CASE REPORT

Ameloblastic Carcinoma of Anterior Mandible – A Case Report

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ABSTRACT

Ameloblastic carcinoma is a rare tumor arising from remnants of the dental lamina, enamel organ, and epithelial lining of odontogenic cysts or basal cells of the epithelium. It is an aggressive neoplasm that is locally invasive with an incidence of <1%. The present case of ameloblastic carcinoma arises in the anterior mandible region in a 69-year-old male patient. Marginal mandibulectomy of the lesion was done for the patient. The patient has been on regular follow-up for the past 2 years with no signs of recurrence or metastasis.

Keywords: Ameloblastomic carcinoma, Malignant ameloblastoma, Odontogenic tumours.

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INTRODUCTION

Ameloblastic carcinoma arise from remnants of odontogenic epithelium. They can also arise as secondary deposits of primary tumors from other parts of body intraosseous carcinomas in jaws are a rare occurrence. Ameloblastic carcinoma demonstrates greater cytological atypia and mitotic activity than ameloblastoma. It is an aggressive neoplasm that is locally invasive and can spread to regional lymph nodes or distant sites such as lung and bones. Treatment is similar to squamous cell carcinoma, but the prognosis is poor.

CASE DESCRIPTION

A 69-year-old male patient came to the Department of Oral and Maxillofacial Surgery, MES Dental College, Malappuram, with a large single swelling in the anterior aspect of the mandible. The patient gives a history of small swelling 8 months back which increased size rapidly 3 months back. The patient also gives a history of loose teeth in the anterior mandible, which got extracted 1 month back at a private dental clinic.

On extraoral examination a gross asymmetry of the face due to swelling over anterior mandible 4 cm from midline bilaterally and 5 cm superoinferiorly from the line joining commissures of lip till chin and it was extending 1 cm inferior to chin also [Figure 1]. Margins of the swelling were indistinct. On palpation, the swelling was firm, non-tender, and no regional lymph node involvement.

On intraoral examination a smooth sessile exophytic growth with the spherical shape of size 4 cm × 4 cm, was seen which completely obliterated the labial vestibule and extending from 45 to 34, extending into the floor of the mouth and infiltrating the lingual frenulum. On palpation, swelling was non-tender, hard, and non-fluctuant. Edges of swelling were indistinct.

On radiographic evaluation, orthopantomograph revealed unilocular radiolucent lesion in the anterior mandible with smooth borders extending between right and left mental foramen, inferiorly extending up to the inferior border of the mandible [Figure 2]. Based on the clinical and radiographic assessment, provisional diagnosis of ameloblastoma was considered. Differential diagnosis was ameloblastic carcinoma, odontogenic myxoma, and odontogenic keratocyst. Incisional biopsy was done under local anesthesia which came out as ameloblastoma.

Marginal mandibulectomy of anterior mandible was planned. The incision was placed from right to left 1st molar region; the lesion was exposed with careful dissection. The entire lesion was resected along with normal 1 cm margin leaving an inferior border of mandible intact [Figure 3]. Gross specimen was pale brown irregular tissue bit measuring 5 cm × 3.5 cm × 2.5 cm [Figure 4]. A pale brown to the dark brown nodular lesion was noted at the raw surface measuring 2.5 cm × 2 cm × 2 cm. The cut bone surface was smoothened, and closure was achieved. The patient is on regular follow-up for the past 2 years with no signs of recurrence or metastasis.

