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Adenomatoid Odontogenic Tumor of Mandible - A Case Report of Unusual Manifestation.

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Abstract

the jaws and more frequently, the anterior maxilla usually in association with the crowns of unerupted teeth. Most of the tumors are diagnosed in second decade of life. A rare case report of AOT associated with an impacted left mandibular canine a 17-year-old female is reported.

Keywords: Adenomatoid odontogenic tumor, follicular, impacted, mandible, canine

Introduction

Odontogenic Tumors are a group of heterogenous uncommon lesions that affect maxillary and mandibular osseous tissue, due they originate from mesenchymal and/or epithelial components from the odontogenesis. OT are lesions that present a wide variety of clinical and his to pathological features, developing from hama r to mas to malignant lesions. [1]

It was first introduced by Dreilbaldt in 1907, as a pseudo adenoamelo blastoma, and first reported by Harbitz in 1915. In 1971, the World Health Organization (WHO) adopted the term 'adenomatoid odontogenic tumor' (AOT). Max and Stern, in 2003, coined the name 'adeno matoid Odonto genic cyst'. [2] The WHO defines it as a tumor composed of odontogenic epithelium in a variety of his to architectural patterns, embedded in a mature connective tissue stroma and characterized by slow but progressive growth. [3]

The AOT is the fourth more common Odon to genic tumor, appearing in 2-7% of cases. It arises from Odonto genic epithelium, which indicates it could originate from the enamel organ, reduced enamel epithelium, Malassez

rests or dental lamina rests, being this the most accepted theory. AOT is a benign, painless, non-invasive, and slow-growing tumor that does not infiltrate the bone. [4]

Sometimes referred to as 'two-third tumor' because it occurs in the maxilla in about two-third cases, about two-third cases arise in young females, two-third cases are associated with an unerupted tooth, and two-third affected teeth are canines.[4]

AOT is a well-defined unilocular lesion surrounding the crown of an unerupted tooth, often misdiagnosed as Odonto genic cysts, such as dentigerous cysts and amelo blast omas. Diagnosis of AOT usually depends on pathological features. Due to tumor growth, the adjacent teeth can be displaced, but teeth root resorptions are rare. There are three variants of AOT: Follicular, extra follicular, and peripheral. The follicular type (peri coronal) is a central intra bony lesion associated with an unerupted tooth, which accounts for about 70% of all cases. The extra follicular type (extra coronal) is also an intra osseous lesion, but unrelated to an unerupted tooth, and represents 25% of all AOTs. The peripheral type (extra osseous) is a rare form that arises in the gingival tissue, and accounts for 5% of all AOTs. [5] Conservative treatment is indicated for AOT because of its low recurrence.[4]

The aim of the present article is to report an Adenomatoid Odontogenic Tumor case, and describes the surgical therapy, clinical course and morphological characteristics of an adenomatoid odontogenic tumour that developed in the mandible of a 17-year-old patient.

Case report

A 17-year-old female patient reported to our Department of Oral and Maxillofacial Surgery at Dr. R Ahmed Dental College and Hospital with chief complaint of swelling in the lower front teeth region since 4-5 years. The swelling increased gradually in size. She had no other chief complaint symptoms, such as pain or numb ness of the lower lip. She had no noteworthy family history of this condition.

Extra orally, there was gross facial asymmetry due to a solitary, diffused nodular swelling of about 3x2 cm on the symphyseal region of the mandible. On palpation of the affected area, the swelling was firm to hard in consistency, fixed to the underlying bone, free from the overlying skin. Intra orally, a solitary swelling was situated in the anterior region of mandible measuring about 3x2 cm labially and 2x2 cm lingually in its greatest dimension, extending from 34 to 42, obliterating the vestibule. The margins were diffuse and the surface mucosa was normal. On palpation, swelling was non-tender, hard in consistency towards the gingival margin and firm towards the buccal vestibule with grade III mobility deciduous canine lateral incisor and permanent central incisor with missing permanent 32, 33. There was expansion of the buccal and lingual cortical plate. The deciduous lateral incisor and canine was over-retained and had no signs of eruption of permanent lateral incisor and canine.

A B C

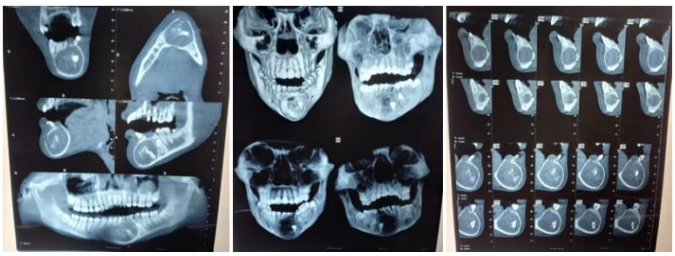
Fig 1: (A) Gross facial asymmetry due to a solitary, diffused nodular swelling of about 3x2 cm on the symphyseal region of the mandible, (B) Intraorally, a solitary swelling was situated in the anterior region of mandible measuring about 3x2 cm labially and (C) 2x2 cm lingually in its greatest dimension, extending from 34 to 42, obliterating the vestibule.

Investigations

CBCT was performed to locate the missing permanent mandibular lateral incisor and canine and to know the nature and extension of the lesion.

CBCT revealed a large unilocular radiolucency measuring about 4x5 cm extending from mesial surface of 34 to mesial surface of 43.

It was oval in shape, with well circumscribed and corticated margins.



D E F

Fig 2: (D), (E), (F). CBCT revealed a large unilocular radiolucency measuring about 4x5 cm extending from mesial surface of 34 to mesial surface of 43.

It was oval in shape, with well circumscribed and corticated margins Routine blood and urine investigations were advised and found to be normal.

