Ameloblastic Fibro-Odontoma in Children: Report of 2 Cases

Thiago de Santana Santos, Msc
Ricardo Wathson Feitosa de Carvalho, DDS
Rafael Linard Avelar, Msc
Emanuel Dias de Oliveira e Silva, DDS
Riedel Frota, PhD *Edvaldo Dória Anjos, MSc*

ABSTRACT

Ameloblastic fibro-odontoma is a rare, mixed, benign, odontogenic tumor of significant prevalence in the mandible, with epithelial and mesenchymal components. It usually affects pediatric patients and is associated with teeth, causing a delay in eruption chronology or an alteration in the dental eruption pathway. It is occasionally diagnosed during radiographic evaluations of these patients. The literature is unclear whether it is a distinct pathological entity or a stage of odontoma. As it is benign and has a low recurrence rate, conservative treatment is recommended. The purpose of this paper was to present 2 cases of ameloblastic fibro-odontoma in the mandibles of children.

(J Dent Child 2011;78(3):173-7)

Received March 29, 2010; Last Revision July 12, 2010; Revision Accepted September 11, 2010.

KEYWORDS: ODONTOGENIC TUMORS, BENIGN JAW LESIONS, ORAL PATHOLOGY

Ameloblastic fibro-odontoma is defined by the World Health Organization (WHO) as a tumor composed of proliferating odontogenic epithelium embedded in cellular ectomesenchymal tissue that resembles dental papilla, with varying degrees of inductive change and dental hard tissue formation.[1] Clinically, this tumor behaves as a slow-growing, well-encapsulated, benign lesion and is often asymptomatic.[2]

Ameloblastic fibro-odontoma occurs predominantly in children and teenagers, with no predilection for gender.[3] It is frequently associated with erupted or displaced teeth and can reach large sizes.[4] In many cases, it is discovered through the radiographic evaluation of patients with delayed tooth eruption.[1] Radiography reveals a well-defined, radiolucent area containing various amounts of radiopaque material of irregular size and form, usually in the posterior mandible. The calcified material within may appear as multiple small forms or an opaque solid mass.[5] Management is generally conservative and consists of enucleation of the lesion and curettage of the adjacent bone. The rate of recurrence is low.[6]

Ameloblastic fibro-odontoma is considered a variant of ameloblastic fibroma. If dentin is present, the tumor is an ameloblastic fibrodentinoma. If dentin and enamel are present, the tumor is an ameloblastic fibro-odontoma.[7]

The purpose of this paper was to report 2 cases of ameloblastic fibro-odontoma in the mandibles of children. The clinical, radiographic and histological features are discussed, along with the differential diagnosis and treatment.

CASE REPORT

CASE 1

A 7-year-old male patient was sent to the Oral and Maxillofacial Surgery Department of Tiradentes Dental School, Aracaju, Sergipe, Brazil with a complaint of an increase in volume on the right side of his face that had begun 4 months earlier, with no associated toothache, fever, or pruritus in this period (Figure 1). During the patient history, neither the family nor medical history offered any

Dr. Santos is PhD Student of University of São Paulo, Dr. de Carvalho is Master's Degree Student of Pernambuco Dental School, Dr. Avelar is PhD Student of Pontifícia Católica University, Dr. e Silva is Professor of Pernambuco Dental School, and Dr. Frota is Oral and Maxillofacial Surgeon of Oswaldo Cruz Academic Hospital, all in the Oral and Maxillofacial Surgery Program, Dentistry College of Pernambuco, Recife, Pernambuco, Brazil; and Dr. Anjos is Professor of Tiradentes Dental School, Oral and Maxillofacial Surgery Program, University of Tiradentes, Aracaju, Sergipe, Brazil.
Correspond with Dr. Santos at thiago.ctbmf@yahoo.com.br

relevant data. During the oral examination, edema was noted in the buccal area of the mandible's posterior right region. The tumor was painless upon palpation. The permanent mandibular right first molar was absent, and the other teeth had no mobility. The buccal mucosa was intact and of a normal appearance.

