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

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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

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

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

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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 al[1] 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 al[2] identified 67 (64.4%) compound, 32 (31.0%) complex, and 5 (4.6%) diagnoses of both compound and complex odontomas. However, multiple odontomas with extensive involvement of jaws are found very rarely in humans[3] and their prevalence is unknown.

A review of English literature showed 6 published reports[3-7] of extensive multiple compound odonto-

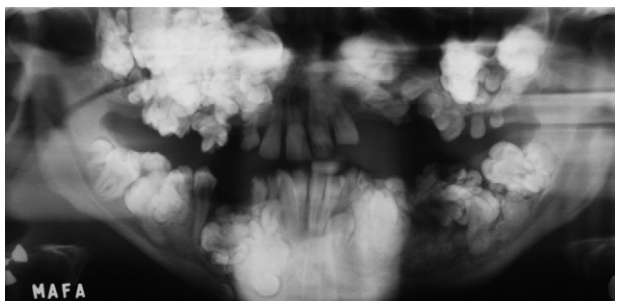

FIGURE 3. Panoramic radiograph, multiple tooth-like opacities in the jaws.

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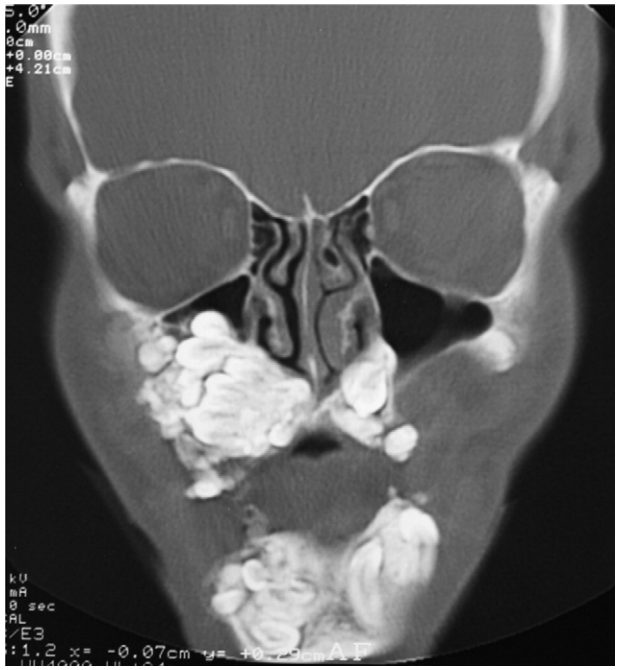

FIGURE 4. Coronal computed tomography, notice the great involvement of the right maxillary sinus.

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

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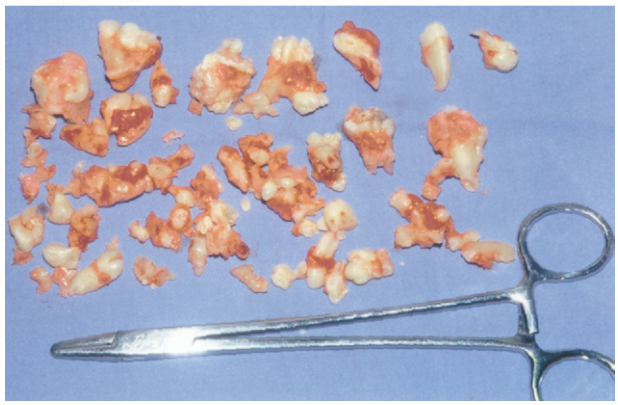

FIGURE 6. Surgical specimens removed from the mandible.

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mas including the present case (Table 1). The cases presented by Iwamoto et al,[8] Melnick,[9] and Lamberg et al[10] 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 Bader[3] and Schimidser.[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 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 Khalid[5] in a 7-year-old Libyan female. In 396 cases of odontoma, Katz[15] found only 5 cases of compound and complex odontomas in association with unerupted primary teeth.

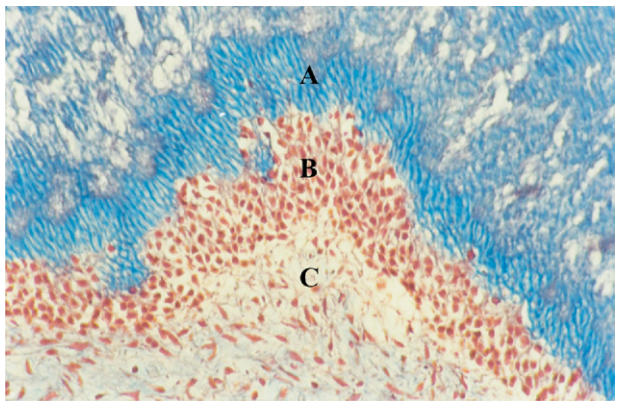

FIGURE 7. Photomicrograph showing dentine (*A*),odontoblasts (*B*) with a soft connective tissue-pulp (*C*). (Tricromic of Masson, original magnification 20x).

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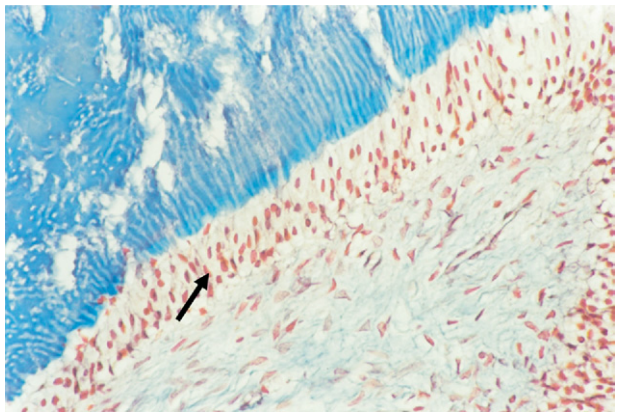

FIGURE 8. Photomicrograph showing loose fibrous matrix tissue, covered by stratified odontogenic epithelium resembling odontoblasts (*arrow*). (Tricromic of Masson, original magnification 40x).

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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 Adekeye[3] 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

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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.

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