CASE REPORT

ACANTHOMATOUS AMELOBLASTOMA OF MANDIBLE –A CASE REPORT

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Abstract

Ameloblastomas are a slow growing, locally invasive benign tumors of jaw. They are odontogenic in origin on the basis of histologic similarities of the tumor and the developing enamel organ. There are six histopathological variants of ameloblastoma among which, acanthomatous type is a rare variant that has distinct features. This report describes a case on an acanthomatous ameloblastoma involving the anterior mandible extending to the body of the mandible occurring in a 60 years old patient.

Key Words: Acanthomatous ameloblastoma, odontogenic, squamous metaplasia.

INTRODUCTION

Odontogenic tumors are lesions originating from epithelial and/or mesenchymal components of the tooth forming apparatus. They are unique to the jaws and if left untreated, often lead to extensive tissue destruction and deformity.[1] Ameloblastoma is a rare, locally aggressive, odontogenic jaw tumor that is a challenge to pathologists because of its diversity of histological features and to the surgeons due to its frequent defiance to complete eradication.[2] Various histological subtypes of the ameloblastomas have been described , among which the Follicular variety (64.9%) is the most prevalent, whereas the Acanthomatous variant (3.9%) is the rarest.[3]

According to Larsonn and Almeron , incidence of the acanthomatous type has been 0.6 cases per million.[3] About 80% of the acanthomatous ameloblastoma occur in the mandible, out of which 70% are located in the area of molars or the ascending ramus, 20% occur in the premolar region and 10% in the anterior region.[3]

An ameloblastoma usually presents as a slow growing mass with displacement of teeth, loose teeth , malocclusion, pathological fracture and rarely with paresthesia.[3] As the tumor enlarges it forms a hard rounded swelling, but later thinning of the cortical bone allows eggshell crackling.[4] Rarely it can ulcerate through the mucosa.[4] Radiographically,ameloblastoma appears as radiolucent lesions that may either have a multilocular or unilocular,soap bubble or honeycomb type of appearance.[3] The roots of the adjacent teeth are frequently displaced or resorbed.[4]

Several microscopic subtypes of conventional ameloblastoma are recognized, out of which the follicular and plexiform patterns are the most common and acanthomatous, granular cell, desmoplastic and basal cell types are less common variants. However, unicysic ameloblastoma is defined as single cystic cavity shows ameloblastomatous proliferation in the lining. All of these variants consist of peripheral columnar or cuboidal ameloblast like cells along with centrally placed loosely arranged, angular stellate reticulum like cells. Cystic degeneration of the stellate reticulum are also found in some areas. In acanthomatous ameloblastoma there is also presence of extensive squamous metaplasia of the central core of the epithelium, often associated with keratin

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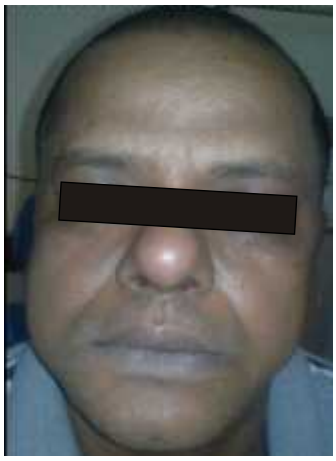

