TREATMENT OF AMELOBLASTIC FIBRO-ODONTOMA ON A 3-YEAR-OLD CHILD: CASE REPORT

Tratamento do fibro-odontoma ameloblástico em paciente pediátrico: relato de caso

Access this article online	
Quick Response Code:	**Website:** https://periodicos.uff.br/ijosd/article/view/65647
	DOI: 10.22409/ijosd.v2i70.65647

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RESUMO

O fibro-odontoma ameloblástico (AFO) é um tumor odontogênico benigno raro com características de fibroma ameloblástico e componentes de esmalte/dentina. Este estudo relata o caso de um menino de 3 anos com edema e atraso na erupção dentária na região posterior da mandíbula. Exames de

imagem revelaram uma massa radiopaca na mandíbula esquerda com infiltração na cortical óssea. O diagnóstico foi confirmado por biópsia incisional. O tratamento incluiu ressecção marginal e curetagem, preservando a cortical lingual. Planejamento cirúrgico utilizou biomodelos 3D, e osteossíntese foi realizada com uma placa em 'L' do sistema 1.5. Em quatro anos de acompanhamento, demonstrou crescimento facial normal, aspecto cicatricial satisfatório e ausência de recidiva. Por fim, destacando a ressecção marginal conservadora como uma opção de tratamento para preservar contorno mandibular, crescimento facial, reduzir riscos de recorrência e múltiplas intervenções.

Palavras-chave: Cistos Odontogênicos; Margens de Excisão; Cirurgia Maxilofacial.

ABSTRACT

Ameloblastic fibro-odontoma (AFO) is a rare benign odontogenic tumor with characteristics of ameloblastic fibroma and enamel/dentin components. This study reports the case of a 3-year-old boy with swelling and delayed tooth eruption in the posterior mandibular region. Orthopantomography revealed a radiopaque mass in the left mandible with cortical bone infiltration. The diagnosis was confirmed through incisional biopsy. Treatment involved marginal resection and curettage, preserving the lingual cortical. Surgical planning was made using 3D biomodels. Osteosynthesis was performed using an L-shaped plate from the 1.5 system. At a four-year follow-up, the patient showed normal facial growth, satisfactory healing, and no signs of recurrence, highlighting conservative marginal resection as an effective approach to preserve mandibular contour, support facial growth, reduce recurrence risk and multiple surgical interventions, such as orthognathic surgeries.

Keywords: Odontogenic Cysts; Margins of Excision; Surgery, Oral.

INTRODUCTION

Ameloblastic fibro-odontoma (AFO) is a mixed benign odontogenic tumour, exhibiting the typical attributes of an ameloblastic fibroma while additionally comprising enamel and dentin components (WHO, 2022).

The two main complaints associated with AFO are swelling and failure of tooth eruption. Clinically, it presents as a painless swelling of the affected area, usually

the posterior portion of the maxilla or mandible. Radiographycally, it is possible to see a well-defined radiolucent area (DE RIU et al., 2010). The objective of this paper is to present a clinical case of ameloblastic fibro-odontoma on a 3-year-old child.

CASE REPORT

A 03-year-old boy was reported to oral and maxillofacial surgery service of Hospital das Clínicas de Teresópolis Constantino Ottaviano (HCTCO) with his legal representative on 2019. The legal guardian of the minor signed the Informed Consent Form (ICF), and the Research Ethics Committee of the Faculty of Dentistry at UNIFESO approved the dissemination and publication of this case report (Protocol Nº 3935955).

The legal representative referred the main complaints were tooth eruption delay and malocclusion. On inspection, a big swelling was presented on the left posterior buccal region (Fig. 1). On palpation, lesion was firm in consistency. Oral panoramic radiograph showed a radiopaque mass with a radiolucent border on the left posterior mandible, covering the mandibular body and ramus, presenting temporomandibular joint involvement (Fig.2A). Based on the 3D reconstruction (Fig.2B), a biomodel was printed for surgical planning and plate's pre-bending, in which was possible to measure the extensive cortical perforation and medullary bone destruction. On the first surgical moment, an incisional biopsy was performed in the posterior region of the mandibular ramus on 2019, which was diagnosed as AFO. The treatment choice was marginal resection and curettage, preserving the lingual cortical, followed by osteosynthesis using a 4-hole 'L'-shaped plate from the 1.5 system, considering the patient's age.