Radiographic examination showed a clear, well-circumscribed, radiolucent lesion, extending from the parasymphysis region to the mandibular incisors and exhibiting an internal radiopaque mass. The lesion was surrounded by healthy bone (Figure 2). The lesion caused the separation of the unerupted permanent mandibular right first molar and second premolar and contained an opaque mass approximately 3 cm at its widest point. There was also an unerupted permanent tooth (presumably the permanent first molar) in the mandible's basilar region.

An incisional biopsy was performed with the histopathological diagnosis of ameloblastic fibro-odontoma (Figure 3). Under general anesthesia, complete removal and curettage of the lesion were performed with great care due to the risk of mandible fracture (Figure 4A). The permanent mandibular right first molar and second premolar had to be removed during surgery because of their union with the tumor mass (Figure 4B). After the immediate postoperative period, the patient did not return for follow-up.

CASE 2

An 8-year-old male patient was sent to the Oral and Maxillofacial Surgery Department of Pernambuco Dental School, Camaragibe, Pernambuco, Brazil with the complaint of an increase in volume on the chin that had begun 6 months earlier, with no associated toothache, fever, or pruritus in this period. When the patient's history was obtained, neither the family nor medical history offered any relevant data. During the extraoral examination, a nevus was found, and the patient was sent for dermatological evaluation, which confirmed the hypothesis of nevus (Figure 5). During the oral examination, there was an increase in volume in the buccal area of the anterior mandible. The tumor was painless upon palpation. The lower left incisor was absent. There was no mobility in the adjacent teeth. The buccal mucosa was intact and of a normal appearance.

Radiographic examination revealed a clear, well-circumscribed, radiopaque lesion extending from the right parasymphysis region to the mandibular body and well defined by surrounding bone (Figure 6A-B). The lesion caused separation of the central and lateral right incisors and right canine and contained an opaque mass approximately 2 cm at its widest point. Computed tomography revealed a lesion with an internal radiodense mass surrounded by a well-defined sclerotic margin of healthy surrounding bone . Some areas of the lesion were not completely filled by the tumor mass. A 3-D reconstruction showed mandible deformation (Figure 7A-D).

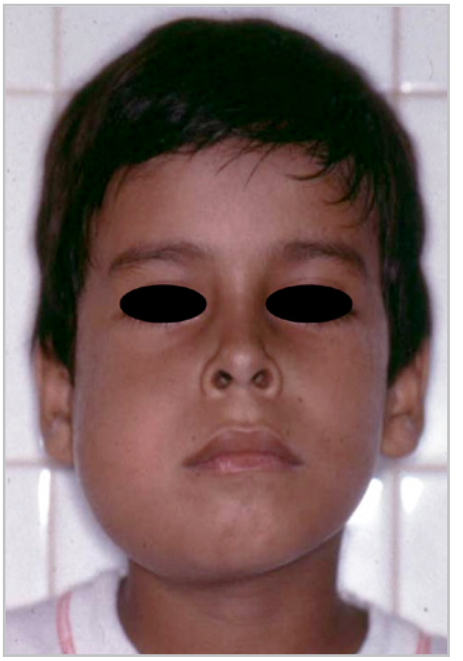

Figure 1. Frontal view showing an increase in volume on the right side of the face.

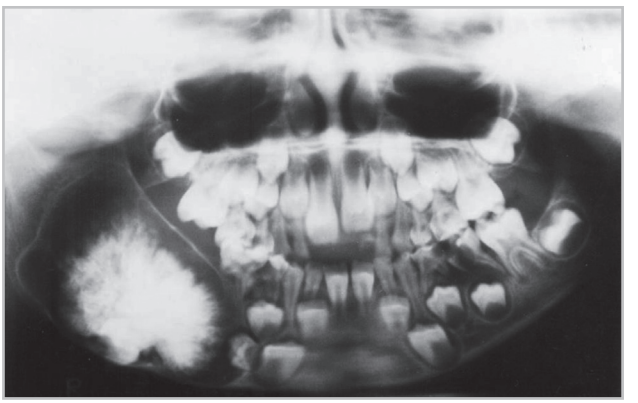

Figure 2. Panoramic radiograph with well-circumscribed radiolucent lesion extending from the parasymphysis region to the mandibular incisors with an internal radiopaque mass delimited by surrounding bone.

