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Case Report

Acanthomatous Ameloblastoma of Mandible: A Rare Clinical Presentation

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Abstract— *Ameloblastoma is a benign locally aggressive infiltrative odontogenic tumour commonly located in the jaw bone. Patients with this tumor usually presents in late stages with facial disfigurement caused by expansion of the jaw bones. The most common site of involvement is mandibular third molar region. Among the histological variants, acanthomatous ameloblastoma is the rarest of all. It may pose a diagnostic challenge when ameloblastoma manifests as a small alveolar swelling in the anterior region of jaws without any distinct features. Here we are reporting such a case of 44-year-old male with a small swelling on right anterior mandible region which was diagnosed as acanthomatous ameloblastoma based on clinical, radiographic and histopathological features.*

Keywords— *Acanthomatous Ameloblastoma, odontogenic tumour*.

I. INTRODUCTION

Ameloblastoma is a benign odontogenic tumour derived from the tooth producing apparatus. In 1930, Ivey and Churchill coined the term "ameloblastoma".[1] It is defined as: Unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent.[2] The source of origin is believed to be epithelium of the enamel organ, residual epithelium from tooth germ or epithelium of odontogenic cyst.[3]

There are intra osseous and peripheral/extraosseous ameloblastoma. Intraosseous ameloblastoma is further categorised into solid or multicystic, unicystic, and desmoplastic. The histological subtypes of ameloblastoma are follicular, plexiform, acanthomatous, granular cell, basal cell and clear cell types.[4] The present case report is one such rare variant of acanthomatous ameloblastoma affecting the mandible which was diagnosed based on clinical, radiographic and histopathological features.

II. CASE REPORT

A 44 year old male patient reported to the department of oral medicine and radiology with a chief complaint of a gradually progressing painless swelling in lower right front tooth region since two weeks. His medical history was non-contributory. Intra oral examination revealed a solitary diffuse swelling of size approximately 2× 2 cms in relation to 43,44. (Figure 1) Swelling was non-tender and hard in consistency. Provisionally considering it as a case of lateral periodontal cyst, a radiological examination with Intra oral periapical radiograph (IOPA), Occlusal radiographs and OPG was done.

Panoramic radiograph and IOPA in relation to 43, 44 revealed an interdental multilocular radiolucency of size around 2×1 cm showing honey comb pattern with displacement of the root of 44. (Figure 2, Figure 3) No root resorption was noted. Occlusal radiograph showed a mild buccal expansion of the mandible in the area of interest.

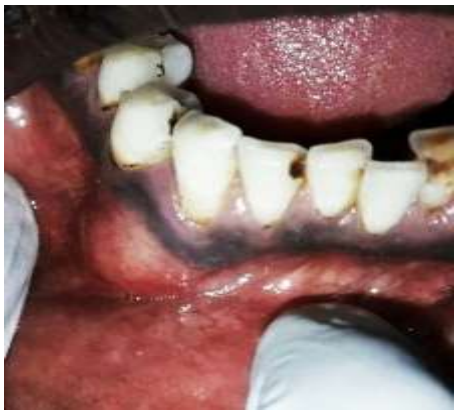

Fig. 1.

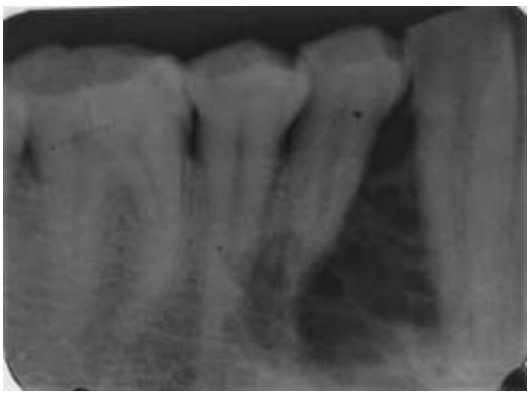

Fig. 2.

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(Figure 4) Excisional biopsy was done and the resected specimen was sent for histopathological examination. Microscopically, haematoxylin and eosin stained section revealed solid epithelial cell nests with peripheral palisading basaloid cells and central squamoid cells. Tumor cells nests were surrounded by fibrous stroma. (Figure 5) Based on the above histological features a diagnosis of acanthomatous ameloblastoma was made. Patient was followed up for one year and to date there has been no evidence of recurrence.

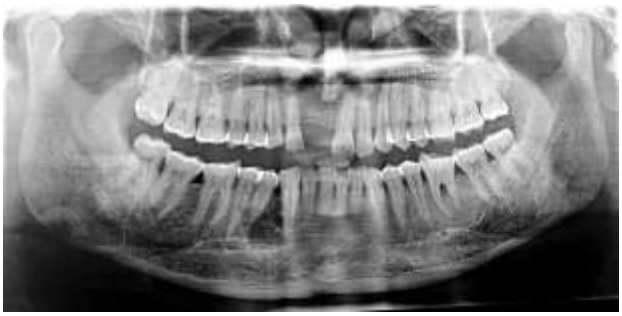

Fig. 3.

