International Journal of Dental Sciences
www.dentaljournal.in
E-ISSN: 2663-4708, P-ISSN: 2663-4694
Received: 04-08-2024, Accepted: 03-09-2024, Published: 18-09-2024
Volume 6, Issue 2, 2024, Page No. 27-30

Ameloblastic fibro-odontoma

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Abstract

Introduction: Ameloblastic fibro-odontoma (AFO) is a relatively uncommon mixed odontogenic tumor usually detected in the early years of life. Common signs and symptoms include asymptomatic swelling, delayed tooth eruption, and a radiographically distinct lesion that appears radiolucent-opaque with well-defined borders. The standard treatment for AFO involves enucleation of the tumor, accompanied by long-term follow-up to monitor for potential recurrence or complications.

Patient case summary: A 10-year-old girl was referred to the Oral Medicine and Radiology department due to a small swelling in the lower right back teeth area, present for 5 to 6 months. A CBCT scan revealed a large, well-defined, cauliflower-like radiopaque lesion in the right ramus region. The lesion was enucleated under general anesthesia, and histopathological analysis showed focal islands of odontogenic epithelium with low cuboidal to columnar cells, along with ameloblastomatous proliferation, calcifications, and dentinoid tissue.

Discussion: Mixed odontogenic tumors are a group of rare lesions that can lead clinicians to consider a variety of differential diagnoses; therefore, it is essential to conduct thorough clinical and radiological investigations, perform proper surgical excision, achieve an accurate histopathological diagnosis, and ensure long-term follow-up in order to develop and implement an appropriate treatment plan for the patient.

Conclusion*:* This case report highlights the importance of appropriate clinical, radiographical and histological correlation for the correct diagnosis and treatment of ameloblastic-fibroodontoma. The possibility of a mixed rare tumour should be kept in mind by the clinician where they deal with the swellings of posterior mandibular region in children.

Keywords: Ameloblasticfibro-odontoma, Odontome, mixed odontogenic tumor

Introduction

The ameloblastic fibro-odontoma (AFO) is an uncommon benign odontogenic lesion characterized as a tumor that shares the general attributes of an ameloblastic fibroma but also incorporates enamel and dentine(1,2) In the recent 2022 World Health Organization (WHO) classification of Odontogenic Tumors, ameloblastic fibro-odontoma (AFO) and ameloblastic fibrodentinoma (AFD) were excluded, as was the case in the 2017 classification. This exclusion is based on the presumption that most cases of AFO and AFD likely represent developing odontomas [11]. Typically, AFO is diagnosed in young patients and does not show a significant predilection for either gender. The two primary clinical complaints associated with ameloblastic fibro-odontoma are swelling and the failure of tooth eruption. Clinically, this condition presents as a painless swelling in the affected area, which is usually located in the posterior region of the maxilla or mandible. Radiographically, AFO is characterized by a welldefined radiolucent area that contains varying amounts of radiopaque material, which are irregular in size and shape [1, 2, 3, 5].

Case report

A 10-year-old girl was referred to Oral medicine and Radiology department with a complaint of swelling on the right side of face since 5-6 months. On extraoral examination well defined swelling present on right side of the face extending from the from of the right medial canthus of the eye -to the tragus of the ear and angle of mandible. (Fig 1). Supero-inferiorly from the zygomatic region to the lower border of the mandible. On palpation swelling was firm to hard, non fluctuant, non compressible, and tender on palpation.