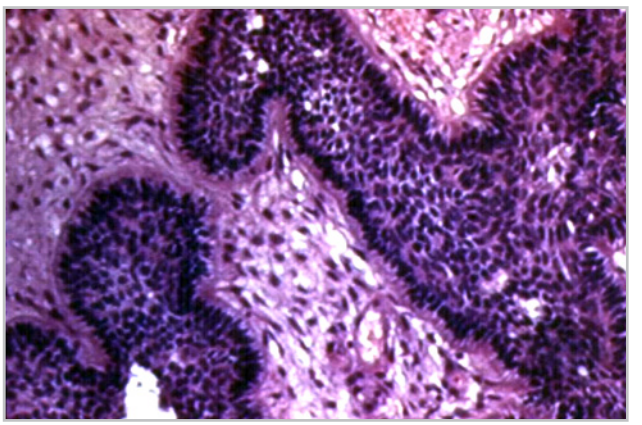

Figure 3. Greater magnification revealing an island of cuboidal to columnar odontogenic epithelium surrounded by loosely arranged stromal spindle cells with angular nuclei and bland cytology (hematoxylin—eosin: 400x).

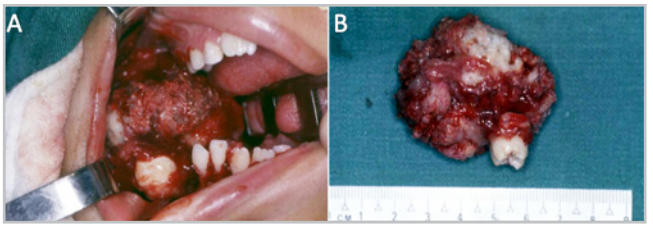

Figure 4. (A) Removal of the lesion. (B) Tumor mass removed.

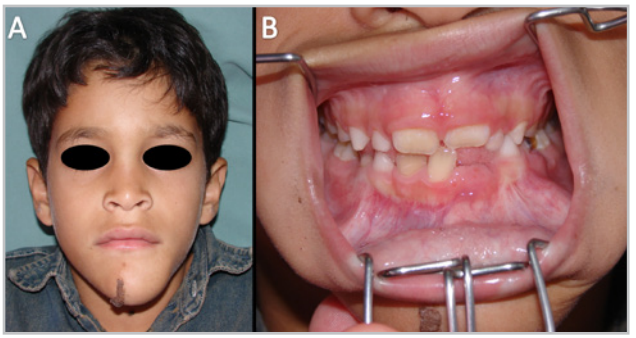

Figure 5. (A) Frontal view showing an increase in volume of the chin. (B) An increase in volume in the buccal area of the anterior mandible.

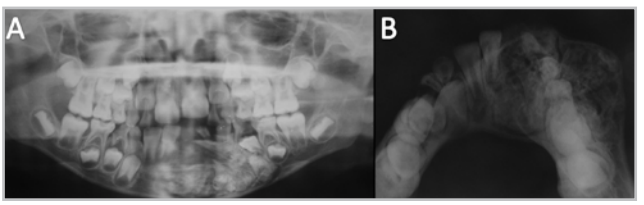

Figure 6. (A) Panoramic radiograph showing a clear, well-circumscribed radiopaque lesion extending from the right parasymphysis region to the mandibular body. (B) Occlusal radiograph showing the anterior extension of the tumor.

