

Ameloblastic fibro-odontoma in a child patient: case report and literature review

Fibro-odontoma ameloblástico em paciente infantil: relato de caso e revisão da literatura

Fibroodontoma ameloblástico en un paciente infantil: reporte de un caso y revisión de la literatura

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Abstract

The ameloblastic fibro-odontoma (AFO) is a mixed odontogenic tumor, with characteristics of ameloblastic fibroma, presenting enamel and dentin, which occurs more frequently in individuals aged 5 to 17 years. This paper reports an extensive case of ameloblastic fibro-odontoma in the mandible of a 3-year-old patient, discussed in comparison to cases selected from a brief literature review on the clinical characteristics, Evolution and therapeutic options for this lesion. In the last years, there was no consensus in the literature concerning its etiopathogenesis and classification, yet recently the AFO was classified as a developing odontoma. This case is in accordance with the 7 cases reported in the literature of AFO in the mandible of children aged 10 years or younger, especially concerning the lesion pattern and evolution and treatment adopted. The patient did not present relapse and exhibited local bone regeneration at the 3-year follow-up.

Keywords: Ameloblastic fibro-odontoma; Odontogenic tumors; Mandibular neoplasms.

Resumo

Fibro-odontoma ameloblástico (FOA) é um tumor odontogênico misto, com características de um fibroma ameloblástico, apresentando esmalte e dentina, que acomete mais frequentemente, pacientes entre 5 e 17 anos de idade. Este trabalho tem por objetivo relatar um caso de extenso fibro-odontoma ameloblástico localizado na mandíbula em um paciente de 3 anos de idade e discuti-lo frente a casos selecionados em uma breve revisão da literatura sobre as características clínicas, comportamentais e opções terapêuticas desta lesão. Nos últimos anos a literatura não apresentava consenso em relação a sua etiopatogenia e classificação, porém, recentemente o FOA foi classificado como odontoma em desenvolvimento. O presente caso está de acordo com os 7 casos relatados na literatura de FOA mandibulares de crianças com idade igual ou inferior a 10 anos, principalmente em relação ao padrão e comportamento da lesão e tratamento escolhido. O paciente não apresentou recidiva e apresenta regeneração óssea local em um pós-operatório de 3 anos.

Palavras-chave: Fibro-odontoma ameloblástico; Tumores odontogênicos; Neoplasias mandibulares.

Resumen

El fibroodontoma ameloblástico (AFO) es un tumor odontogénico mixto, con características de fibroma ameloblástico, que presenta esmalte y dentina, que se presenta con mayor frecuencia en individuos de 5 a 17 años. En este trabajo se reporta un extenso caso de fibroodontoma ameloblástico en la mandíbula de un paciente de 3 años,

discutido en comparación con casos seleccionados de una breve revisión de la literatura sobre las características clínicas, evolución y opciones terapéuticas de esta lesión. En los últimos años, no hubo consenso en la literatura sobre su etiopatogenia y clasificación, sin embargo, recientemente el AFO fue clasificado como un odontoma en desarrollo. Este caso concuerda con los 7 casos reportados en la literatura de AFO en la mandíbula de niños de 10 años o menos, especialmente en lo que respecta al patrón de lesión y evolución y tratamiento adoptado. El paciente no presentó recidiva y presentó regeneración ósea local a los 3 años de seguimiento.

Palabras clave: Fibroodontoma ameloblástico; Tumores odontogénicos; Neoplasias mandibulares.

1. Introduction

The ameloblastic fibro-odontoma (AFO) is a benign and rare odontogenic tumor, more frequently observed in individuals aged 5 to 17 years. It is considered a mixed tumor, because it presents characteristics of an ameloblastic fibroma, yet associated with variable degrees of enamel and dentin maturation (Buchner et al., 2013; De Riu et al., 2010).

The AFO usually affects the posterior mandibular region, presenting as an asymptomatic volume increase, possibly causing facial asymmetry, delayed tooth eruption and altered positioning of adjacent teeth (Manor et al., 2012). Radiographically, it presents as a unilocular, well-defined radiolucent area, with different levels of radiopacity, possibly associated with the crown of an impacted tooth. The radiopacity may be predominant, thus similar to an odontoma. The treatment involves surgical curettage/enucleation with removal of involved teeth in cases of extensive tumors. The relapse is uncommon and malignant transformation is rare, being more common in adult patients (Boxberger et al., 2011).