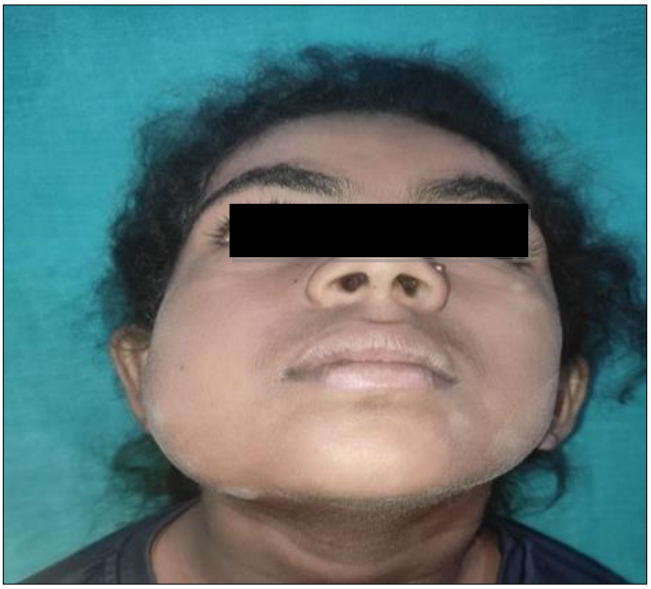

Fig 1: Extra-oral photograph showing a swelling extending from right corner of mouth to the angle of mandible and tragus of ear

Radiographical investigation

Cone beam computed tomography radiograph showed a large well defined cauliflower like radiopaque lesion in right body and ramus region.

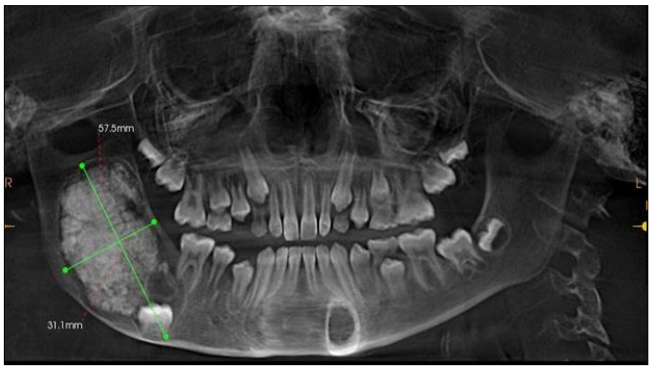

Fig 2: Reconstructed OPG View of CBCT Showing The lesion is extending from supero-inferiorly distal of 46 to 6mm short of the sigmoid notch, with a dimension of approx. 57.5mm and AP from medial border of anterior 1/3rd of ramus to the 2 cm from angle of mandible with dimension approx. 31.1mm

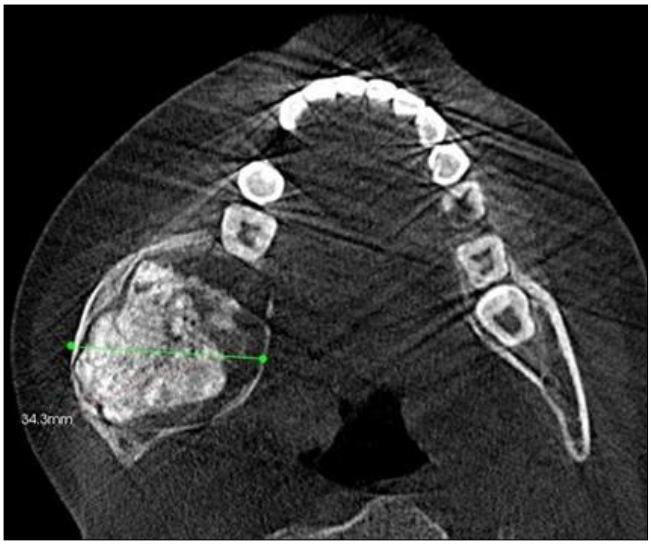

Fig 3: Axial View of CBCT Showing Buccolingually, showed an expansion of 34.3mm.