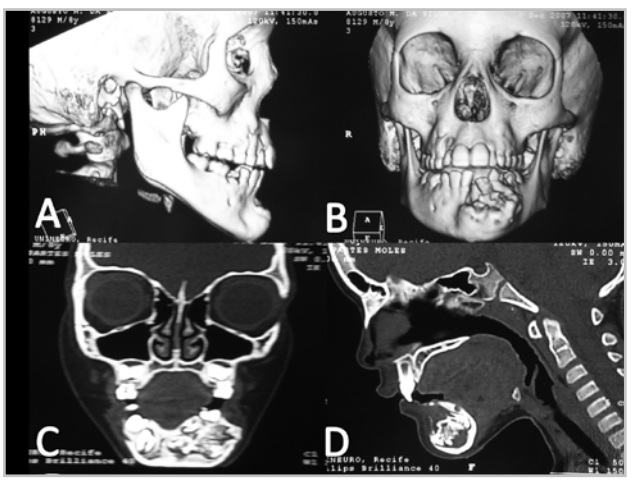

Figure 7. A lesion in computed tomography showing mandible deformation. (A) Lateral view of reconstruction. (B) Frontal view of reconstruction. (C) Crown view. (D) Sagittal view.

An incisional biopsy (Figure 8a) was performed in 2 different regions of the tumor, with the histopathological diagnosis of an ameloblastic fibro-odontoma (Figure 8b). Under general anesthesia, the complete removal and curettage of the lesion were performed (Figure 9a-b). The patient has been under the care of the clinic for 3 years, with no clinical or tomographic signs of recurrence to date (Figure 10a-b).

DISCUSSION

Ameloblastic fibro-odontoma is a rare, benign, mixed, odontogenic tumor that accounts for approximately 3% of all odontogenic tumors. Average age at diagnosis is 9 years.[2,8] There is no predilection for gender.[3,5] Both cases described here involved males with similar ages (7 and 8 years).

There has been much discussion in the literature regarding the proper classification of this tumor.[9] One point of discussion is the discrimination between neoplasm and hamartoma.[10] According to Philipsen et al. and Slootweg, ameloblastic fibro-odontoma has a hamartomatous nature, whereas ameloblastic fibroma has a neoplastic nature.[8,11] According to Oghli, ameloblastic fibroodontoma is currently designated as a separate entity,but may be histologically indistinguishable from an immature complex odontoma.[12] The relative arrangement of the soft tissues and the stage of development of the teeth involved are useful criteria for the diagnosis. According to the revised WHO classification, ameloblastic fibro-odontoma is a benign tumor without invasive growth, by contrast to ameloblastoma.[1] Osseous expansion with no destruction of the adjacent structures in both cases described demonstrates the benign characteristic, with no locally aggressive behavior. The authors in the literature believe that the best way to make this distinction can and should be based on the clinical and radiologic correlation.

When analyzing the frequency of lesions, epidemiological studies are needed to delineate the demographic profile of a pathologic lesion. Sousa et al., carried out a review of 2,356 biopsies from pediatric patients and found that odontoma was the most frequent tumor in the group of odontogenic tumors.[13] Adebayo et al. analyzed 318 cases of odontogenic tumors treated at the maxillofacial surgery unit of the hospital of the University of Ahmadu Bello, Kaduna, Nigeria, and found that ameloblastic fibro-odontoma was the least frequent odontogenic lesion.[14] The authors of the present article published a previous paper with a review of 238 odontogenic tumors in northeastern Brazil, only one of which was an ameloblastic fibro-odontoma, underscoreing the rare occurrence of this tumor.[15]

Although this lesion can occur in the maxilla, the mandible is the most frequent site.[5,16] According to Neville et al. and al-Sebaei and Gagari, this tumor occurs most commonly in the mandible's posterior region, as in the first case reported here.[5,17]

Ameloblastic fibro-odontoma is usually associated with teeth, causing delayed eruption and changes in the eruption pathway.[4,18] The tumor mass has slow progression and generally produces an asymptomatic increase in volume.[5] When large, however, it may cause discomfort. Tooth mobility may also be present.[19] Both cases reported here

had unerupted teeth in the region of the growing tumor mass, but without symptoms. There was also no tooth mobility in either patient.

