-like structures penetrating the oral mucosa in some areas.

Radiographic aspects of compound odontomas are characteristic. They show calcified structures resembling teeth in the center of a well-defined radiolucent lesion.¹² The compound odontomas are surrounded usually by a narrow radiolucent zone and are associated more often with unerupted teeth.¹² It is possible, based on the radiographic features, to diagnose the tumor as a compound odontoma.¹⁴

The conservative surgical removal of compound odontomas continues to be the treatment of

Table 1. PUBLISHED REPORTS OF MULTIPLE COMPOUND ODONTOMAS IN THE LITERATURE

Authors	Age (yr)	Gender	Location	Systemic Symptoms	Treatment
Bader (1967)	Newborn	F	Both jaws	Yes	Surgical excision
Malik and Khalid (1974)	7	F	Both jaws	No	Surgical excision
Mani (1974)	19	M	Both jaws	No	Symptomatic
Schimidser et al (1975)	4	M	Both jaws	Yes	Surgical excision
Ajike and Adekeye (2000)	15	F	Both jaws	No	Surgical excision
Bordini Jr (2007)	17	M	Both jaws	No	Surgical excision

Bordini et al. Multiple Compound Odontomas. J Oral Maxillofac Surg 2008.

choice.^{3,11-14,17,18} Although every effort should be made to preserve impacted permanent teeth, in this case, all of them were removed due to their position and close association with the lesions.

Acknowledgment

The authors thank Professor Silvio Baras from the Department of Stomatology, Faculty of Dentistry, Federal University of Paraná for his cooperation during surgical procedure. Before completing the manuscript of this article, he died. We dedicate this article to his memory.

References

- Fernandes AM, Duarte ECB, Pimenta FJGS, et al: Odontogenic tumors: A study of 340 cases in a Brazilian population. *J Oral Pathol Med* 34:583, 2005
- Owens P, Schuman NJ, Mincer HH, et al: Dental odontomas: A retrospective study of 104 cases. *J Clin Pediatr Dent* 21:261, 1997
- Ajike SO, Adekeye EO: Multiple compound odontomas in the facial bones: A case report. *Int J Oral Maxillofac Surg* 29:443, 2000
- Bader G: Odontomatosis (multiple odontomas). *Oral Surg Oral Med Oral Pathol* 23:770, 1967
- Malik SA, Khalid M: Odontomatosis (multiple odontomas)—A case report. *Br J Oral Surg* 11:262, 1974
- Mani NJ: Odontoma syndrome: Report of unusual case with multiple multiform odontomas of both jaws. *J Dent* 2:149, 1974
- Schimidser R, Hausamen JE: Multiple odontogenic tumors and other anomalies. *Oral Surg Oral Med Oral Pathol* 39: 249, 1975
- Iwamoto O, Harada H, Kusukawa J, et al: Multiple odontomas of the mandible: A case report. *J Oral Maxillofac Surg* 57:338, 1999
- Melnick M: Odontomatosis. *Oral Surg Oral Med Oral Pathol* 40:163, 1975
- Lamberg MA, Syryanen SM, Ripatti LT, et al: Multiple odontomas. Review of the literature and report of a new case. *Proc Finn Dent Soc* 80: 169, 1984
- Miki Y, Oda Y, Iwaya N, et al: Clinicopathological studies of odontoma in 47 patients. *J Oral Sci* 41:173, 1999
- Oliveira BH, Campos V, Marçal S: Compound odontoma—Diagnosis and treatment: Three case reports. *Pediatr Dent* 23:151, 2001
- Yeung KH, Cheung RCT, Tsang MMH: Compound odontoma associated with an unerupted and dilacerated maxillary primary central incisor in a young patient. *Int J Pediatr Dent* 13:208, 2003
- Philipsen HP, Reichart PA, Praetorius F: Mixed odontogenic tumours and odontomas. Considerations on interrelationship. Review of the literature and presentation of 134 new cases of odontomas. *Oral Oncol* 33:86, 1997
- Katz RW: An analysis of compound and complex odontomas. *ASDC J Dent Child* 56:445, 1989
- Kintarak S, Kumplanont P, Kietthubthwe S, et al: A nodular mass of the anterior palatal gingival. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 102:3, 2006
- Shelton JT, Owens BM, Schuman NJ: Compound odontoma associated with an impacted permanent central incisor. *J Tenn Dent Assoc* 77:46, 1997
- Kamakura S, Matsui K, Katou F, et al: Surgical and orthodontic management of compound odontoma without removal of the impacted permanent tooth. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 94:540, 2002