Case Report

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Adenomatoid Odontogenic Tumour Associated with Dentigerous Cyst: A Rare Case Report

Abstract:
Adenomatoid Odontogenictumour is an uncommon benign lesion of odontogenic origin which affects young individuals mostly in second decade of life, with a female predominance. AOT is most commonly located in anterior maxilla usually associated with impacted canine. Through this paper we present a rare entity – Adenomatoid Odontogenictumour with dentigerous cyst associated to impacted canine and lateral incisor in anterior maxilla in a 15 year old male patient.

Keywords: Adenomatoid Odontogenictumour, Dentigerous cyst, Impacted tooth, Hybrid AOT, pseudoameloblastoma.

Introduction

Adenomatoidodontogenic tumor is a benign tumour of odontogenic origin and was first described by Driebaldt in 1907 as a pseudoameloblastoma. Philpsen and Birn in 1977 proposed the widely accepted name adenomatoidodontogenic tumor.[1] AOT constitutes about 2 – 3% of all the odontogenictumor[2] usually found in young patients, predominant in females with a Male : Female ratio of 1:1.9.[3] AOT occurs both intraosseosly and extraosseosly[1] and was subdivided in 3 variants -follicular, extra follicular and peripheral[4] they all share a histological characteristic which is indicative of a common origin from remnant of dental Lamina.

The most common is follicular type (73%) and it is mostly associated to impacted tooth. Extrafollicular type (24%) has a central lesion and no connection with the tooth whereas peripheral type (3%) is the rarest, according to literatures available.[5,6]

Association of AOT with odontogenic cyst is very rare. Literature shows that only 14 cases of AOT with Dentigerous cyst have been reported.[2] Therefore this case report is a rare entity of AOT with Dentigerous cyst in anterior maxilla of a male patient.

Case Report:

A 15 years old male patient reported to the OPD of Oral Medicine and Radiology with chief complaint of swelling in the right upper face region since 4 ½ months.

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The swelling was initially small and gradually increased in size over a period of 4 ½ months. It was associated with pain which was dull and intermittent. No history of pus discharge, hot foamentation or trauma. Medical and dental histories were non – contributing.

Local Examination:

Extra oral findings revealed facial asymmetry with a well-defined swelling on the right mid face region measuring approximately 2X3 cm in size extending superoinferiorrly 1.5 cm below infraorbital margin to the corner of mouth and mediolaterally from ala of the nose to 3 cm anterior to the tragus of ear. On palpation swelling was firm in consistency. The overlying skin was afebrile without changes in colour and texture.

Intraoral findings revealed a well-defined swelling extending from the right maxillary central incisor to the 1st premolar, obliterating the right buccal vestibule. Measuring approximately 1cm x 0.5cm in size. The right maxillary lateral incisor and canine were missing and the overlying mucosa was stretched. On palpation the swelling was firm in consistency, with mobility in first and second premolar. Both buccal and palatal cortical plates showed expansion.

Fine needle aspiration cytology revealed straw coloured fluid was aspirated. On the basis of clinical examination a provisional diagnosis of Adenomatoid Odontogenic Tumour, and a differential diagnosis of dentigerous cyst, ameloblastoma and calcifying epithelial odontogenictumour was made.

Pulp vitality was done on the teeth, associated with the swelling tested to be vital.

Radiographic examination:

The intraoral periapical radiograph of maxillary lateral and canine region revealed a well circumscribed unilocular radiolucency in respect to [12, 13] region and was associated with impacted [12, 13] and distally displaced crown of [12]. In addition specks of calcification in the radiolucent area were present.

On seeing the radiolucency and impacted tooth further occlusal maxillary topographic and Orthopantomogram (OPG) radiographs were advised. OPG was suggestive of unilocular radiolucency surrounded by thin radiopacity with impacted 12, 13 and displaced 14 (mesially displaced root)with flecks of calcification [Figure 1]. Occlusal maxillary cross sectional radiograph revealed thinning with expansion of buccal cortex extending from 12 to 14 region.

The patient was advised surgery under general anaesthesia, the lesion was surgically enucleated along with impacted 12 and 13 and was sent for histologic analysis.

Figure 1: Photograph of orthopantomogram showing unilocular radiolucency with impacted 12, 13 and displaced 14 (distally displaced root) with flecks of calcification

Histopathological Analysis:

Under scanner view, section shows highly cellular stroma. On higher magnification, sheets of polyhedral odontogenic epithelial like cells throughout stroma of varying shape were seen. These odontogenic cells in certain areas exhibit whorled and rosette like patterns (Figure 2).