In many cases, these lesions are diagnosed through radiography in patients with delayed tooth eruption.[19] Radiography generally reveals a well-defined radiolucent area containing various amounts of radiopaque material of irregular size and form. The ratio of radiopaque to radiolucent areas differs from one lesion to another; sometimes the mineralized element in the tumor predominates and the lesion may resemble an odontoma.[5,2] In the second case reported here, the radiographic aspect led to the diagnostic hypothesis of odontoma. Computed tomography, however, suggested ameloblastic fibro-odontoma due to the aspect of a radiodense mass in the middle of the tumor.

Traditionally, ameloblastic fibro-odontoma has been classified as a benign, mixed, odontogenic tumor. The term "epithelial odontogenic tumor with odontogenic mesenchyme" is currently becoming more widely accepted and avoids potential controversy over the nature of the tumor. The term "ameloblastic fibro-odontoma" represents a histological combination of ameloblastic fibroma and a complex odontoma. This lesion exhibits the same benign biological behavior as an ameloblastic fibroma. By contrast, the term "ameloblastic odontoma" refers to a tumor exhibiting a histological combination of ameloblastoma and complex odontoma, which behave in the invasive manner of classic ameloblastomas.[21]

Histologically, ameloblastic fibro-odontoma has characteristics similar to those of an ameloblastic fibroma and complex odontoma.[7,19] It exhibits odontogenic epithelial islands and strings in a mesenchymal stroma rich in cells, with enamel and dentin matrix formation in close relation to epithelial structures. It is questionable whether these lesions are distinct pathologies or variants of ameloblastic fibro-odontoma.[5] According to al-Sebaei and Gagari, ameloblastic fibroma, ameloblastic fibro-odontoma and odontoma should be considered distinct pathologies.[17] According to Silva et al., however, such lesions should be classified in the odontoma group, like complex and simple odontomas.[22]

It is impossible to differentiate ameloblastic fibro-odontoma, ameloblastic fibroma, and ameloblastic odontoma by radiology alone. Therefore, a histopathological examination is essential.[5,7,19,22] In the cases described here, the histological characteristics of the lesions were similar to those of ameloblastic fibroma and the diagnosis was based on histological and radiographic features, in which a calcified mass in the interior was visible where the biopsies were performed. On the other hand, dental hard tissue was absent in the histological slides, probably due to the region of the preparation of histological sections. The radiological and histological information, however, was sufficient to elucidate the diagnosis.

Ameloblastic fibro-odontoma has a benign biological behavior with a well-defined limited mass in the radiographic image and is easily separated from the surrounding bone. Consequently, a conservative approach involving enucleation of the lesion and mechanical curettage of the surrounding bone is the treatment of choice.[3,5,6,17] This is unlike the treatment some authors recommend for ameloblastic fibroma, with wide excision of the tumor unless the extent of the surgery would result in significant cosmetic deformity.[23] In the first case reported here, the lesion was associated with the unerupted permanent mandibular first molar, which was removed together with the premolars. The first molar was removed because of its relation to the tumor mass, and the premolars were removed because of their unfavorable location, which led to the eruption of other delayed teeth. Hawkins and Sadeghi describe a more conservative treatment in relation to the removal of other teeth.[20] The treatment employed in the second case was conservative and preserved the adjacent structures.

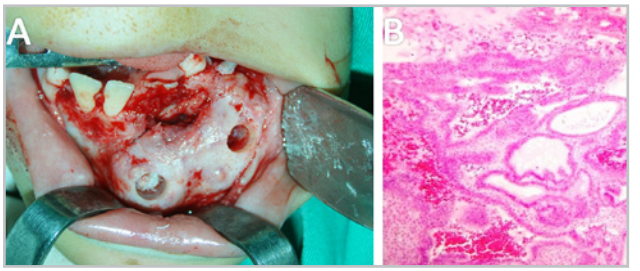

Figure 8. (A) Incisional biopsy with a trephine drill. (B) Low-power magnification of tumor showing fibrous stroma containing strands and nests of odontogenic epithelium (hematoxylin-eosin: 100x).