DISCUSSION

Ameloblastomas are benign odontogenic tumor arising from remnants of the dental lamina, enamel organ, and

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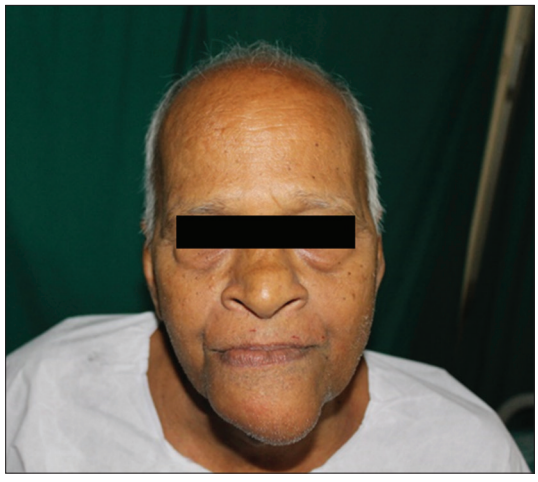

Figure 1: Extraoral swelling over anterior mandible

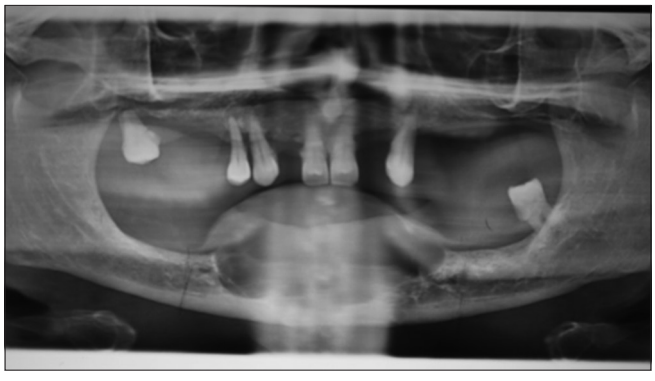

Figure 2: OPG showing radiolucency in midline

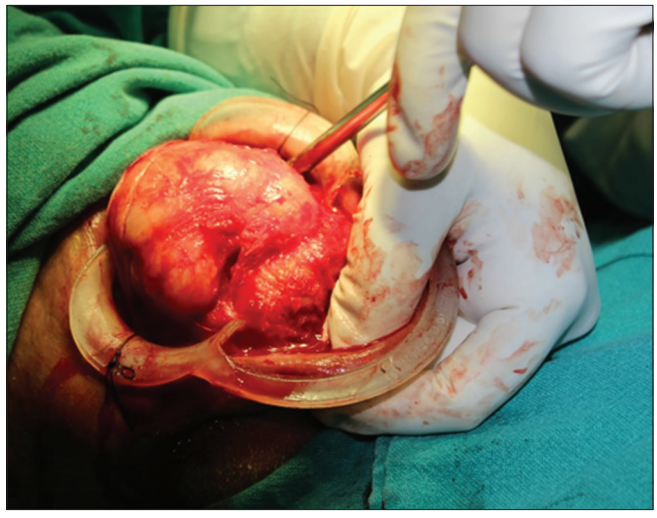

Figure 3: Tumour exposed via crestal incision

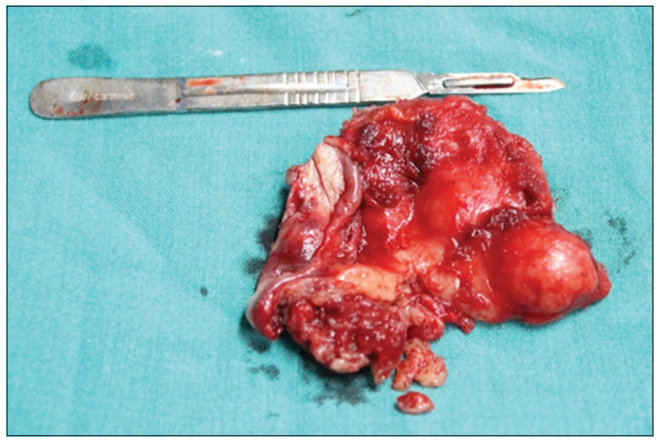

Figure 4: Gross specimen of resected tumour

epithelial lining of odontogenic cysts or basal cells of the epithelium. In 1983, Shafer introduced the term "ameloblastic carcinoma" to describe ameloblastomas in which there had been histologic malignant transformation.[1] A meloblastic carcinoma is a very rare tumor, and its incidence is <1%.[2] It can either be primary type arising *de novo* or secondary type where it arises from pre-existing ameloblastoma. There is another entity called metastasizing ameloblastoma which shows histologic features of classic ameloblastoma and has metastatic deposits elsewhere.

