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Adenomatoid Odontogenic Tumor (An Uncommon Tumor): A Case Report

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Abstract

Adenomatoid odontogenic tumor (AOT), which appears mostly in young females with highest occurrence in the maxillary region, is a hamartomous benign neoplasm of odontogenic origin. It is a slow growing, asymptomatic lesion but hampering the esthetics. It is mainly related to non-erupted canines. Lesions of this type can be classified as follicular, extra follicular and peripheral lesions. Treatment of these lesions is enucleation and curettage of the affected area. Recurrence is rare. A case of adenomatoid odontogenic tumor in a twelve-year-old female which was associated with an impacted maxillary left canine teeth has been reported in this paper.

Keywords: Canine; Impacted; Maxilla; Tumor

Introduction

Adenomatoid odontogenic tumors (AOTs) are rare, slow growing, benign, odontogenic, and epithelial tumors. They are characterized by slow but progressive growth without any pain.[1] Adenomatoid Odontogenic Tumor is also called "tumor of two third" because of the occurrence of two third of these cases in young females in the maxillary region which are associated with unerupted canines and are mainly diagnosed in the second decade of life. Here, we describe a follicular type of adenomatoid odontogenic tumor in the anterior maxilla of a twelve year old female patient. [2-5]

Case report

It is one of the rare tumors of the oral cavity. This tumor needs to be diagnosed at an early age and treated accordingly so as to prevent significant facial deformity that may occur in later stages of life. A twelve year old female patient presented to our outpatient department with the chief complaint of swelling on the left side of face for six months. She gave a history of gradual increase in the size of swelling without a history of pain. Extra orally, a swelling with a hard and smooth surface of size approximately 4cm x 3cm was noted on anterior maxilla extending anteroposteriorly from ala of the nose to about 3cm ahead of the tragus of ear , and superoinferiorly about 2cm below the infraorbital rim to ala tragal line with the obliteration of the nasolabial fold on the left side. On intra-oral examination, swelling of approximately 3cm x 2cm was detected extending from left central incisor to left second premolar with firm and smooth surface. The left upper canine was missing. There was no evidence of oro-nasal and oro-antral communication and palatal mucosa was intact. On radiographic examination, well-circumscribed unilocular radiolucent area was seen, involving impacted upper left canine. Also, the floor of the left maxillary sinus appears to be displaced upward in the radiograph. On the basis of clinical and radiographic findings, differential diagnosis of adenomatoid odontogenic tumor, unicystic ameloblastoma and dentigerous cyst were made. Enucleation of the lesion was done under general anesthesia and the specimen was sent for histopathological examination. A final diagnosis of Follicular variant of Adenomatoid odontogenic tumor was confirmed.