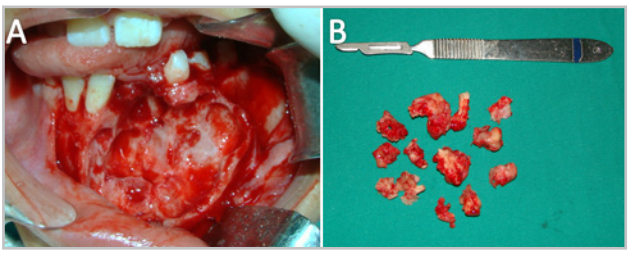

Figure 9. (A) Removal of the lesion. (B) The tumor mass removed.

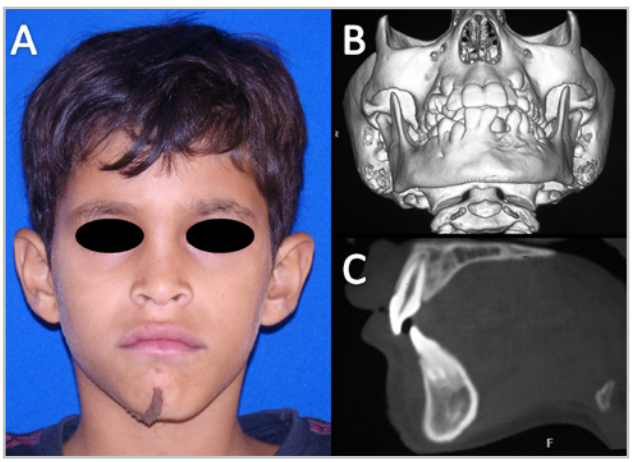

Figure 10. Three-year follow up. (A) Frontal view. (B) Frontal view of the reconstruction (computed tomography). (C) Sagittal view (computed tomography).

According to Dhanuthai and Kongin, recurrence is rare following a conservative approach with enucleation and curettage of the lesion.[3] There are reports, however, of sarcoma degeneration.[4,24] Ueki et al., report a case of malignant ameloblastic fibrosarcoma in a dog, with lung, liver, and orbit cavity metastasis that led to the death of the animal.[4] Sozeri et al., published a case of an ameloblastic fibrosarcoma in the mandible of a 5-year-old child; despite repeated surgical interventions, the tumor recurred 3 times within a year and a half, but no metastases were observed.[24] The treatment of choice in such cases seems to be radical surgery.

Although the rates of recurrence and sarcomative transformation are low, cases have been reported.[4,24] This makes long-term follow up essential.[17] The patient in the first case reported here did not return for follow up. The patient in the second case has been under the care of the clinic for 3 years, with no clinical or tomographic signs of recurrence to date. This case, however, requires a longer follow-up period to ensure the lack of recurrence.

In conclusion, ameloblastic fibro-odontoma exhibits clinical and radiographic features that may not be conclusive, and its diagnosis is only confirmed by histological examination. It remains a controversial tumor, as some authors claim that ameloblastic fibroma and complex odontoma are different stages of the same pathology, while others believe that these lesions are different pathologies. The most important point is that, whatever the classification, the treatment must be conservative in children, and aggressive surgery is not justified, considering the benign behavior, low recurrence rate, and potential of cosmetic deformity in young patients. Moreover, long-term follow up is recommended.