Fig. 1 : Extraoral photograph of the patient

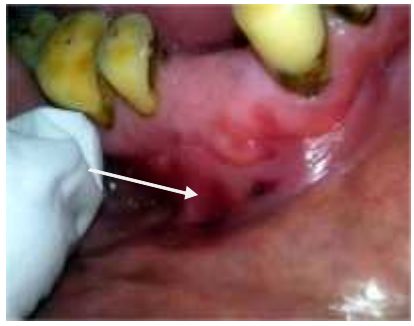

Fig.2 : Intraoral photograph showing a swelling involving the labial mucosa

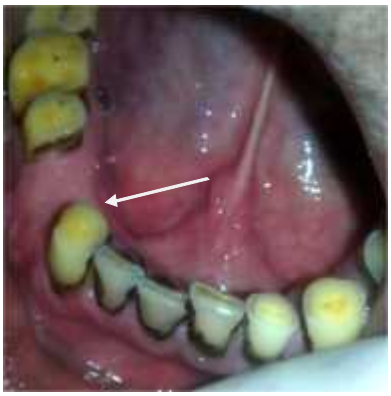

Fig.3 : Intraoral photograph showing a swelling involving the lingual mucosa

Fig.4 : Blood tinged aspirated Fluid

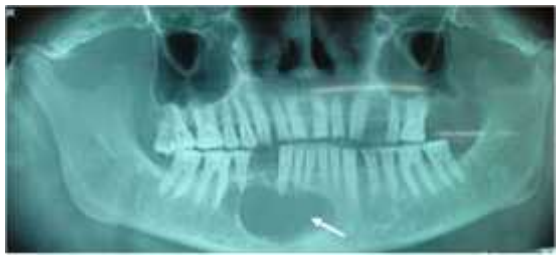

Fig.5 : Orthopantomograph showing multilocular radiolucency involving the mandible extending from the 41 to 46 with a smooth periphery

formation. Such a lesion sometimes may be confused with squamous cell carcinoma or squamous odontogenic tumor.[4,5] Rarely, an ameloblastoma exhibits frank malignant behavior with development of metastasis. The frequency of malignant behavior in an ameloblastoma is difficult to determine but probably occurs in far less than 1% of all ameloblastoma.[5]

Management of ameloblastoma has been controversial because of the unique biological behavior of this lesion as a slow-growing, locally invasive tumor with a high rate of recurrence. Recurrence rates of ameloblastoma are reportedly as high as 15-25% after radical treatment and 75-90% after conservative treatment. Therefore, wide resection of the jaw in accordance with the treatment of malignant tumors is usually recommended for ameloblastomas.[6]

Here, we are presenting a case of 60 year old otherwise healthy male with swelling involving right posterior region of mandible which was diagnosed as acanthomatous ameloblastoma based on clinical, radiographic and histopathological features.

CASE REPORT

A 60 years old male patient, from a semi urban area had reported to the Department of Oral & Maxillofacial Pathology, GNIDSR, Kolkata, with the chief complaint of a painless swelling involving the right side of lower jaw region since past 03 months. The swelling was slow enlarging and asymptomatic. Initially, it was small in size, but had later grown gradually to attain the present dimension.

The past medical and dental histories as well as the family and surgical histories were non-contributory. The patient was of average height and weight. Extra oral examination revealed a diffuse swelling involving the right side of the body of the mandible (Fig. 1). On palpation, the swelling was non-tender and firm in consistency. No regional lymph nodes were palpable. Intraoral examination showed the presence of a swelling involving the alveolar ridge of the mandible extending from the 42 to 46 region, measuring about 4cm x 3cm with obliteration of the labial and lingual vestibules (Fig.2, 3).On palpation, the swelling was firm,non-tender and bony hard in consistency. No paresthesia

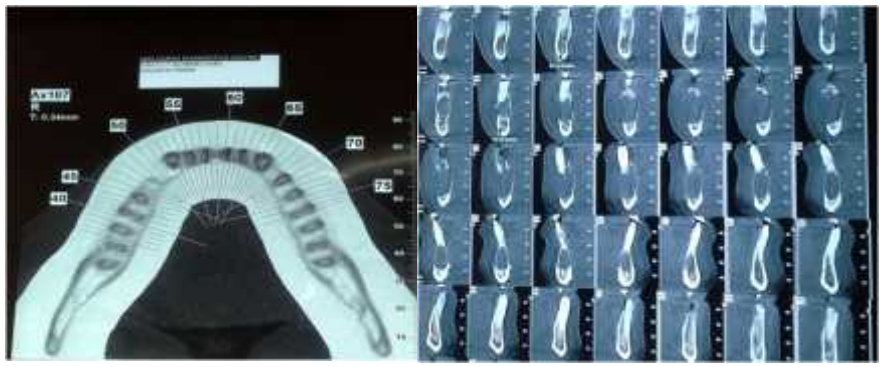

Fig. 6 : CBCT of mandible showing destruction of the buccal cortical plate & thining of lingual cortical plate of the affected site