The treatment, initially, consisted of an incisional biopsy under local anaesthesia which his to pathologic study revealed presence of multiple bits of soft tissue, showing encapsulated tumor mass.

This mass contained spindle shape cell, duct like structure, within scanty connective tissue stroma. In the superficial areas focal calcification is seen. The overall his to pathological features are suggestive of Adenomatoid Odontogenic Tumor

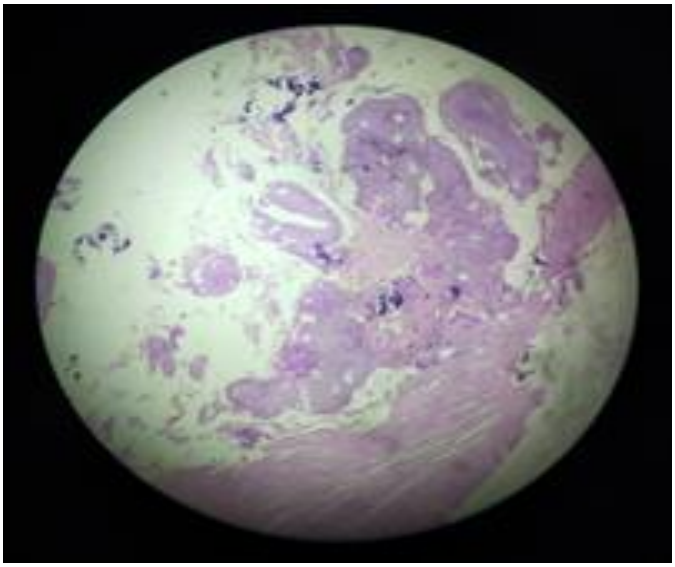


Fig 3: (G)Histopathology revealed cuboidal to columnar cells arranged in the form of nests and rosettes. Duct-like pattern, whorled arrangement of cells, and tubular appearance is evident. Convoluted structures were noted and at the periphery of the lesion tumor cells are arranged in a strand-like configuration. Some amount of calcification was also observed.

Differential diagnosis

On the basis of the clinical findings a provisional diagnosis of dentigerous cyst was given and the following other lesions were thought of while listing the differential diagnosis: calcifying odontogenic cyst, central ossifying fibroma, ameloblastoma, calcifying epithelial odontogenic tumour, central cemenifying fibroma.

Treatment

The surgery was performed under general anaesthesia. Vestibular incision was given from region of 33 to 43. Blunt dissection was done to expose the lesion. The bone appeared to be thinned but was intact. Lesion was enucleated completely. There were no apparent in filtrations of the surrounding bones. The retained canine was completely embedded in the cyst. The oral defect was closed by primary intention.

Discussion

AOT is a benign, hamartomatous, non-aggressive lesion with slow but progressive growth. Adenomatoid Odonto genic tumors (AOT) are benign slowly progressing growths that account for 2.2-7.1% of all odontogenic tumors. Overall, AOT has low incidence. [6] AOT often presents as a painless swelling of the jaw, usually occurring in

H I J

Fig 4: (H) and (I) shows en masse enucleation of the lesion with the impacted tooth, (J) clean cavity after removal of mass in toto

K L

Fig 5: (K) shows removal of lesion in toto and, (L) on cut opening of the lesion impacted lateral incisor can be seen along with calcified deposits.

the anterior region of the maxilla than in the mandible and the anterior jaw is much more affected than the posterior area, approximately twice as often in females as in males. [4]

According to Philipsen and Reichart the AOT appears in three clinico – to po graphic variants: follicular, extra follicular, and peripheral. The follicular and extra follicular variants are both intra bony and account for approximately 96% of all AOTs of which 71% are of follicular type. [7]

Radio graphically, the intraosseous variants share some features as well-defined radiolucency with small radiopacities, adjacent tooth displacement, size usually no more than 3 cm; root resorption and cortical perforation are rare. Despite their similarities, the follicular type is associated to an included tooth, being the canine the more frequent; while the extrafollicular type is not Associated to an included tooth and can appear bet ween, above, below or superimposed to the roots of permanent teeth. In suspicion of presence of AOT, peri apical radiography shows better the calcified deposits within the lesion. [4]

Clinical features generally focus on complaints regarding a missing tooth. The lesion usually present as asymptomatic swelling which is slowly growing and often associated with an unerupted tooth. However, the rare peripheral variant occurs primarily in the gingival tissue of tooth-bearing areas. Unerupted permanent canine are the teeth most often involved in AOTs. [8]

The differential diagnosis of the AOT depends on the variant found. The peripheral variant is differentiated only with gingival pathologies such as epulis and gingival fibroma. The diagnosis is purely his to pathological.[9] As most of the cases of the AOT are associated to an unerupted tooth, the differential diagnosis most include the dentigerous cyst; nonetheless, when the radiolucency exceeds the cementum-enamel junction it has to be ruled out. Because of the small radiopacities within the lesion, the epithelial odontogenic tumor and the epithelial Odon to genic cyst should be considered as well as the ossifying fibroma and Cement ossifying fibroma. Presenting as a unilocular radiolucency it could appear as an unicyclic ameloblastoma. [10]

As the tumour was well encapsulated and benign in nature and was separated easily from the bone,

conservative surgical enucleation was the treatment of choice.

Conclusion

AOT is a rare slow-growing painless, non-invasive tumor, most often misdiagnosed as a dentigerous cyst. Although it affects young individuals, mainly females, commonly found in the anterior maxilla and associated with an impacted canine, in our case anterior mandibular region was affected. The radiograph to shows radiopacities in AOT as discrete foci having a flocculent pattern within radiolucency even with minimal calcified deposits. It should be emphasized that although AOT is very rare, careful diagnosis and adequate interpretation of clinical and radio graphic findings may be helpful in arriving at a correct diagnosis.

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