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A Rare Case of Acanthomatous Ameloblastoma

Dr.Guruaribam Victoria Devi[1], Dr .Amudhan .A[2]
[1]*Postgraduate, Department of Oral Medicine and Radiology ,SreeBalaji Dental College and Hospital, BIHER ,Pallikaranai,Vellacherry Main Road, Chennai, Tamil Nadu ,600100*
[2]*Associate professor, Department of Oral Medicine and Radiology ,SreeBalaji Dental College and Hospital, BIHER ,Pallikaranai,Vellacherry Main Road, Chennai, Tamil Nadu,600100*
Corresponding Author: Dr .Amudhan .A

Abstract: *Odontogenictumorscomprise a complex group of lesions of diverse histopathologic types and clinical behavior. Ameloblastoma is the most frequent odontogenictumor arising from dental epithelium, and is characterized by its histological resemblance to the enamel organ of the developing tooth germ, yet enamel formation is not observed. Thorough imaging investigation, clinical examination and histopathological analysis is mandatory for a definitive diagnosis. Here we present a case report of a 20 year female patient with mild discomfort in the lower left third molar region without any extra oral swelling which was thought to be a case of impacted third molar which was later diagnosed as ameloblastoma after, radiographic and histopathological analysis.*

Keywords: *Ameloblastoma, Odontogenic, Acanthomatous*

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I. Introduction

Odontogenictumorscomprise a complex group of lesions of diverse histopathologic types and clinical behavior. Some of these lesions are true neoplasms and may rarely exhibit malignant behaviour. Epithelial odontogenictumors are composed of odontogenic epithelium without participation of odontogenicectomesenchyme. [1]

The most frequent odontogenictumor arising from dental epithelium is the ameloblastoma, and is characterized by its histological resemblance to the enamel organ of the developing tooth germ, yet enamel formation is not observed.[2] It has been postulated that the epithelium of origin is derived from one of the following sources: (a) cell rests of the enamel organ, (b) epithelium of odontogenic cysts, (c) disturbances of the developing enamel organ, (d) basal cells of the surface epithelium, or (e) heterotropic epithelium in other parts of thebody.[3]

Here, we are reporting a case of 20 year old female patient with a mild pain in the lower left back teeth region with no extra-oral swelling.

II. Case Report

A 20 year old female patient came to the Department of Oral Medicine and Radiology with a chief complaint of mild pain and swelling in the lower left back teeth region since 2-3 days. History revealed that the patient noticed a small swelling in the gums of the left lower back teeth region only 2-3 days back .There was association of a very mild,intermittent type of pain in the region. Medical, family, and personal histories were non‑contributing. No extra-oral swelling and facial asymmetry was noted. Intra-oral examination of the hard tissues revealed clinically missing,18,28,38 and 48. Intra oral examination of the soft tissue (Figure:1)revealed a solitary diffuse swelling of size approximately 1.5 X 2cm in relation to the distal aspect of 37 region extending anterio-posteriorly from the distal aspect of the 37 till the retromolartrigone area and mediolaterally from the buccal gingiva of 37(distal surface) to the lingual aspect of 37 region with lingual and buccal cortical expansion .On palpation,the swelling was soft in consistency ,non-tender and non-compressible.No blood or pus exudate was noted. Mild obliteration of the lower left buccal vestibule in the 37,38 region.

A provisional diagnosis of erupting 38 was suggested and IOPA and OPG radiographs was taken. IOPA (Figure:2) revealed a well-defined radiolucency in the distal and periapical aspect of 37 with mild resorbtion of the mesial root of 37 with periodontal ligament widening and loss of lamina dura. 38 was displaced towards the base of the mandible.

OPG(Figure:3) revealed a multilocular radiolucency of approximately 5X7cm in size in the left ramus of the mandible from the distal and periapical aspect of 37 and extending anterioposteriorly towards the left condylar head.

Further the patient was advised to take a computed radiograph for detailed analysis of the lesion. CT (Figure4,5,6,7) revealed an expansilemultilocular lytic lesion measuring about 4.5X1.8X3.5 cms(Craniocaudal x Transverse x Anteroposterior) in the ramus and angle of the left mandible. Significant cortical thinning and areas of cortical break and unerupted 38 was also evident. Based on the above discussed findings a provisional diagnosis of odontogenickeratocyst in relation to the unerupted 38 was suggested.

An incisional biopsy was done and the resected specimen was sent for histopathological examination.

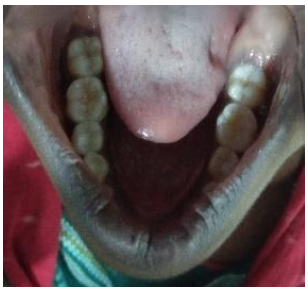

Figure 1.

