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HAMARTOMATOUS GROWTH IN A CHILD, AMELOBLASTIC FIBRO-ODONTOME: A CASE REPORT

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ABSTRACT

Introduction: Ameloblastic fibro-odontoma is a rare, mixed, benign, odontogenic tumor with significant prevalence in the mandible. It is characterized by histologic features of ameloblastic fibroma with the formation of enamel and dentine. It usually affects pediatric patients and is associated with teeth, leading to delay in eruption or a deviation in the dental eruption pathway. **Case Report:** This is a case report of Ameloblastic fibro-odontoma and its management in a 11 year old boy. Intra-oral examination revealed an edema in the buccal area of the mandible's posterior right region. Radiographic examination showed an expansile, mixed radiolucent-radiopaque lesion with well-defined corticated border. Surgical enucleation and curettage was performed under general anesthesia. Histopathologic findings led to diagnosis of Ameloblastic fibro-odontoma. **Conclusion:** Although Ameloblastic fibro-odontoma is a rare tumor, its prevalence is more in children's jaw. Conservative surgical treatment allowed the normal development with no sign of recurrence observed during the 6-month follow-up period.

INTRODUCTION

According to World Health Organization (WHO), Ameloblastic fibro-odontoma (AFO) is defined as a tumor comprising of proliferating odontogenic epithelium embedded in cellular ectomesenchymal tissue that mirrors dental papilla, with varying degrees of inductive change and dental hard tissue formation.[1] Hooker coined the term AFO which had originally been termed ameloblastic odontoma.[2] Prevalence of AFO is relatively rare, it is about 3.1% of all odontogenic tumors[3], with occurence predominantly in children and teenagers, no predilection for gender.[4] There is frequent association with erupted or displaced teeth which can reach large sizes.[5] Radiographic examination of AFO consists of a well-defined unilocular or multilocular radiolucent lesion that carries part of irregular radiopaque particles, usually in the posterior mandible.[6,7]

AFO is managed conservatively which consists of enucleation of the lesion and curettage of the adjacent bone. The prognosis of the lesion is excellent with low rate of recurrence.[8] The purpose of this article is to describe a case of AFO that affected the left mandibular region of a patient that was treated with surgical approach without any sign of alteration or recurrence after 12 months of follow-up.

Case Report

The parent of an 11 year boy was referred to the Department of Pediatric and Preventive Dentistry with complaint of swelling on left side of the jaw. The patient's past medical history was unremarkable. Extra-oral examination showed a painlful swelling on the left lower border of mandible, with firm consistency on the palpation, which caused little facial asymmetry. Intra-oral examination edema was noted in the buccal area of the mandible's posterior right region. The permanent mandibular right first molar was absent, and the alveolar mucosa was irregular with hard tissue projection resembling bone. (Figure: 1a) The panoramic radiograph revealed an expansile, mixed radiolucent-radiopaque lesion with well-defined corticated border. There was also an unerupted presumably first permanent molar in the mandible's basilar region. (Figure: 1b)

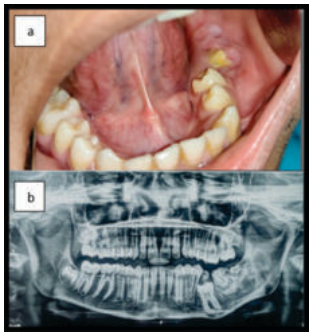

Figure 1a,b: Intraoral Photograph and Pre-operative Orthopantomogram

Cone beam computed tomography revealed presence of an unilocular lesion in left mandibular ramus region measuring 15.3mm antero-posteriorly, 28.0mm supero-inferiorly and 10.9mm mesioditally in its largest dimension extending from angle region inferiorly to the subcondylar region superiorly. (Figure: 2) A well-defined radiodensity, morphologically resembling multiple tooth which is embedded in the lower left third molar region of mandible suggestive of an odontome. The radiographic diagnosis was made of complex odontome associated with odontogenic cyst.