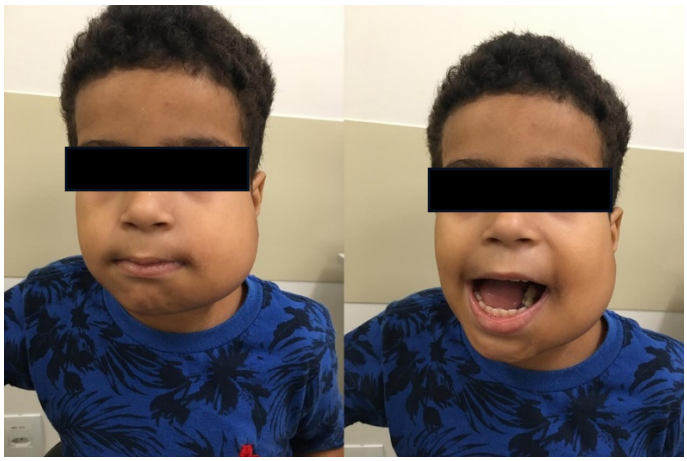

Fig. 1: patient presenting significant swelling with hemifacial enlargement on the left side.

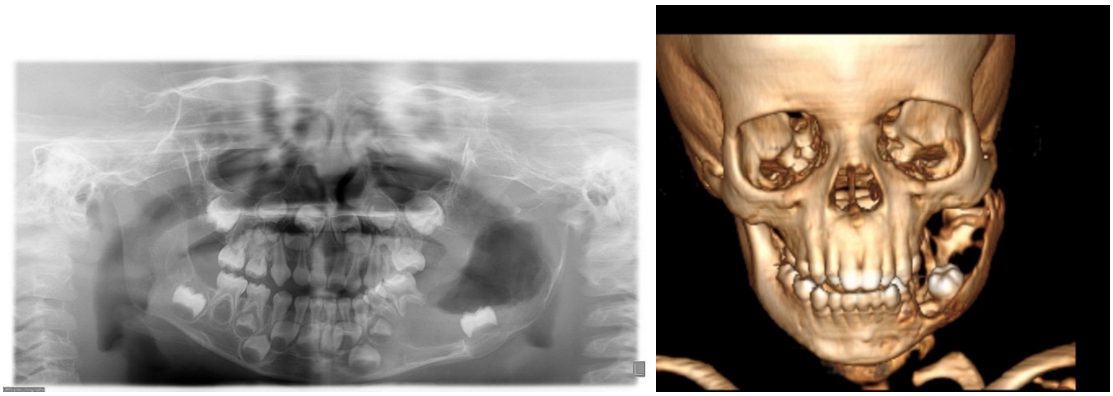

Fig. 2: A. Oral panoramic radiograph B. 3D reconstruction

The procedure was performed under general anesthesia and nasotracheal intubation, with left Kocher access for marginal resection and curettage, preserving the lingual cortical.

Using a #15 blade, a mesial incision on the skin, which divides the lower lip and extends downward to the center of the hyoid bone, added to a lateral incision bellow the margin of the mandible, extending upwards and backwards the hyoid bone, along the submandibular-cervical encounter area to 0,5cm behind and bellow the jaws's angle, up to the level of the mastoid process's apex (Fig. 4).

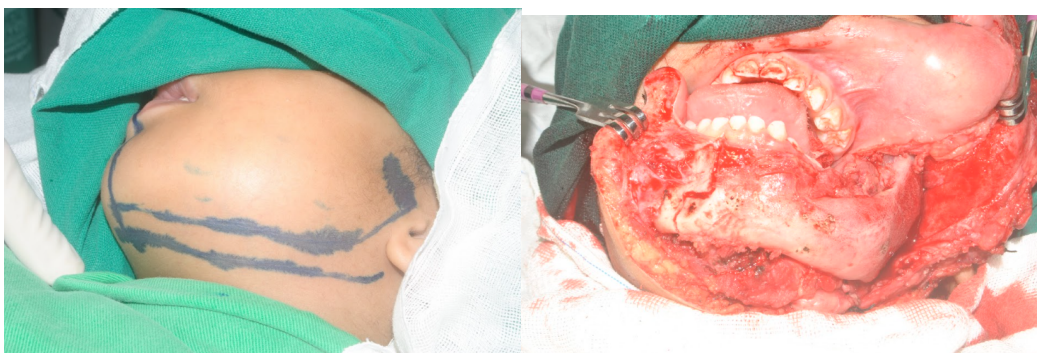

Fig. 4: A. Kocher approach delimitation. B. Tumor exposure

The dissection proceeds by incorporating muscular structures. Anteriorly, mentalis muscle, depressor labii inferioris muscle, and depressor anguli oris muscle. Posteriorly, the buccinator and masseter muscles. On the lingual mandible surface, the anterior digastric portion, mylohyoid, geniohyoid, and genioglossus muscles were detached with an electric scalp, followed by the medial pterygoid muscle.

Following this, the periosteum is dissected on the vestibular and lingual surface. Subsequently, the anterior portion of the mandible is divided medially between elements 81 and 71 using a surgical microsaw system.

The intraosseous lesion was exposed (fig. 4B). There after, marginal resection of the tumor was performed including a safety margin of 1,0cm and curettage, preserving the lingual cortical, along with the extraction of the involved teeth. Internal fixation was made with two screws on each segment, using a four-hole L-shaped plate from the 2.0 system, considering the patient's age and height. A 3D biomodel of the mandible was printed allowing for pre-bending of the plate prior to the operation, thus reducing operation duration.