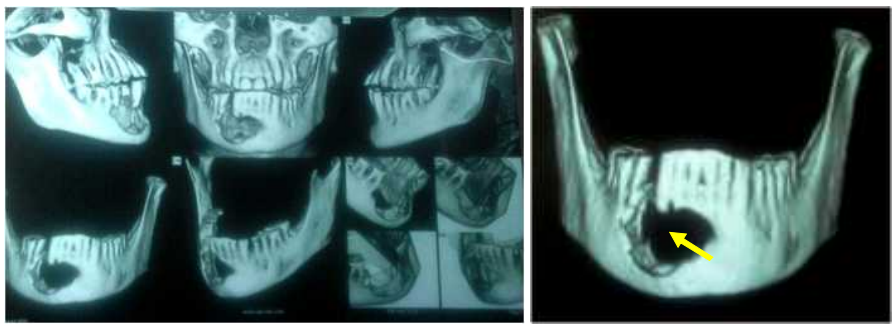

Fig. 7: 3-D reconstuction CT image showing destruction of mandible in relation to 41 to 46 region

was noted. On aspiration, blood-tinged fluid was obtained (Fig. 4).

Orthopantomograph (OPG) revealed a multilocular radiolucency involving the mandible extending from the 41 to 46 region, with destruction and thinning of the lower border of the mandible (Fig.5). Cone Beam Computed Tomography (CBCT) of mandible and 3-D reconstruction CT images also revealed a large osteolytic lesion involving the body of mandible, extending from the 41 to 46 region. The tumor mass was seen mostly extending toward the labial aspect causing marked expansion and destruction of both the labial and lingual cortices (Fig.6, 7).

Considering the history, clinical and radiological findings, a provisional diagnosis of ameloblastoma or odontogenic keratocyst was made. Differential diagnoses included odontogenic myxoma, central giant cell granuloma, neurofibroma,central hemangioma etc.

An incisional biopsy was performed under local anesthesia. Microscopic features of the H & E stained section showed solid epithelial cell nests with peripheral palisading columnar cells and central stellate reticulum like cells (Fig.8a,b,c). Many cell nests also showed squamous differentiation with well-formed keratin production (Fig.8d). Focal areas of cystic changes were also seen. No features of malignancy were seen. According to clinicopatholgical correlation, the features were corroborative to acanthomatous ameloblastoma.

On the basis of above diagnosis the patient was referred to the department of oral and maxillofacial surgery for further management and treatment.

DISCUSSION

An ameloblastoma is a true neoplasm of enamel organ type tissue which does not undergo differentiation to the point of enamel formation.[7] It accounts for 1% of all the tumors of the jaw encountered from the third to the fifth decades of life, with no significant sex predilection.[3] Acanthomatous ameloblastoma occurs in older patients, rather than in the younger population. The acanthomatous type of ameloblastoma is considered to be rare, with more involvement of the mandible (81%) than the maxilla (19%).[8] In the present case, acanthomatous ameloblastoma was seen in a 60-year-old male patient involving the anterior mandible, extending to the body.

Patients having this neoplasm usually presented with a slow-growing mass, malocclusion, loose teeth, pain or more rarely paresthesia, however, many lesions are detected incidentally on radiographic studies in asymptomatic patients. The lesions usually progress slowly, but are locally invasive and will infiltrate through the medullary spaces and can erode cortical bone. If left untreated, they can resorb the

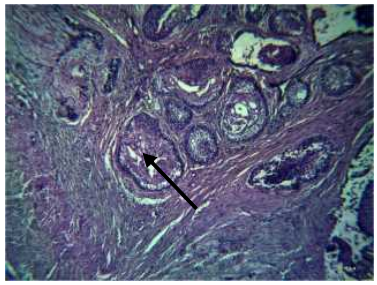

Fig. 8a : Low power photomicrograph showing islands of proliferating odontogenic epithelium