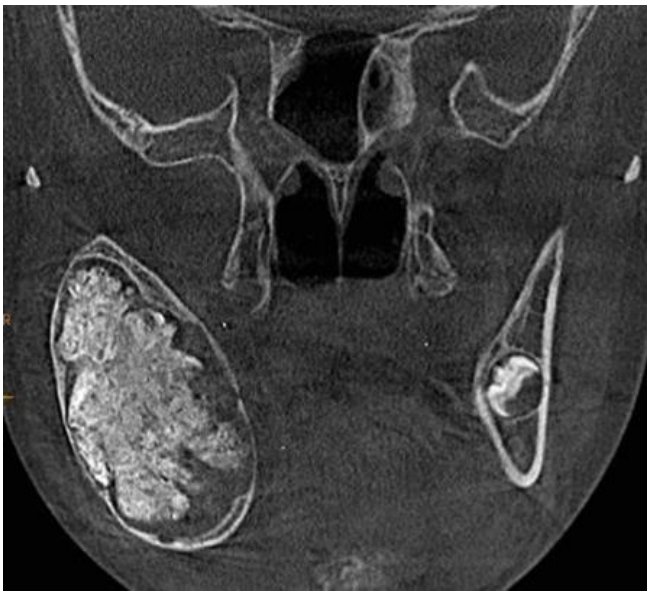

Fig 4: Coronal View of CBCT Showing Cauliflower like growth of lesion

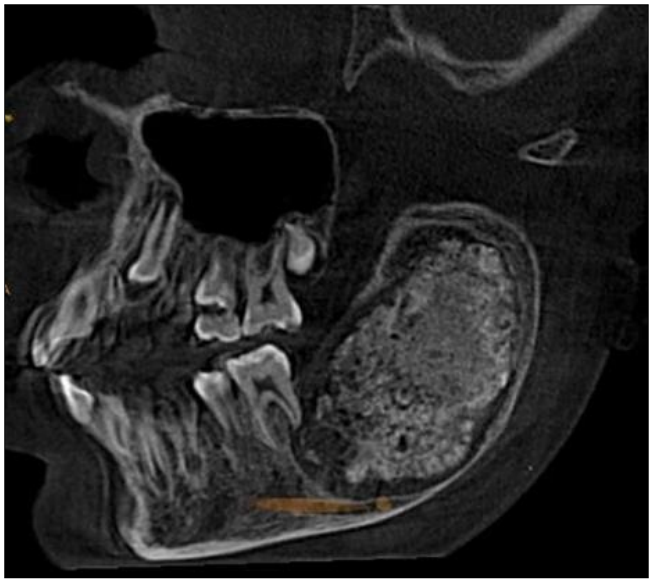

Fig 5: Sagittal View of CBCT showing Inferior displacement of the IANC, Tooth bud of Mandibular 2nd Molar and Apical root resorption of the distal root of Mandibular 1st Molar.

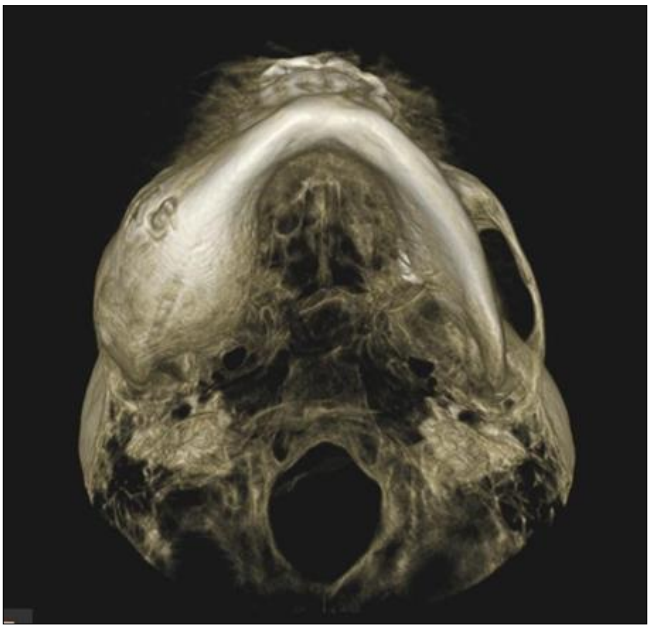

Fig 6: Histopathology (10×) decalcified section showing areas of enamel and dentine in close relationship with the ameloblastic epithelium CBCT(3D reconstruction) demonstrating massive expansion of the buccal and lingual cortex

Radiolucent peripheral rim surrounding radiopaque cauliflower growth noted. The internal structure is radiopaque, the original osseous structure is completely lost. Lesion has caused expansion of the buccal and lingual cortex. Apical root resorption of the distal root of right mandibular first molar was seen. Considering the clinical and radiological picture the possible differential diagnosis was Complex odontoma, Adenomatoid Odontogenic tumor (AOT), Calcifying odontogenic cyst (COC), Ameloblastic fibro-odontoma, Calcifying epithelial odontogenic tumour (CEOT).