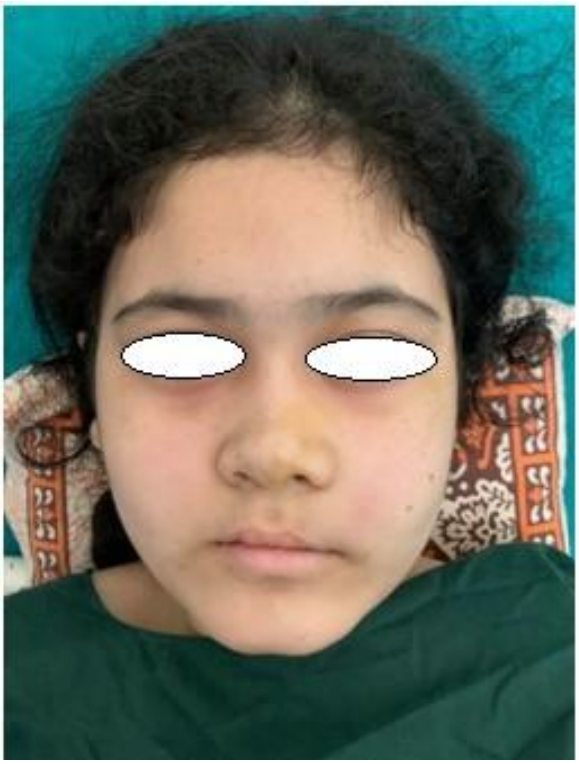


Figure 1: Preoperative Extraoral Photograph

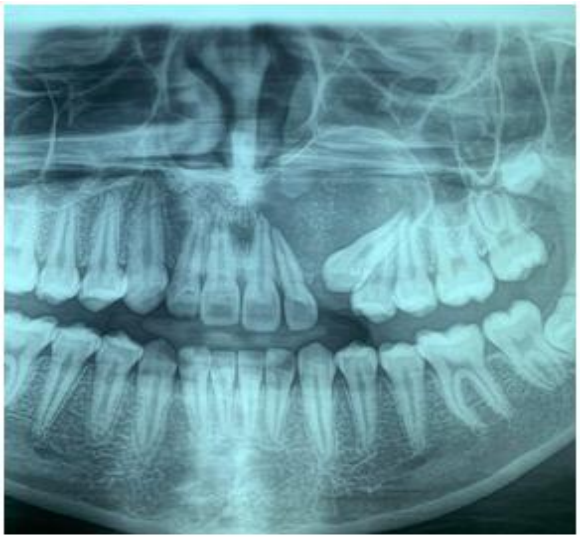
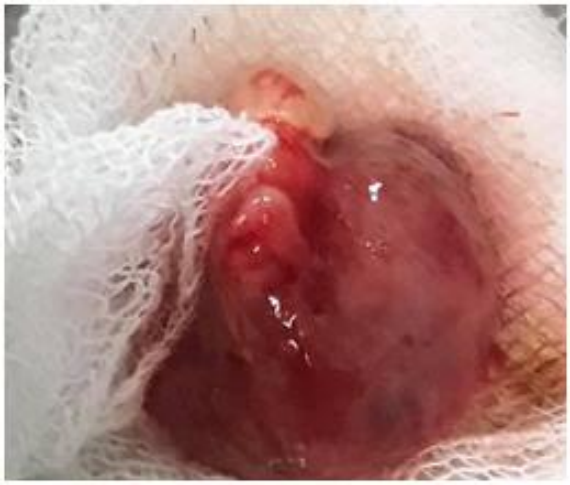


Figure 2: Preoperative panoramic radiography showing well circumscribed radiolucency around the impacted left maxillary canine



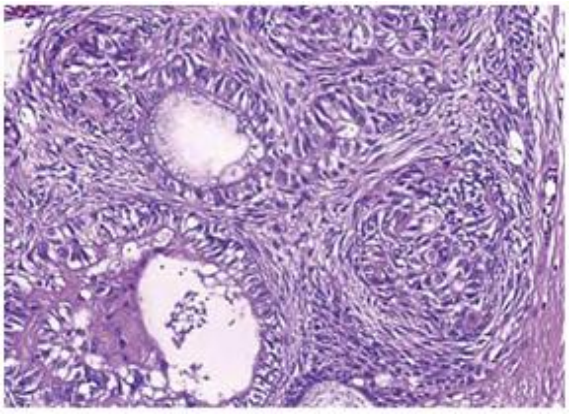


Figure 3: Excised Specimen of size 3cm x 2cm
Figure 4: Histopathological section of excisional biopsy along with impacted canine specimen (10X)

Discussion

Adenomatoid odontogenic tumor (AOT), first described by Dreibaldt as 'Pseudoadenoamelo-blastoma' in 1907 and consecutively by Harbitz as 'Cystic Adamantoma' in 1917, is an uncommon benign epithelial lesion of odontogenic origin. [1, 4, 6] in 1948, it was considered a distinct entity by Stafne and in 1969, Philipsen and Birn called it as 'Adenomatoid Odontogenic Tumor'. [1] AOT was describes as 'A tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive changes in the connective tissue' by World Health Organization. [7]

Dental lamina, enamel organ, reduced enamel epithelium with its remnants have been cytologically related to AOT. Whether to consider AOT as a hamartoma or neoplasm is still a topic of debate. Radiographically, AOT resembles a dentigerous cyst, which is usually unilocular and radiolucent. However, fine calcifications (snowflake), a feature consistent with AOT is often seen on radiographs that may be helpful in differentiating an AOT from a dentigerous cyst. The unilocular cyst is well demarcated with a smooth cortical border. Most lesions are located on pericoronal and juxta coronal area. Divergence of roots and displacement of teeth without root resorption are often seen. [5] In this case, similar features were present