2. Methodology

This paper reports a case of extensive ameloblastic fibro-odontoma in a child patient and reviews the literature on the treatment options (Pereira et al., 2018). The literature review included searches in the databases *Medline* and *Lilacs* and manual search in the main journals related to the field (*Oral Surgery*, *Oral Medicine*, *Oral Pathology*, *Oral Radiology*; *International Journal of Oral and Maxillofacial Surgery*; *Journal of Oral and Maxillofacial Surgery*; *Head & Neck*; *Medicina Oral, Patología Oral Y Cirugía Bucal*; *Journal of Cranio-Maxillo-Facial Surgery*; *European Archives of Paediatric Dentistry* and *Journal of Dentistry for Children*). The MeSH terms used were *ameloblastic fibro-odontoma OR ameloblastic fibroodontoma OR ameloblastic fibro odontoma OR ameloblastic fibro-odontome OR ameloblastic fibroodontoma*. The survey included all studies conducted in humans, published in English language, in the period July 2004 to July 2019, addressing AFO affecting the mandible of child patients (up to 10 years of age). The legal guardian for the patient signed the consent form, allowing participation in the study prior to the start of treatment, as well as the procedures followed were in accordance with ethical standards in human research according to the Helsinki Declaration of 1975, as revised in 1983.

3. Results and Discussion

Case report

Male patient, aged 3 years, was evaluated presenting a painless swelling on the left mandibular body, causing facial asymmetry. Intraoral examination revealed increased volume on the left mandibular body, posterior to tooth 75, involving the buccal and lingual regions.

The computed tomography evidenced an extensive, well-defined, unilocular, hypodense lesion containing scarce hyperdense material, measuring 3.8x2.7x5.4 cm. The lesion involved the left mandibular body, extending from the deciduous mandibular second molar to the ipsilateral mandibular body, surrounding the coronoid process up to the subcondylar region, with expansion of buccolingual cortical plates throughout its extent. The lesion was associated with the developing permanent

first molar, which was displaced to the mandibular base. Also, the deciduous second molar presented root resorption (Figure 1). Before first surgery those responsible for the patient signed informed consent.

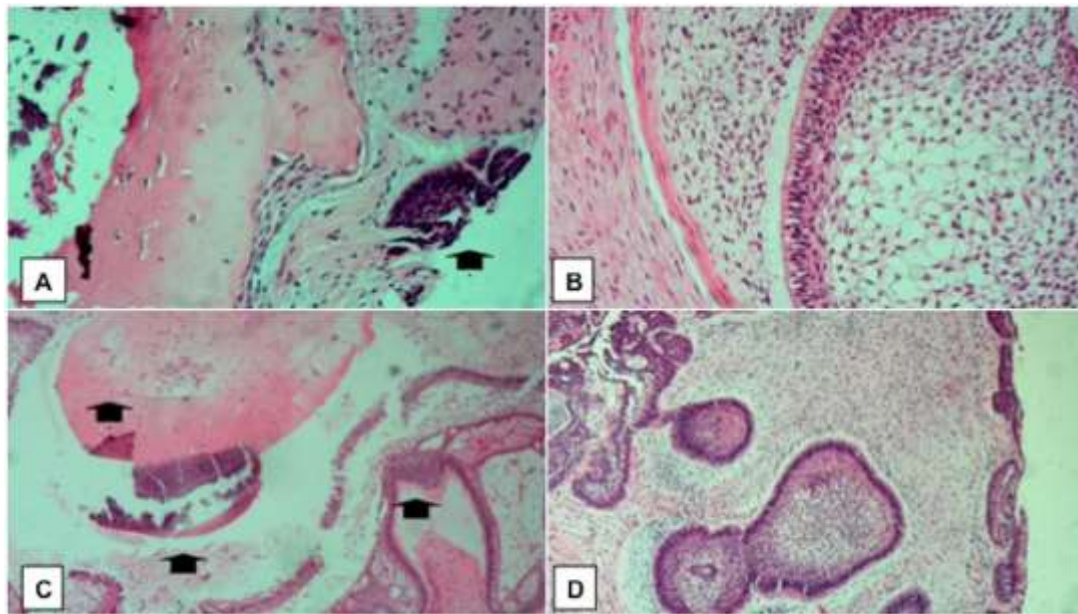
Figure 1: Initial cone beam computed tomography (CBCT). **A:** Axial reconstruction evidencing unilocular well-circumscribed lesion, containing calcified material compatible with odontogenic tissue. Observe the expansion of cortical plates in buccolingual direction. **B:** Coronal reconstruction exhibiting extensive lesion involving the left mandibular body and ramus. **C:** 3D reconstruction.