The extrinsic muscles of the tongue inserted on the inferior border of the mandible were sutured with Vicryl 3.0. Access closure was performed according to the anatomical layers, including the fascia and platysma, subcutaneous tissue, and skin with simple sutures, on the deep planes using Vicryl 3.0, and Nylon 5.0 on the skin.

The patient was discharged on day 02 post-operation and continued an oral liquid diet. On the first post-operative examination, the dentoalveolar mucosa appeared normally colored and demonstrated satisfactory healing, with slight swelling on the left submandibular region. At one year follow-up, no limitation on mouth opening was observed (Fig. 5). Thus, after six months of bone repair, plate removal was performed.

Fig. 5: One year follow-up pictures. A) front view with open mouth; B) Left profile view; C) inferior-superior view.

At a two-year follow-up examination, a slight laterognatia towards the left side was recognizable. Jaw movements and occlusion were normal, with no speech impairment. At the present moment, the patient is under control, with no signs of tumor recurrence. Patient was referred to receive orthodontic and pediatric dental treatment to ensure good jaw and facial growth and symmetry, still remaining on ambulatory care. Control orthopantomography demonstrated satisfactory bone callus formation.

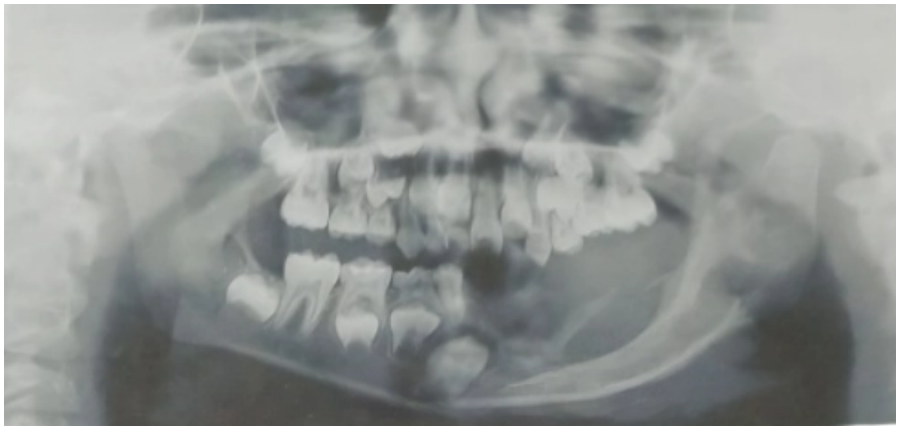

Fig. 6: Four-year control orthopantomography

DISCUSSION

The 4th edition of WHO classification of head and neck tumors indicates that AFO are not considered independent lesions, but a developing odontoma (EL-NAGGAR et al., 2017). However, there is still confusion regarding the nature of these lesions. The concept of ameloblastic fibro-odontoma being a maturation stage of the odontoma is not universally accepted (CHRCANOVIC & GOMEZ, 2017).

Given the difficulty in distinguishing a true tumor from a developing odontoma, the true nature of the lesion cannot be easily determined (DE RIU et al., 2010; CHRCANOVIC & GOMEZ, 2017). Nevertheless, some researchers acknowledge that AFO can reach extensive sizes and exhibit characteristics of true neoplasms (CHRCANOVIC & GOMEZ, 2017).

Several factors determine the treatment modality choice. The continuous growth and facial bone physiology in children characterized by a higher percentage of cancellous bone, bone turn-over, and periosteal activity affect the management approach considerably (KUMAR et al., 2014). Despite the general agreement for the use of conservative therapy in young patients, the AFO necessitates a long follow-up due to its controversial nature, and this must be considered when choosing the treatment modality, according to patient's profile and limitations (DE RIU et al., 2010; KUMAR et al., 2014).

The choice of combining radical and conservative therapy as an initial approach eliminates the need for multiple surgical interventions in recurrent lesions, thereby reducing morbidity and multiple hospitalization. Additionally, maintaining the mandibular contour ensures satisfactory facial growth and minimizes the likelihood of orthognathic surgeries to correct facial asymmetries (TROSMAN & KRAKOVITZ, 2015; CASTELLON et al., 2018).

Authors have reported cases of malignancy from ameloblastic fibro-odontoma to ameloblastic fibrosarcoma (HOWELL & BURKES, 1977; FRIEDRICH et al., 2001). Ameloblastic fibrosarcomas arise from ameloblastic fibromas or ameloblastic fibro-odontomas about 50% of the time (NEVILLE et al., 2009; CASTELLON et al., 2018)

CONCLUSION

A case of AFO is rare due to underreported number of cases in literature. This article presents a case report of AFO with emphasis on the treatment of this lesion. The modality of choice in this presented case involved a kocher approach for marginal resection, curettage and lingual cortical preservation as a satisfactory treatment option.

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