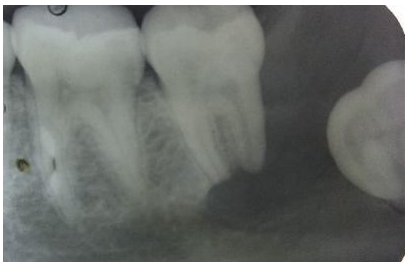

Figure 2

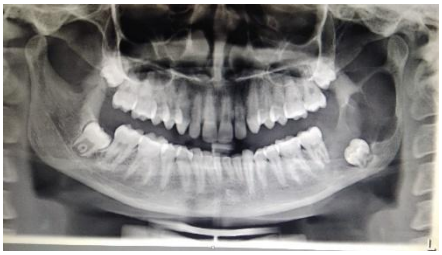

Figure 3

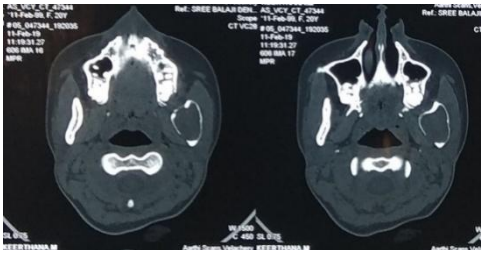

Figure 4

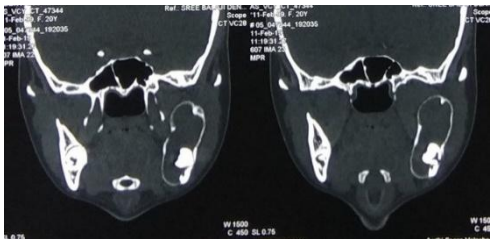

Figure 5

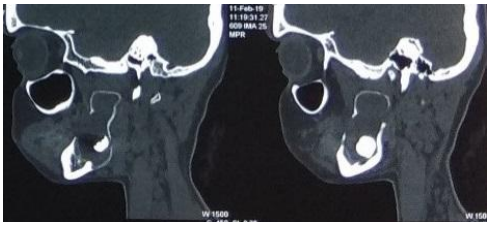

Figure 6

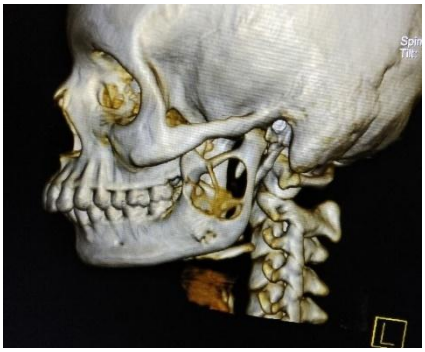

Figure 7

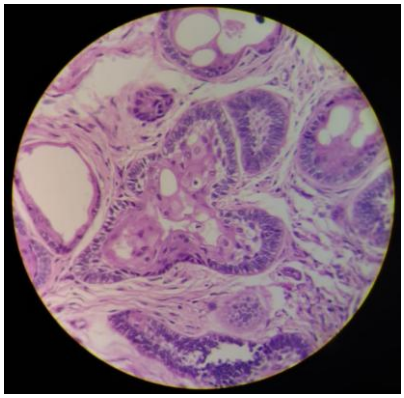

Figure 8

III. Histological Findings

The hematoxyline and eosin stained histopathological sections of the specimen showed cystic lining epithelium of 1-2 cell layer thick exhibiting ameloblastomous changes like tall columnar cells ,hyperchromatic nuclei and reversal of polarity.Subnucleolar vacuolization with central stellate reticulum like cells. Areas of normal surface epithelium with parakaratenised stratified squamous type. Few islands show cystic degeneration and squamous metaplasia with keratin formation .Connective tissue showing numerous odontogenic epithelium arranged in the form of islands and strands exhibiting features like palisaded basal layer,hyperchromatic nuclei with central stellate reticulum like cells .The dense connective tissue showed increased cellularity with diffuse dense chronicinflammatory cell infiltrate. (Figure 8) Correlating all the clinical,radiographic and histopathological analysis, a final diagnosis of acanthomatousameloblastoma was given.

On the basis of above diagnosis patient was planned for surgery. Hemi-mandibulectomy was performed. Reconstruction was carried out with stainless steel reconstruction plate and advancement local flaps. Patient was followed up for one year and there was no signs of recurrence.

IV. Discussion

Ameloblastoma is classified according to WHO and the International Agency for Research on Cancer, 2003 as a benign tumor with odontogenic epithelium, mature fibrous stroma and without odontogenic ectomesenchyme.[5]It is further classified into: Solid/multicystic, extraosseous/peripheral, desmoplastic and unicystic. The multicysticameloblastoma has histological variants including follicular, plexiform, acanthomatous, granular cell, desmoplastic and basal cell types.[6]Follicular ameloblastoma is the most prevalent histological variant (64.9%), followedby the plexiform (13.0%), desmoplastic (5.2%) and acanthomatous (3.9%) varieties.[7]

In acanthomatousameloblastoma, the cells occupying the position of the stellate reticulum undergo squamous metaplasia and keratin formation within the island of tumor.[3]

About 80% of all cases occur in mandible, of which 70% cases are seen in the ramus which was also evident in our case.[4]

Slow-growing mass, malocclusion, loose teeth or sometimes paresthesia and pain are some of the signs and symptoms .But many lesions are detected incidentally on radiographic studies in asymptomatic patients. In our case report also, there was no extra-oral swelling and it was an incidental radiographic finding.

According to literature, ameloblastoma is noted for its persistent slow growth, local invasiveness and high rate of recurrence. When the tumour enlarges, it may cause thinning of the cortical bone resulting in an egg shell crackling.[10]

Assessment of cellular proliferative activity of the odontogenic cysts and tumors is mainly done by the Ki-67 index which plays an important prognostic marker of tumor recurrence, biological behaviour, and local invasiveness.[9]Surgery with wide resection is the treatment of choice. Majority of the surgeons recommend a margin of 1.5–2 cm beyond the radiological limit to ensure all microcysts are removed.[8] Lifelong follow up is always recommended. Rare malignant transformation has also been documented.

In our case the patient was mostly asymptomatic apart from the mild gingival swelling in the third molar region which was thought to be an erupting 38. This particular case shows the importance of taking radiographs and correct histopathological analysis to come a correct diagnosis.

V. Conclusion

In the jaws various pathological conditions are manifested. Ameloblastomas are benign odontogenictumors that rarely become malignant. The acanthomatous subtype is rare entity.

Thorough imaging investigation and clinical examination is mandatory for correctly identifying the internal architecture and its adjacent anatomical structures to assist in treatment planning. It can be concluded that diagnosis based on histopathology is the definitive diagnosis.

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