One of the earliest reviews on ameloblastic carcinomas was performed by Slootweg and Muller in 1984, who reviewed 42 cases and reported two of their own.[3] There are about 92 cases reported from 1984 to 2012 in the scientific literature.[4]

The age of presentation ranges from 7 to 91 years. Males are more frequently affected with male:female ratio of 2.3:1.[4] The mandible is more commonly affected than the maxilla. Our present case was consistent with the literature. Both in maxilla and mandible posterior region were favored than anterior region. In contrast to the literature, the anterior mandible was involved in our report, which is again a rare feature. Cases have also been reported in the maxillary sinus, anterior skull base, temporomandibular joint, etc. Ameloblastic carcinomas are more aggressive than ameloblastoma. Pain, swelling, paraesthesia, and ulcerations, are commonly seen followed by facial asymmetry, tooth mobility, trismus, dysphagia, cortical plate perforation, etc. Rapid growth and perforation of the cortex are characteristics of this lesion.[5]

In the radiographic examination, features were similar to solid/multicystic ameloblastoma and appeared usually as an ill-defined radiolucent lesion, either unilocular or multilocular.[6] Dispersed radio opacity was observed, which is due to dystrophic mineralization which is not seen in ameloblastoma. The affected teeth show displacement, root resorption, loss of lamina dura, and displace normal anatomic boundaries such as the floor of the nose, maxillary sinus, and mandibular canal.[7] In our case, also the patient had a history of tooth loosening slight marginal sclerosis without periosteal new bone formation is also usually noted.

Histologically, it resembles ameloblastoma but shows cellular pleomorphism, nuclear hyperchromatism, mitosis, cellular atypia vascular, and neural

invasion of tumor cells. These features were clearly observed in our case.

Ameloblastic carcinoma usually metastasis to lung, other sites include lymph nodes, brain, and bone. Multiple sites can also be involved; hence, assessment of these sites is mandatory.

Surgical resection is the treatment of choice. *En bloc* removal with 1–2 cm of normal bone margin is the safest surgical modality to ensure disease-free survival.[8] Same treatment modality was employed in our case also. This method has resulted in local recurrence rates of <15%.[9] In maxilla surgical resection is still difficult as the growth is rapid and its extension to the maxillary sinus, orbit, and other vital structures. Neck dissection should be carried out if there are signs of lymph node involvement. The efficacy of post-treatment chemotherapy and radiotherapy is not clear since there is no evidence of definitive success after these modalities.[10] However, radiation therapy and chemotherapy should be considered if it is a locally invasive lesion or metastatic lesion not amenable to surgical resection.[11]

AC can recur locally 0.5–11 years after definitive therapy. Distant metastasis may appear 4–12 years postoperatively and is common in lung, bone, liver, and brain.[12] Post-operative reconstruction should be delayed up to 2 years to account for the extremely rapid rates of recurrence.[6] The present case had a follow-up of 2 years, which showed no signs of recurrence. Periodic assessment with a follow-up period of 10 years is mandatory.

To conclude, cases of ameloblastoma should be carefully studied regarding the changes in growth pattern to detect any malignant transformation. Even though ameloblastic carcinoma is rare tumor, we should always rule out the chance of it, if a patient comes with loose tooth associated with rapidly growing and persistent jaw swellings. Complete history, radiographic, and histopathological evaluation are a must for all jaw tumors with suspicion of malignancy. Since there is a chance of metastasis for this carcinoma, we should consider the general system examination to rule out the same.

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