REFERENCES

1. Barnes L, Eveson J W, Reihcart P, Sidransky D, eds. World Health Organization International Histological Classification of Tumors. Lyon , France: IARC Press; 2005.
2. Yagishita H, Taya Y, Kanri Y, et al. The secretion of amelogenins is associated with the induction of enamel and dentinoid in an ameloblastic fibro-odontoma. J Oral Pathol Med 2001;30:499-503.
3. Dhanuthai K, Kongin K. Ameloblastic fibro-odontoma: A case report. J Clin Pediatr Dent 2004;29:75-7.
4. Ueki H, Sumi A, Takaishi H, Ito H, Oyamada T, Yoshikawa H. Malignant ameloblastic fibro-odon-toma in a dog. Vet Pathol 2004;41:183-5.
5. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. New York, NY: Elsevier; 2009.
6. Sivapathasundharam B, Manikandhan R, Sivakumar G, George T. Ameloblastic fibro-odontoma. Indian J Dent Res 2005;16:19-21.
7. Pereira KD, Bennett KM, Elkins TP, Qu Z. Ameloblastic fibroma of the maxillary sinus. Int J Pediatr Otorhinolaryngol 2004;68:1473-7.
8. Philipsen HP, Reichart PA, Praetorius F. Mixed odontogenic tumors and odontomas. Considerations on inter-relationship: Review of the literature and presentation of 134 new cases of odontomas. Oral Oncol 1997;33:86-99.
9. Olech E, Alvares O. Ameloblastic odontoma: Report of a case. Oral Surg Oral Med Oral Pathol 1967;23: 487-92.
10. Schmidseder R, Hausamen JE. Multiple odontogenic tumors and other anomalies: An autosomal dominantly inherited syndrome. Oral Surg Oral Med Oral Pathol 1975;39:249-58.
11. Slootweg PJ. Analysis of the inter-relationship of mixed odontogenic tumors: Ameloblastic fibroma, ameloblastic fibro odontoma, and the odontomas. Oral Surg 1981;51:266-76.
12. Oghli AA, Scuto I, Ziegler C, Flechtenmacher C, Hofele C. Ameloblastic fibro-odontoma: A large ameloblastic fibro-odontoma of the right mandible. Med Oral Patol Oral Cir Bucal 2007;12:E34-7.
13. Sousa FB, Etges A, Corrêa L, Mesquita RA, Araújo NS. Pediatric oral lesions: A 15-year review from São Paulo, Brazil. J Clin Pediatr Dent 2002;26:413-8.
14. Adebayo ET, Ajike SO, Adekeye EO. A review of 318 odontogenic tumors in Kaduna, Nigeria. J Oral Maxillofac Surg 2005;63:811-9.
15. Avelar RL, Antunes AA, Santos TS, Andrade ESS, Dourado E. Odontogenic tumors: Clinical and pathology study of 238 cases. Rev Bras Otorrinolaringol 2008;74:668-73.
16. Skiavounou A, Iakovou M, Kontos-Toutouzas J, Kanellopoulou A, Papanikolaou S. Intraosseous lesions in Greek children and adolescents: A study based on biopsy material over a 26-year period. J Clin Pediatr Dent 2005;30:153-6.
17. al-Sebaei MO, Gagari E. Ameloblastic fibro-odontoma. J Mass Dent Soc 2001;50:52-3.
18. Flaitz CM, Hicks J. Delayed tooth eruption associated with an ameloblastic fibro-odontoma. Pediatr Dent 2001;23:253-4.
19. Chang H, Precious DS, Shimizu MS. Ameloblastic fibro-odontoma: A case report. J Can Dent Assoc 2002;68:243-6.
20. Hawkins PL, Sadeghi EM. Ameloblastic fibro-odontoma: Report of a case. J Oral Maxillofac Surg 1986;44:1014-9.
21. Riu GD, Meloni SM, Contini M, Tullio A. Ameloblastic fibro-odontoma: Case report and review of the literature. J Craniomaxillofac Surg 2010;38:141-4.
22. Silva GCC, Jham BC, Silva EC, Horta MCR, Godinho SHP, Gomez RS. Ameloblastic fibro-odontoma. Oral Oncol Extra 2006;42:217-20.
23. Zallen RD, Preskar MH, McClary SA. Ameloblastic fibroma. J Oral Maxillofac Surg 1982;40:513-7.
24. Sözeri B, Ataman M, Ruacan S, Gedikoglu G. Ameloblastic fibrosarcoma. Int J Pediatr Otorhinolaryngol 1993;25:255-9.