Histopathology

On gross examination the specimen consisted of a hard tissue mass with a soft tissue attachment (Fig 9). Histopathologic examination of the soft tissue revealed focal

islands of odontogenic epithelium, characterized by low cuboidal to columnar cells with ameloblastomatous proliferation (Fig. 8). Islands of central stellate reticulum-like cells, with associated dentin, cementum, and pulpal tissue in a loose stroma were seen. Additionally, calcifications and dentinoid tissue were observed (Fig. 7).

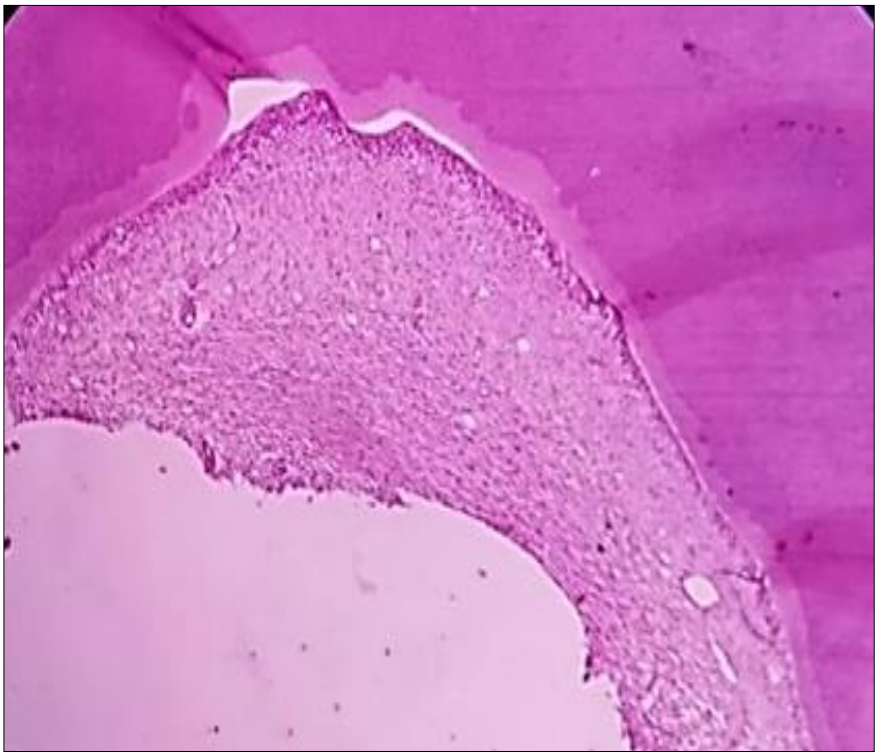

Fig 7: Histopathology (10×) decalcified section showing areas of enamel and dentine in close relationship with the ameloblastic epithelium