Source: Authors.

The first surgical stage comprised incisional biopsy of the lesion, under general anesthesia. Aspiration before biopsy was negative, thus incisional biopsy was performed by intraoral access on the left mandibular body region, posterior to the deciduous second molar. The lesion fragment was submitted to histopathological analysis. The histological analysis evidenced proliferation of islands and cords of odontogenic epithelium, exhibiting as peripheral palisade with freely arranged central cells, similar to the stellate reticulum of the enamel organ embedded in stroma rich in myxoid cells similar to the dental papilla. There was also amorphous eosinophilic tissue compatible with dentin formation. The diagnosis was conclusive for ameloblastic fibro-odontoma (Figure 2).

Figure 2: Photomicrograph of lesion diagnosed as ameloblastic fibro-odontoma. **A:** Presence of amorphous eosinophilic tissue comparable with dentin formation with mineralized structures (arrow). (40x) **B:** Odontogenic epithelium in palisade with freely arranged central cells, similar to the stellate reticulum of the enamel organ (40x). **C:** Dentin and presence of mineralized structures (10x). **D:** Proliferation of islands and cords of odontogenic epithelium embedded in stroma rich in myxoid cells similar to the dental papilla (4x).



Source: Authors.

The definitive treatment was performed on a second surgery by lesion enucleation. Surgery was performed under general anesthesia, with intraoral buccal access on the left mandibular body, with complete lesion removal and extraction of the permanent first molar, which remained in the cavity after lesion removal without stability, which precluded its maintenance. The buccal and lingual cortical plates were maintained, assuring the integrity of mandibular contour (Figure 3).

Figure 3: Surgical treatment of the lesion. **A:** Aspect of lesion after incision, displacement and osteotomy for identification. **B:** Surgical bed after excision and curettage of the lesion in close association with tooth 36. **C:** Specimen sent to histopathological analysis and tooth 36.