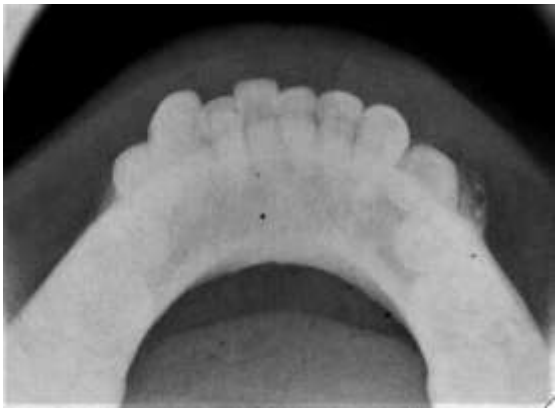

Fig. 4.

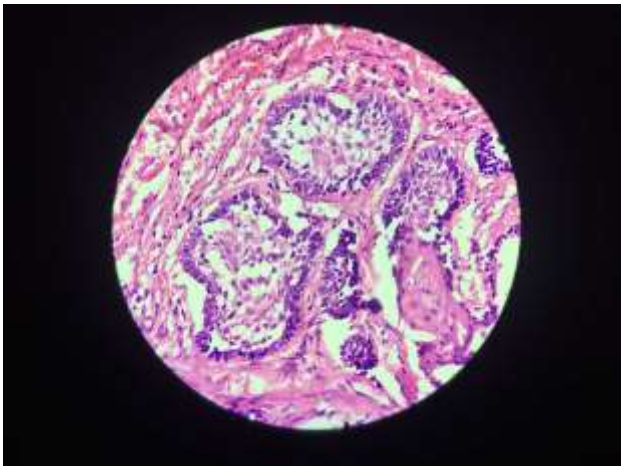

Fig. 5.

III. DISCUSSION

Ameloblastoma accounts for 1% of all tumors in head and neck region and around 11% of all odontogenic tumors. Average age reported is in the third to fifth decade of life with no significant sex predilection. About 80% of the ameloblastomas occur in the mandible, out of which 70% are located in the area of the molars or the ascending ramus, 20% occur in the premolar region, and 10% in the anterior region.[5] The patient reported here, had a swelling in the canine premolar region which is a very rare site of involvement.

As literature states ameloblastoma is noted for its persistent slow growth, local invasiveness and high rate of recurrence. As the tumour enlarges, it may cause thinning of the cortical bone resulting in an egg shell crackling.[6] If left untreated, ameloblastoma can resorb the cortical plate and extend into adjacent soft tissues.[5] In our case the patient only reported regarding a slowly progressive swelling. The tumor dimension was very small. Other associated features with the tumor such as tooth mobility, pain/paraesthesia, displacement of teeth etc were also absent. Based on clinical presentation, the lesion was initially thought to be a lateral periodontal cyst. Lateral periodontal cyst will have a round to oval unilocular cystic appearance. Initial radiographic examination in our case showed a small multilocular radiolucency with honey comb pattern in the interdental region of 43 and 44 with displacement of the root of 44. The tumour was probably diagnosed at an early stage since the size of the lesion was small, there was minimal expansion and no evidence of root resorption.

The radiographic appearance of ameloblastoma can vary according to the type of tumour. In the early stage the lesion may appear cystic, unilocular and may resemble a dentigerous or a residual cyst. Later it becomes multilocular with internal compartments separated by radiopaque septa or trabeculae. The classic radiographic appearance of a mixed cystic or solid form of ameloblastoma is a multilocular radiolucency with a "soap bubble" or "honeycomb" appearance.[7] The tumours are usually expansile bucco-lingually with greatest degree of expansion occurring along the vector of tumour growth. It may cause extensive root resorption, either blunting of root apex/knife-edge root resorption.

Most literatures shows that follicular ameloblastoma is the most prevalent histological variant (64.9%) and acanthomatous (3.9%) is the rarest.[4] The follicular histopathologic form consists of loosely arranged epithelial nests resembling stellate reticulum of an enamel organ surrounded by well organised single layer of cuboidal or tall columnar cells. In acanthomatous ameloblastoma, the cells occupying the position of the stellate reticulum undergo squamous metaplasia and keratin formation within the island of tumor as seen in the present case.[2] Some authors have stated that formation of squamous metaplasia may be due to chronic irritation of calculus and oral sepsis.[2] The mixed cystic and solid form demonstrates more aggressive behaviour and a higher rate of recurrence than unicystic and desmoplastic ameloblastoma.[9]

Based on tumor size, location, histopathology, and clinical/radiographic presentation, treatment modalities range from curettage, enucleation, enucleation plus curettage to en bloc resection. Attempts to remove the tumour by curettage often leave small islands of tumour within the bone, which later manifests as recurrences. 50% of all recurrences are

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reported to be within 5 yrs. postoperatively.[10] The mainstay of treatment is surgery with wide marginal resection since tumour margins usually extends beyond the apparent radiographic boundaries seen in plain radiographs. Lifelong follow up is strongly recommended. Rare malignant transformation has also been documented.

IV. CONCLUSION

There are various pathological conditions which may manifest as a jaw swelling. Although ameloblastoma is a very common odontogenic neoplasm, it is very unusual for the tumor to present as a small swelling in the anterior region of the jaws as in the present case. Acanthomatous subtype is also a rare entity giving more chances for the tumour to be misdiagnosed. Hence, we recommend that it would be ideal to consider acanthomatous ameloblatoma in the differential diagnosis of swellings of the mandibular anterior region and histopathological examination is inevitable in acquiring a correct diagnosis.

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