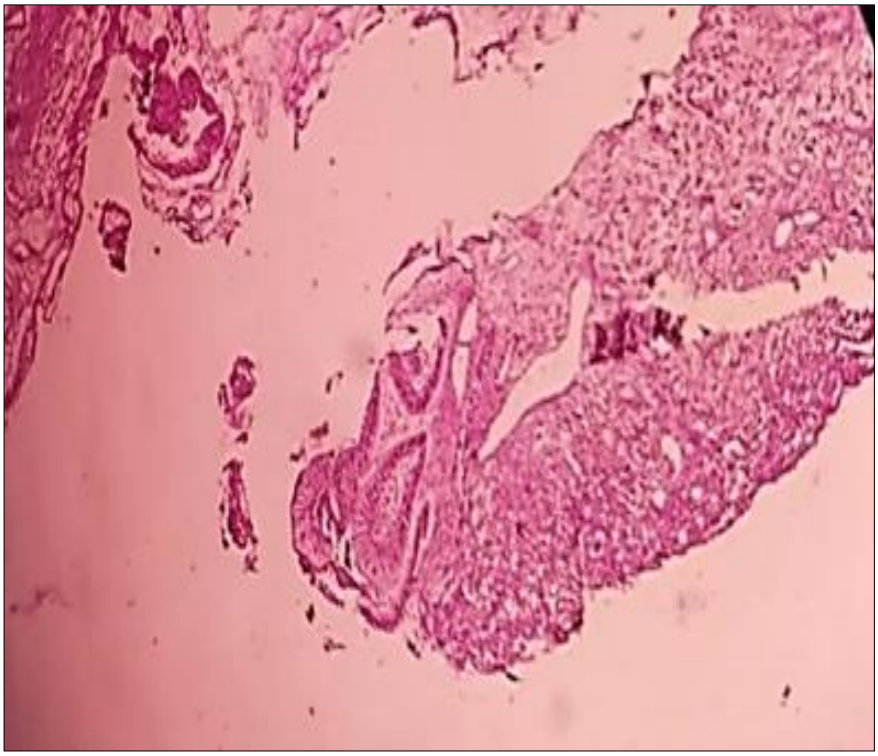

Fig 8: Histopathology (10×) the histopathologic examination of the soft tissue revealed focal islands of odontogenic epithelium–low cuboidal to columnar cells- ameloblastomatous proliferation.

Discussion

Ameloblastic fibro-odontoma (AFO) is a rare type of radiolucent-opaque odontogenic tumor [1, 3]. According to a recent comprehensive study, AFO has been reported with a prevalence range of 0– 3.4%. Typically, it is observed in individuals in their first and second decades of life, which might be a characteristic of the lesion [1, 4, 5].

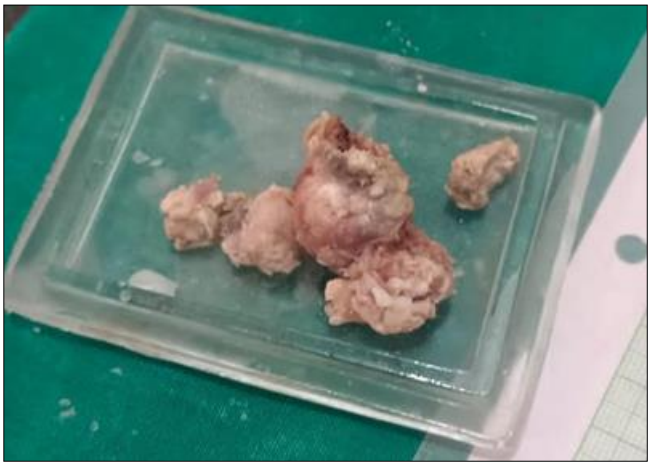

Fig 9: Excised lesion

However, ameloblastic fibro-odontoma can also occur in older adults [6, 7]. Previous research by Philipsen *et al*. indicated that the mean age of AFO patients is lower compared to those with ameloblastic fibroma (AF) and ameloblastic fibro-dentinoma. This finding supports the idea that age is a critical factor in diagnosing AFO [7]. Common signs and symptoms of AFO include asymptomatic swelling, delayed tooth eruption in the affected area, and radiolographically well-defined radiolucent-opaque appearance. This appearance is similar to other odontogenic formations, such as complex odontoma, calcifying epithelial odontogenic tumor, and adenomatoid odontogenic tumor. However, the final diagnosis is determined through microscopic evaluation, which reveals islands of odontogenic epithelium embedded in cellrich ectomesenchyme, similar to dental papilla. AFO can be differentiated from ameloblastic fibroma radiologically by multilocular radiolucency with sclerotic borders appearance and histological features characterized by epithelial islands and cords submerged in ectomesenchyme that bear resemblance to the dental papilla and enamel organ but without actual hard tissue formation (12 It is also distinguishable from ameloblastic fibro-dentinoma, not only due to the presence of dental structures resembling dentine, but also because it contains enamel-like tissues. Therefore, the formation of AFO may be attributed to enamel matrix production, one of the most important features of this lesion [1].

The treatment of ameloblastic fibro-odontoma typically involves a conservative surgical approach. Infrequent recurrences of ameloblastic fibro-odontoma have been linked to insufficient surgical removal during the initial treatment. Some studies suggest that conservative enucleation is sufficient. There is ongoing debate in the literature regarding whether to extract or retain the associated tooth bud in cases of AFO. Most articles advocate for the removal of the associated tooth bud to prevent recurrence [13, 14].

Conclusion

Clinicians should consider the possibility of a rare radiopaque-lucent odontogenic tumor when encountering a radiopaque mass in the posterior mandible of children. While most of these benign lesions heal well with complete excision, long-term follow-up is necessary to ensure proper recovery and monitor for any potential recurrence.

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