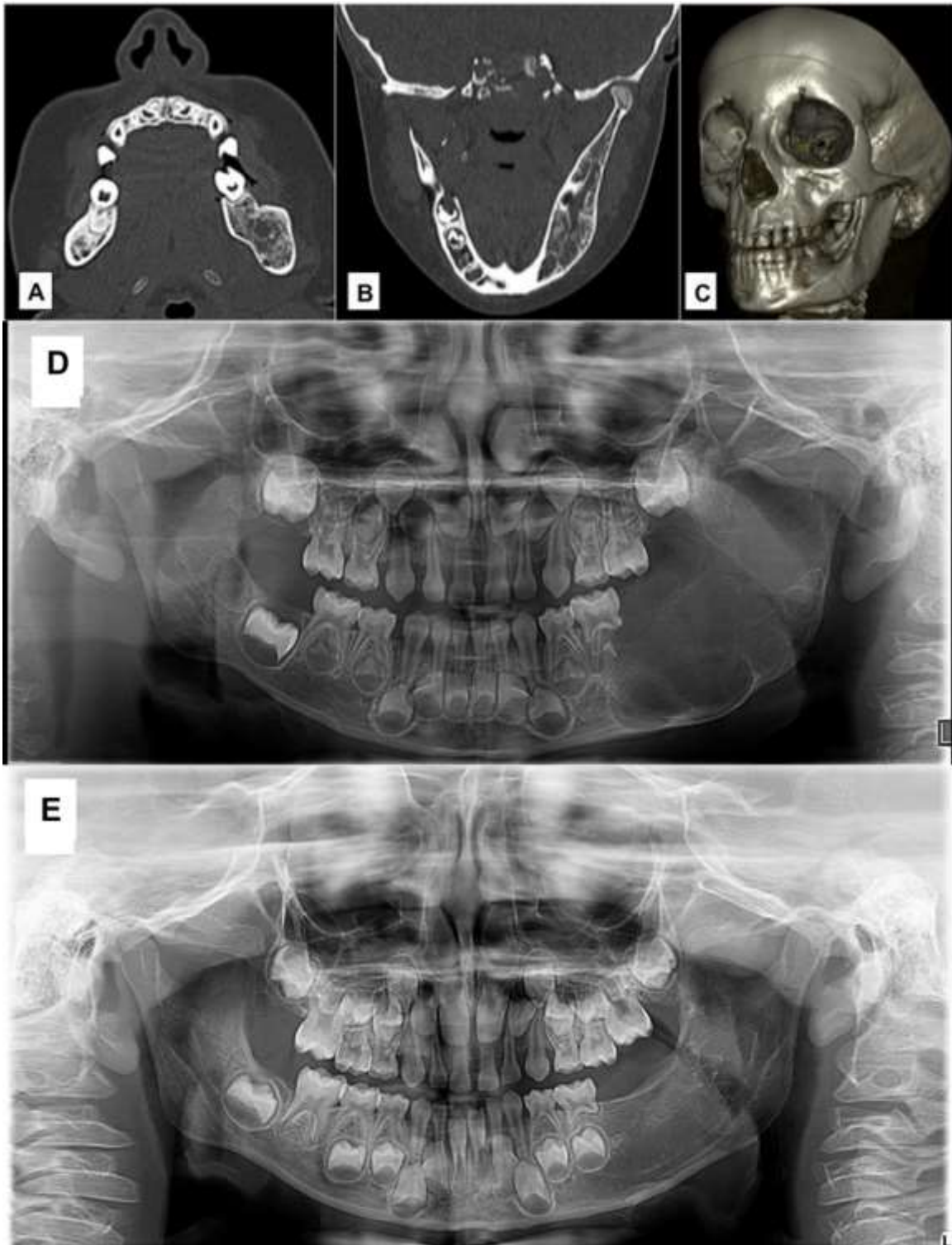


Source: Authors.

The lesion was sent to histopathological analysis, which confirmed the initial diagnosis of AFO. At 7 months postoperatively, there was evident reduction of extraoral asymmetry, with ongoing local bone repair, good intraoral healing aspect without clinical or tomographic signs suggestive of lesion relapse (Figure 4 A, B e C).

The panoramic radiograph obtained at 3-year follow-up evidenced complete bone repair at the site of lesion removal, without signs of relapse, maintenance of bone development and absence of the tooth bud of the mandibular left second molar (Figure 4 E).

Figure 4: **A, B, C:** Cone beam computed tomography at 7 months postoperatively. observe ongoing local bone repair, reduced expansion of cortical plates and absence of tomographic signs suggestive of lesion relapse. **D:** Panoramic radiograph at 30 days postoperatively evidencing radiolucent area in left mandibular body and ramus compatible with the area of lesion enucleation. **E:** Panoramic radiograph at the 3-year follow-up, evidencing complete bone repair at the lesion region, with normal bone contours, without signs of relapse and absence of teeth 36 and 37.



Source: Authors.

4. Discussion

Hamner and Pizer in 1968 (Hamner & Pizer, 1968) described that the AFO presents the same benign biological evolution as ameloblastic fibroma, yet it presents changes that led to dentin and enamel formation.

Histologically, the differential diagnosis between AFO and ameloblastic fibroma is based on the presence or absence of enamel and dentin (Santos et al., 2011; Soares et al., 2006). However, other mixed tumors should also be considered, such as calcifying cystic odontogenic tumor, adenomatoid odontogenic tumor, calcifying epithelial odontogenic tumor and complex odontoma (Pontes et al., 2008; Zouhary et al., 2008).

In the present case, the ectomesenchyme was rich in cells with presence of cells with different shapes (spherical, stellate, fusiform), which indicates significant cell activity. This finding may explain the more aggressive biological evolution observed in the present case, in agreement with the cases reported (Lucio et al., 2013; Reichart et al., 2004; Reis et al., 2007; Soares et al., 2006).

The histogenesis and classification of AFO is controversial. It has been proposed that the mixed odontogenic tumors known as ameloblastic fibroma, ameloblastic fibro-dentinoma, ameloblastic fibro-odontoma and complex odontoma represent a continuous maturation of development from one to another, respectively. Consequently, since the AFOs present the components of an ameloblastic fibroma and a complex odontoma (Boxberger et al., 2011), it has been hypothesized that the AFO is an intermediate stage as the ameloblastic fibromas evolve to a complex odontoma, which according to some authors (Dolanmaz et al., 2008) indicates that they should not be considered as distinct disorders.