Under general anesthesia, complete removal and curettage of the lesion were performed with great care due to the risk of mandible fracture. The specimen was sent to histopathological analysis, with the diagnosis of an ameloblastic fibro-odontoma (Figure: 3). During the 6 month follow up period no sign of recurrence was detected and soft tissue healing was uneventful. The patient is being followed up postoperatively for the eruption of first permanent molar, if not then orthodontic extrusion will be planned.Figure 2: CBCT showing cyst and presence of odontome

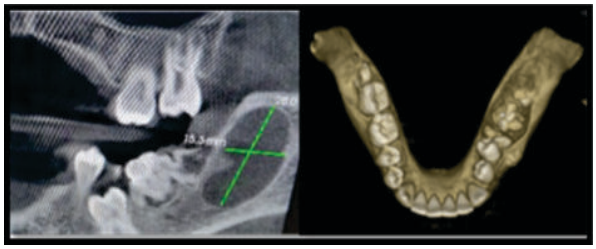

Figure 2: CBCT showing cyst and presence of odontome

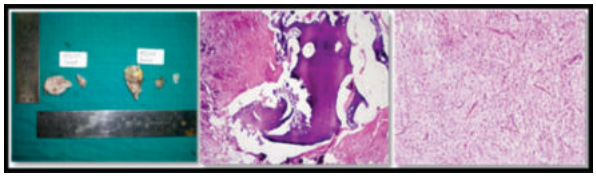

Figure 3: Excised Gross tissue specimen with Photomicrograph of hard tissue and soft tissue(10x)

DISCUSSION

AFO is a benign tumor comprising of odontogenic epithelium proliferation embedded in an ectomesenchymal tissue with dentin and enamel formation as a result of inductive properties of this tissue.[9] In a study, Buchner reviewed published cases series of totally 114 patients it was revealed that the mandible-to-maxilla ratio was 1.85:1 and almost 80% of the lesions were located in the posterior region of the jaws.[10] AFO is usually associated with teeth, leading to delayed eruption and changes in the eruption pathway.[5] The progression of tumour5 mass is slow and generally produces an asymptomatic increase in volume.[7] When large, however, it may cause discomfort. Tooth mobility may also be present.[11]The present case had unerupted teeth in the region of the growing tumour mass, but with symptoms, with no tooth mobility.

Radiographic diagnosis generally reveals a well-defined radiolucent area composed of various amounts of radiopaque material of irregular size and form.[7] For the evaluation of the nature and behaviour of lesions, age of the patient and size tumor are the most important factors that must be considered at initial detection phase. In our case, histopathologic assessment showed typical specification of both ectomesenchymal and epithelial components which led to diagnosis of AFO. Management of AFO is with conservative surgical approach.[12] Recurrence of AFO is rare, most of the recurrences are related to incomplete surgical removal.[13]In a case conducted by *Reis et al*. AFO was treated with enucleation and impacted lower left first permanent molar was preserved and complete eruption was reported without any sign of recurrence. Some clinical reports have achieved success without dental management followed by tooth eruption with no sign of recurrence.[14]

However there are certain reports of sarcoma degeneration, *Ueki et al.* reported a case of malignant ameloblastic fibrosarcoma in a dog, with metastasis that led to the death of the animal.[5] *Sozeri et al.* reported a case of an ameloblastic fibrosarcoma in the mandible of a 5-year-old child, inspite of repeated surgical interventions, the tumor recurred 3 times within a year and a half, but without metastases. The treatment of choice in such cases is radical surgery.[15]

In the present case, lesion enucleation was complete, and the impacted tooth was preserved. The parents were instructed about the need to avoid hard foods, local care and prevention of any type of torque or local impact. Six months after complete lesion removal, the patient did not present signs of relapse and has been followed by clinical and imaging examination.

CONCLUSION

Ameloblastic fibro-odontoma exhibits clinical and radiographic features that may not be confirmatory, and its diagnosis is only be confirmed by histological examination. It remains a disputed tumor, since some authors claim that ameloblastic fibroma and complex odontoma are different stages of the same pathology, whereas others believe that these lesions are different pathologies. The main principal is that, whatever the classification, the treatment must be conservative in children, an 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.

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