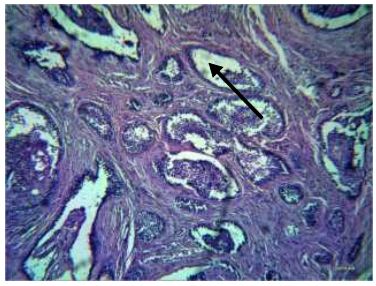

Fig. 8b : Low power photomicrograph showing proliferating odontogenic epithelium having cystic changes

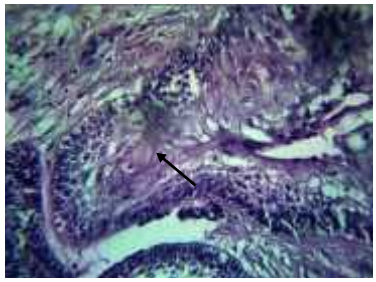

Fig. 8c : High power photomicrograph showing squamous metaplasia within the islands of proliferating odontogenic epithelium

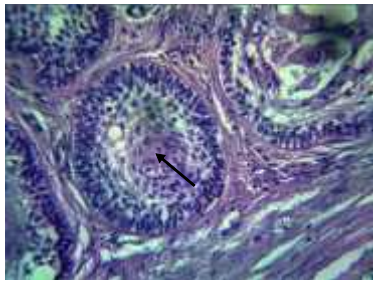

Fig. 8d : High power photomicrograph showing islands of odontogenic epithelium with keratin formation

cortical plate and extend into adjacent tissue1. Here in this case, the patient reported with a slowly progressive swelling and difficulty in mastication.

Radiographically, ameloblastomas appear as radiolucent lesions that may either have a multilocular or unilocular, soap bubble / honeycomb type of appearance.[1] In later stages, they may expand the cortical plate giving rise to a paper thin appearance on a panoramic radiograph as well as on Cone Beam Computed Tomography (CBCT).[9] These characteristic radiographic features were also evident in our case.

Amongst the six histological subtypes follicular ameloblastoma is the most prevalent histological variant (64.9%) followed by the plexiform (13.0%), desmoplastic (5.2%), and acanthomatous (3.9%) varieties. The histopathological features of an acanthomatous ameloblastoma include cells occupying the position of stellate reticulum, undergoing squamous metaplasia, sometimes with keratin formation in the central portion of the tumor islands.[10] In the present case, histopathological evaluation revealed the presence of islands of proliferating odontogenic epithelium containing a core of loosely arranged angular cells resembling stellate-reticulum, surrounded by a single layer of tall columnar cells with nuclei showing reverse polarity, associated with squamous metaplasia and keratin production. Special investigation like immunohistochemical analysis was not performed in this present case as the clinical, radiological and histopathological findings were corroborative to the diagnosis of acanthomatous ameloblastoma. Therefore, a final diagnosis of acanthomatous ameloblastoma was made on the basis of clinical, radiological and histopathological parameters.

The treatment of choice is complete surgical resection.[10] In our case, the patient was also advised for surgical resection and reported to the Oral Surgery Department for further management & treatment.

SUMMARY AND CONCLUSION

Ameloblastomas are uncommon benign odontogenic neoplasms that rarely become malignant, accounting for less than 1% of all ameloblastomas. Careful clinical examination combined with thorough imaging modalities to evaluate the general aspects of the lesions and the margins, as well as its internal architecture and its relationship to adjacent anatomical structures are very important for proper diagnosis. These informations coupled with histopathological confirmation of the diagnosis will allow for the selection of the best individual therapeutic approaches, increasing the treatment efficacy in patients diagnosed with this tumor. Thus, the

clinicopathological, diagnostic and treatment modalities of acanthomatous ameloblastoma, in general and a lesion involving the mandible, in particular is discussed herewith.

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