The tumor may be presented as partly cystic while in some cases, the solid lesion presents itself only as a mass in the wall of a large cyst. Some eosinophilic uncalcified and amorphous material can be found, which is called tumor droplets. [2, 8, 9] In this case the mass was a cystic non solid mass involving the impacted tooth and similar histological features were seen as described by WHO. Conservative surgical enucleation and curettage is the treatment modality of choice since all the variants of AOT are benign and well encapsulated. The tumor should be removed in Toto. Recurrence of this tumor is extremely rare. Also, cosmetic disfigurement can be avoided if patient of AOT is diagnosed at early age and provided proper treatment because cortical expansion is very common in AOT. [5, 10] This was a classical follicular variant of AOT with no recurrence after six month follow up period. Patient was satisfied with her facial appearance and no facial asymmetry was seen.

Conclusion

AOT rarely recurs and if it is removed in-Toto, better results are obtained. This also helps satisfy the patient toward the efforts of the clinician. Enucleation and Curettage for AOT has been the most common treatment modality, yet requires histological diagnosis so as to carry out minimally invasive surgery.

Conflict of interest

The author declares no conflict of interest.

Acknowledgments

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References

1. Philipsen HP, Reichart PA, Nikai H. The Adenomatoid Odontogenic Tumour (AOT): An Update. Oral Medicine & Pathology. 1997;2(2):55-60. DOI: 10.3353/omp.2.55
2. John JB, John RR. Adenomatoid Odontogenic Tumor Associated with Dentigerous Cyst in Posterior Maxilla: A case Report and Review of literature. J Oral and Maxillofacial Pathology: JOMFP. 2010;14(2):59. DOI: 10.4103/0973-029X.72502
3. Nair, M., Ajila, V., Hegde, S., Babu, S., & Ghosh, R. (2021). Cone Beam Computed Tomography Findings of a Large Adenomatoid Odontogenic Tumour in the Anterior Mandible- A case report . Annals of Medical Research, 24(1), 0075–0078. https://annalsmedres.org/index.php/aomr/article/view/2133
4. Dayi E, Gürbüz G, Bilge OM, Çiftcio, gcarlu MA. Adenomatoid Odontogenic Tumour (adenoame-loblastoma). Case Report and Review of the literature. Australian Dental Journal. 1997;42(5):315-318. DOI: 1834-7819.1997.tb00136.x
5. Neha S, Santosh M, Sachin MG, Poonam SR, Simranjit S, Abdul KA. Adenomatoid Odontogenic Tumour: An Enigma. the Saudi Dental J 2018; 30(1):94-96. DOI:10.1016%2Fj.sdentj.2017.10. 005
6. Grover S, Rahim AMB, Parakkat NK, Kapoor S, Mittal K, Sharma B, et al. Cystic Adenomatoid Odontogenic Tumor. Case Reports in Dentistry. 2015. DOI: 10.1155/2015/503059
7. Manjunatha B, Mahajan A, Mody BM, Shah V. Adenomatoid Odontogenic Tumor (AOT) Arising from a Dentigerous Cyst: Literature Review and Report of a Case. J Maxillofacial and Oral Surgery. 2015;14(2):393-397. DOI: 10.1007%2Fs12663-012-0369-3
8. Katpar S, Kashif M, Azad N, Qureshi NR. Maxillary Adenomatoid Odontogenic Tumour-An Uncommon Oral Pathology, Reported Locally. J Liaquat Uni Med Health Sci. 2010;9(3):155-158.
9. Motamedi M, Shafeie H, Azizi T. Salvage of an Impacted Canine Associated with an Adenomatoid Odontogenic Tumour: A Case Report. British dental J. 2005;199(2):89-90. DOI: 10.1038/sj.bdj.4812522
10. Handschel JG, Depprich RA, Zimmermann AC, Braunstein S, Kübler NR. Adenomatoid Odontogenic Tumor of the Mandible: Review of the Literature and Report of a Rare Case. Head & Face Medicine. 2005;1(1):1-5. DOI: 10.1186/1746-160x-1-3