Others suggested that the AFO is hamartomatous and represents a complex immature non-neoplastic odontoma (Philipsen et al., 1997; Sloomweg, 1981). In some studies, the term AFO represents a histological combination of ameloblastic fibroma and complex odontoma (Chang et al., 2002; Sloomweg, 1981). However, the concept that these lesions present continuous differentiation is not widely accepted, since other investigators suggest that they are different pathologies (Eversole et al., 1971).

Specifically concerning the ameloblastic fibrodentinoma (AFD), according to the WHO classification of 1971 it was considered a distinct disorder. In the WHO classification of 1992, it was considered a variant of AFO, while in the WHO classification of 2005 it was considered a variant of ameloblastic fibroma. In the recent WHO classification of 2017 (Wright & Vered, 2017), it was concluded that there was little evidence to justify the classification of AFD and AFO as independent disorders, and it was decided to group them as developing odontomas.

Despite the recent change in classification, the present search was based on papers published in the last 15 years with only cases of AFO, selecting the papers according to the classification into force at this period. Therefore, it was decided to exclude the cases of AFD in the selection of cases, alike (Reichart et al., 2004) suggested in their analysis of AFOs, to actually achieve parameters for comparison and discussion.

The initial search retrieved 1,392 papers, from which the study included only papers published in English language, in the last 15 years, conducted on humans, yielding 556 papers. These were analyzed by their titles and only 47 papers were considered for reading of abstracts, excluding the papers not related to this subject. After reading the 47 abstracts, papers including cases of AFO associated with other lesions, located in the maxilla or affecting patients older than 10 years were excluded. Thus, 8 papers were selected and read in full text, adding up to 10 cases (Table 1).

Table 1. Demographic and clinical features of ameloblastic fibro-odontoma (AFO) described in this paper and in the literature from 2004 to 2019.

Author, Year	Gender	Age (year)	Location	Evolution time (months)	Lesion size (cm)	Locularity	Tooth relation	Cortical bone	Treatment	Follow-up time (months)	Recurrence
<i>Present case</i>	M	3	Posterior	NR	3.8 x 2.7 x 5.4	Unilocular	Unerupted + Root resorption	Expansion	Excision + curettage + tooth removal	36	None
<i>Kalra et al, 2018</i>	F	9	Posterior	NR	NR	Unilocular	Unerupted + Displacement + lesion + root resorption + presented with mixed radiodensities and a central radio-opacity, which looked like a tooth bud	Expansion	Excision + curettage + third molar removal	12	None
<i>Abdulla et al, 2017</i>	M	6	Posterior	1	NR	Unilocular	Unerupted	NR	Excision + curettage + tooth removal	24	None

<i>Mashhadiabba s et al, 2017</i>	F	7- mont h-old	Anterior	NR	NR	Unilocular	Unerupted + Displacement	Expansion	Excision + curettage	12	None
<i>Lucio et al., 2013</i>	M	3	Posterior	3	10	Multilocul ar	Unerupted	Expansion	Excision + curettage + tooth removal	11	None
<i>Pontes et al., 2012</i>	M	9	Posterior	3	NR	Unilocular	Unerupted	NR	Excision + tooth removal	12	None
<i>Pontes et al., 2012</i>	M	6	Posterior	12	NR	Unilocular	Unerupted	Expansion + perforatio n	Excision + tooth removal	14	None
<i>Santos et al., 2011</i>	M	7	Posterior	4	NR	Unilocular	Displacement	Expansion	Excision + curettage + tooth removal	NR	NR
<i>Santos et al., 2011</i>	M	8	Anterior	6	NR	Unilocular	Displacement	Expansion	Excision	3	None
<i>Cavalcante et al., 2009</i>	M	8	Posterior	9	NR	Unilocular	Unerupted	Expansion	Excision + curettage+ tooth removal	96	None
<i>Reis et al., 2007</i>	F	6	Posterior	NR	NR	Unilocular	Unerupted	Expansion	Excision	24	None

M: Male; **F:** Female; **NR:** Not reported. Source: Authors.

