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Case Report

Ameloblastic Fibro-Odontoma: A Rare Case Report

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Abstract: Ameloblastic fibro-odontoma (AFO) is one of the rare mixed radiolucent and radiopaque odontogenic tumors in children and are often asymptomatic. AFOs are found by routine clinical and radiological examination or when they cause obvious intra- or extra-oral swelling. We rport an unusual case of Ameloblastic fibro-odontoma of 06-year old male patient.

Keywords: Benign tumors, Mixed odontogenic tumors.

INTRODUCTION

Ameloblastic fibro-odontoma is a rare benign mixed odontogenic tumor, comprising 1-3% of all odontogenic tumors [1]. The term ameloblastic fibroodontoma was first purposed by Hooker in 1967 [2]. Ameloblastic fibro-odontoma is defined by the WHO as a neoplasm composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles dental papilla; there are varying degrees of inductive changes, resulting in dental hard tissue formation [3]. It may inhibit tooth eruption or displace involved teeth, although teeth in the affected area remains vital. The lesions are usually diagnosed during the first and second decade of life. It occurs with equal frequency in the maxilla and the mandible and with equal frequency in males and females. Radiographically, it shows a well defined, radiolucent area containing various amounts of radiopaque material of irregular size and form [4]. The AFO shares similarities with the ameloblastic fibroma and the ameloblastic fibro-dentinoma but may be distinguished from these two entities by the presence of dentin and enamel on histological examination [5, 6]. Here, we present a case of ameloblastic fibroodontoma affecting the maxilla in a 06-year-old male children.

CASE REPORT

06 -year-old male patient referred to department of oral & maxillofacial surgery for the treatment of swelling on left side of face which was present since 5-months. His medical, family, and social history were unremarkable and there was no history of trauma. On oral examination revealed a large lobulated lesion extending from the upper lateral incisor to the first molar region [Figure - 1]. The swelling measured about 12 × 7 cm; there was obliteration of the labial and buccal vestibule, the lesion was reddish, nontender, and soft to firm in consistency. A panoramic radiograph [Figure - 2] revealed a large expansile radiolucent lesion, with the teeth pushed towards the periphery of the lesion. OPG & radiolucent mass with a radiopaque boarder in the left posterior maxilla covering left maxillary tuberosity with scatter foci & radiopaque material.



Fig-1: Preoperative view



Fig-2: OPG showing, On left side of maxilla, radiolucent lesion with scattered radio-opaque material

On the basis of the clinical and radiographical finding, the possible differential diagnosis was ameloblastic fibro-odontoma, calcifying epithelial odontogenic tumour, adenomatoid odontogenic tumour immature complex odontoma. Under (AOT) and general anaesthesia with oral intubation, a crevicular incision was made from the left maxillary primary lateral incisor region to the maxillary tuberosity with a vertically releasing incision from the mesial end of right primary lateral incisor. A full thikness elevated and a thin bone covering the surface of the lesion was removed. The lateral wall of the maxillary sinus was removed and the tumor was identified from the maxillary sinus the lesion was enucleated and the defect was not filled with any material [Figure-3]. The cavity was irrigated carefully and the debris was removed. The flap was repositioned in the same position and suturing was done with vicryl 3.0 suture material, avoiding the risk of an oro-antral communication[Figure- 4].



Fig-3: Intraoperative view



Fig-4: After Suturing

Microscopic examination of the surgical specimen showed islands of odontogenic epithelium, with peripheral columnar cells resembling ameloblasts and central stellate reticulum-like cells. The mesenchymal component was an embryonic fibrous connective tissue with delicate fibrils resembling dental papilla. Calcified structures resembling dysplastic dentin with enamel space were seen in close proximity to the epithelium. There was no evidence of a malignant growth were observed. Based on the histopathological findings, diagnosis of ameloblastic fibro-odontoma was made [Figure- 5].

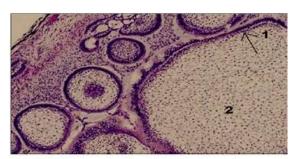


Fig-5: Histopathological view

DISCUSSION

Ameloblastic fibro-odontoma is one such example, involving both the epithelial and mesenchymal elements, with inductive changes [7]. This tumor has been confused earlier with other odontogenic mixed tumors [8]. It was identified by a variety of terms until Hooker distinguished it from ameloblastic odontoma and described it as a separate entity [9]. It is a slow growing lesion and it occur before 20 years of age [10].

The two most common presenting complaints are swelling and failure of tooth eruption. The lesion may displace erupted teeth although the teeth remain vital.

One area of discussion is the nature of AFO with regard to the discrimination between neoplasia and hamartoma [11]. Both Philipsen *et al.* and Slootweg indicated that the AFO has a hamartomatous character,

whereas the ameloblastic fibroma has a neoplastic nature [12, 13].

Histologically, AFO is characterized by proliferation of ectomesenchymal component, which resembles dental papilla, exhibiting stellate and spindle-shaped cells, and an odontogenic epithelial component [14, 15]. Rarely, tumors with the histomorphology of AF may form dysplastic dentin, and are called ameloblastic fibrodentinomas or dentin plus enamel and be classified as ameloblastic fibro-odontoma. Presence of different amounts of mineralized products of odontogenesis (dentin and enamel matrix) is the key to distinguish AFO from AF. The islands and strands of odontogenic epithelium are often in close relation with the hard tissue.

Treatment of odontogenic tumors is based on the biological and clinical behavior. Many authors have reported that AFO is not aggressive and can be treated adequately with surgical curettage of the lesion without removing the adjacent teeth [16, 17]. Chen et al. studied seven cases of AFO: five patients were initially treated by enucleation or curettage; one by segmental resection; and one by hemi-mandiblectomy. There was recurrence in two of the five patients with follow-up data [18]. Some authors suggested that in case of association with unerupted or even erupted teeth, the teeth should be removed in order to avoid recurrence, especially when dealing with larger AFOs [19]. However, this topic is controversial, since some authors believe that teeth do not interfere with tumor enucleation; therefore, there is no reason for removal, with possibility of further spontaneous eruption.

Here, we recommend tumor enucleation as first step in the treatment & the associated tooth bud has to be removed in order to avoid recurrence but more extensive treatment or resection only in the cases of repeated recurrence or evident malignancy.

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