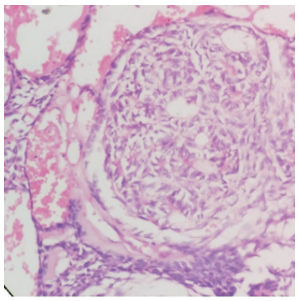

Figure 2: Photomicrograph under scanner view, section shows highly cellular stroma. On higher magnification, sheets of polyhedral odontogenic epithelial like cells throughout stroma of varying shape were seen. These odontogenic cells in certain areas exhibit whorled and rosette like patterns. In other areas section shows tubular duct like structures lined by single layered row of cuboidal epithelial cells.(H&E stain) (40x)

In other areas section shows tubular duct like structures lined by single layered row of cuboidal epithelial cells. In another section under scanner view, cystic lining is seen overlying connective tissue stroma. On higher magnification, 1-2 layered squamous epithelium is seen. Cells are loosely attached and the connective tissue stroma shows dense inflammatory infiltrate comprising of interlacing collagenous bundle with small blood vessels, fibroblasts and extravasated RBC's (Figure 3).

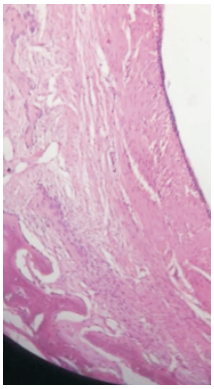

Figure 3: Photomicrograph - under scanner view, showing cystic lining overlying connective tissue stroma. On higher magnification, 1-2 layered squamous epithelium is seen. Cells are loosely attached and the connective tissue stroma shows dense inflammatory infiltrate comprising of interlacing collagenous bundle with small blood vessels, fibroblasts and extravasated RBC's. (H&E stain) (40x)

On the basis of histopathological analysis final diagnosis of AOT associated with dentigerous cyst was made.

Discussion :

AOT are uncommon non – aggressive tumours of odontogenic epithelium in variety of patterns mixed with mature connective tissue stroma, constituting 2-3% of all oral tumours.[2,7] In the present case the patient was a male aged 15 years with a lesion in anterior maxilla and impacted maxillary lateral incisor and cuspid, which goes in accordance with the literature available.

Garcia Pola et al. (1998), described the proliferation of AOT in the epithelial border of a dentigerous cyst, Santos et al.(2007) reported a case of AOT which developed in the fibrous capsule of the dentigerous cyst. Whereas Cassiano Francisco Weege et al, reported a case of AOT which was associated with dentigerous cyst.[6,9] Most commonly AOT is associated with dentigerous cyst occurring in the anterior maxilla as in our case and other areas of the jaw such as the angle of the mandible.

Radiologically AOT usually surrounds an impacted, unerupted tooth and is seen as well-definedunilocularradiolucency withcorticted borders and in few cases is associated to small radiopacities. AOT most oftenly appears enveloping the crown as well as the root of the involved tooth but in cases of dentigerous cyst the root is not enveloped.[5]In our case a corticated radiolucency with small radio-opacities was seen enveloping the crown and root of impacted canine and lateral incisor.

A review of literature of Clinical and radiographic details of previously reported cases in literature.[11-20]

Sl.no	Author	Year	Age/ Gender	Clinical Finding	Site	Radiographic Details
1.	Valderrama	1988	16/F	Impacted 14	Maxilla	Unilocular
2.	Warter et al.	1990	8/M	Impacted 13	Maxilla	Unilocular
3.	Tajima et al.	1992	15/M	Unerupted crown of 28	Maxillary Sinus	Radiopaque mass
4.	Garcia-Pola Vallejo et al.	1998	12/M	Impacted 23	Maxilla	Unilocular
5.	Takahashi et al.	2001	22/M	Impacted 28	Maxilla	Unilocular
6.	Bravo et al.	2005	14/M	Impacted 23	Maxilla	Unilocular
7.	Nonaka et al.	2007	13/F	Impacted 23, 24	maxilla	Unilocular
8.	Sandhu et al.	2010	25/F	Impacted 13	Maxillary sinus	Unilocular
9.	John and John	2010	39/F	Impacted 27	Maxilla	Unilocular
10.	Moosvi et al.	2011	13/F	Impacted 32	Mandible	Unilocular

AOT'sand Dentigerous cysts are benign encapsulated lesions, are successfully treated by enucleation and curettage with low recurrence rate.[10,21]

Conclusion:

This case of AdenomatoidOdontogenicTumour associated with dentigerous cyst emphasizes the importance of proper diagnosis of neoplasm's arising from odontogenic tissues. Thus meticulous histopathological evaluation should be conducted in all cases of enucleated cyst for appropriate diagnosis. AOT with dentigerous cyst gives a clue of a hybrid kind of AOT other than the 3 variants

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