The AFO is a rare benign tumor, accounting for less than 3% of all odontogenic tumors. The lesion affects mainly children (mean age 9.6 years) (Boxberger et al., 2011; Buchner et al., 2013). In the present case, the patient was 3 years old, younger than the mean age reported in the literature. When compared to cases selected from our survey, only other case reported (Lucio et al., 2013) was younger than 6 years, being a multilocular case (Lucio et al., 2013). Even though some authors do not report a clear gender predilection (H. A. R. Pontes et al., 2012; Reis et al., 2007), the male gender was more prevalent in our study, accounting for 72.72% of cases, corroborating the proportion (1.85:1,00) reported in previous studies (Boxberger et al., 2011; Buchner et al., 2013; Chrcanovic & Gomez, 2017).

According to the literature, the posterior mandibular region is usually the most affected (more than 80% of cases), and presents as radiographic characteristics being a mixed and unilocular lesion. Also, a size of 3.3 cm is considered average for the AFO, possibly varying from 0.8 to 14 cm in the greatest diameter (Boxberger et al., 2011; Buchner et al., 2013). In the present study, including the reported case, almost all cases of child patients with AFO in the mandible presented mixed lesion, located at the mandibular posterior region, except for some cases (Mashhadiabba et al., 2017; Santos et al., 2011) in the anterior region; and unilocular, except for one case radiographically presenting as multilocular.

Concerning the size, the lesion presented in this study and that reported (Lucio et al., 2013) had greatest diameters of 5.4 cm and 10 cm, respectively. Both are above the average described in the literature. The other reports do not present values corresponding to the lesion dimension (Abdulla et al., 2017; Cavalcante et al., 2009; Kalra et al., 2018; Mashhadiabba et al., 2017; H. A. R. Pontes et al., 2012; Reis et al., 2007; Santos et al., 2011)

The expansion of cortical plates is usually slow, without related painful symptomatology. Cortical perforation is uncommon and may cause significant facial asymmetry (Boxberger et al., 2011; Buchner et al., 2013). In most cases reported, alike in the present case, there was expansion of cortical plates, and only one case reported (H. A. R. Pontes et al., 2012) had cortical perforation, corroborating the literature.

In the present study, including the reported case, there have been reports of tumors that caused displacement of adjacent teeth (Kalra et al., 2018; Mashhadiabba et al., 2017; Santos et al., 2011), and other studies demonstrated lesions associated to an unerupted tooth crown (Abdulla et al., 2017; Cavalcante et al., 2009; Kalra et al., 2018; Mashhadiabba et al., 2017; H. A. R. Pontes et al., 2012), corroborating previous studies (Manor et al., 2012).

The treatment of choice for AFO is conservative combined with curettage, due to the benign evolution of the tumor, and relapse is uncommon (De Riu et al., 2010). However, there is no consensus concerning the maintenance or not of associated teeth. In some situations, surgical enucleation of AFO and maintenance of any involved teeth are indicated. Alike the present case, other authors chose to remove the involved teeth (Abdulla et al., 2017; Cavalcante et al., 2009; Kalra et al., 2018; Pontes et al., 2012). This is indicated in cases of extensive AFOs presenting teeth in regions with difficult access, since the tooth maintenance may impair the complete removal of neoplastic tissue, with persistence of tumor cells that may cause relapse (Chang et al., 2002; Lucio et al., 2013). Regardless of the tooth maintenance at the affected site, the recurrence of AFO is associated with inadequate surgical removal and occurs if the remaining tumor persists on the resection margins or involved tooth, especially in cases of large tumors (Friedrich et al., 2001). In the present case, lesion enucleation was complete, and the involved tooth was removed due to the tumor extension and bone involvement. There was risk of pathological mandibular fracture, yet the presence of basal cortical bone enhanced the prognosis. The parents were instructed about the need to avoid hard foods, local care and prevention of any type of torque or local impact. Three years after complete lesion removal, the patient did not present signs of relapse and has been followed by clinical and imaging examination.

5. Conclusion

The AFO was classified as a distinct pathology for several years and is currently considered a developing odontoma. Regardless of the classification, the treatment of choice for AFO in children should be conservative due to the benign nature and low relapse rate of the lesion. When there is a tooth related with the lesion, it may be removed or not, depending on the possibility of its maintenance in position, assuring that there would be no neoplastic remnants after complete lesion removal. A thorough diagnosis, including all clinical, tomographic and histopathological aspects, is fundamental for the adequate treatment of this pathology.

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