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**EDİTÖRE MEKTUP / LETTER TO THE EDITOR**

# Recurrent ameloblastoma of anterior mandible

## Ön mandibula'nın tekrarlayan ameloblastomu

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To the Editor,

Recurrent tumours present an important challenge to the surgeon in terms of management. Among odontogenic tumours, ameloblastoma has a high recurrence rate. Management of ameloblastoma of the mandible is therefore controversial. Though the treatment of choice is surgical excision; curettage, enucleation, marsupialisation, marginal resection and en bloc resection have been used [1]. However, the extent of surgery is determined by the age of patient and the location, size and variant of the tumour. Treatment must, therefore, remove the tumour completely and, at the same time, cause least possible morbidity [1].

First described by Cusack in 1827, the term 'ameloblastoma' is derived from the old French word 'amel' meaning enamel, and the Greek word 'blastos', meaning germ or bud [2]. Ameloblastoma is an epithelial odontogenic neoplasm that is non-mineralized. It is usually benign but locally aggressive in nature [3]. Recurrence rates vary with the treatment modality with around 15% reported for marginal resection. Recurrence has been reported as much as 16 years after surgical treatment [4]. The present report discusses recurrent ameloblastoma in the anterior mandible following surgical excision.

A 54-years-old female reported with a complaint of painless swelling in the lower jaw since 1 year. Patient first noticed the swelling 1 year back which gradually progressed to the present size. There was no history of pus discharge or paresthesia. Patient gave history of similar swelling in the same region 5 years ago, which was surgically treated by curettage and excision and diagnosed as ameloblastoma. On extraoral examination a diffuse swelling measuring around 5x3 cms in size was present in the symphysis region of mandible. Skin over the swelling was normal with no secondary changes. Superiorly the swelling extended 1 cm below the lower lip and inferiorly it was 2 centimeters below the lower border of mandible. Laterally swelling was in line with the angle of the mouth on both the sides (Figure 1A). On palpation, it was bony hard in consistency and non tender. Intraoral examination revealed a smooth surfaced diffuse swelling on the lingual aspect of the mandible measuring approximately 6x8 centimeters in size with obliteration of lower labial sulcus. Bilaterally it extended to the mandibular molar region. The swelling was smooth, bony hard in consistency and non-tender (Figure 1B). Mandibular anteriors and premolars showed delayed response on electric pulp vitality test. Based on the history and clinical features, a provisional diagnosis of recurrent ameloblastoma of anterior mandible was given. Differential diagnosis of central giant cell granuloma and adenomatoid odontogenic tumour was considered. Orthopantomograph showed multilocular radiolucency measuring approximately 5x8 centimetres in size extending superiorly from the crest of the alveolar ridge up to inferior border of

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mandible and mediolaterally from the right mandibular first molar to the left mandibular first molar region. The radiolucency gave a characteristic soap bubble appearance. Thinning and perforation of the inferior border of mandible was seen along with displacement of teeth associated with the radiolucency (Figure 1C).

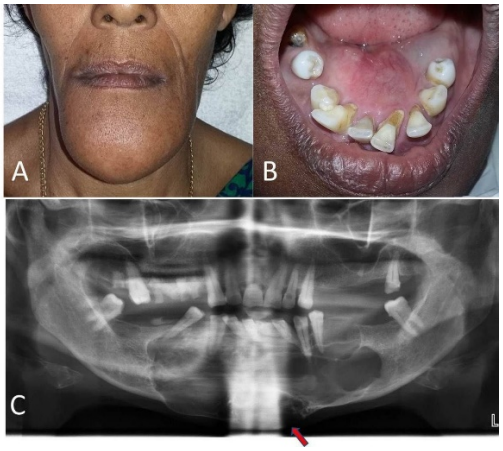


**Figure 1A. Clinical photograph showing swelling in the anterior mandibular region. 1B Intraoral photograph showing swelling in the anterior mandibular region extending to the molar region bilaterally.1C Panoramic radiograph showing a multilocular radiolucency with typical soap bubble appearance and perforation of the lower border of the mandible (red arrow).**

Computed tomography showed perforation of the lingual cortex of mandible. (Figure 2A) The 3-D reconstruction showed multiple septations in symphysis area with buccal cortical perforation and displaced lower anteriors (Figure 2B). Wide surgical excision followed by reconstruction with free fibular graft was done (Figure 3A and 3B). Histopathological examination of H & E stained sections showed odontogenic epithelial islands in connective tissue stroma. Odontogenic islands had peripheral columnar cells with hyperchromatic nuclei with reverse polarity. Inner/central cells showed squamous metaplasia with few keratin pearls. Connective tissue had collagen fibres, fibroblasts and chronic inflammatory infiltrate like lymphocytes and plasma cells. Based on the above features, a diagnosis of acanthomatous ameloblastoma was given (Figure 4). The patient is on regular follow up.

Ameloblastoma is usually unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent.[5] Ameloblastoma was initially considered to be a type of odontogenic cyst. It was first described in a case report by Cusak in 1827. In 1885, the term 'Adamantinoma' was given by Malassez. The name 'Ameloblastoma' was suggested in 1960 by Ivy & Churchill [3].

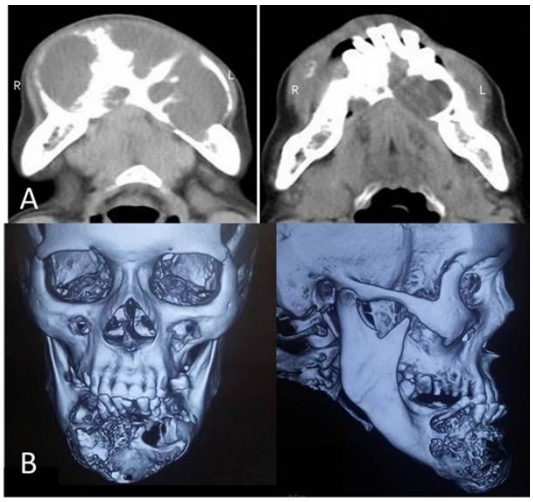


**Figure 2A. Computed tomography- axial sections showing expansion, thinning and perforation of the labial cortex and perforation of the lingual cortex. 2B 3D CT showing multiple septations in symphysis area with buccal cortical perforation.**

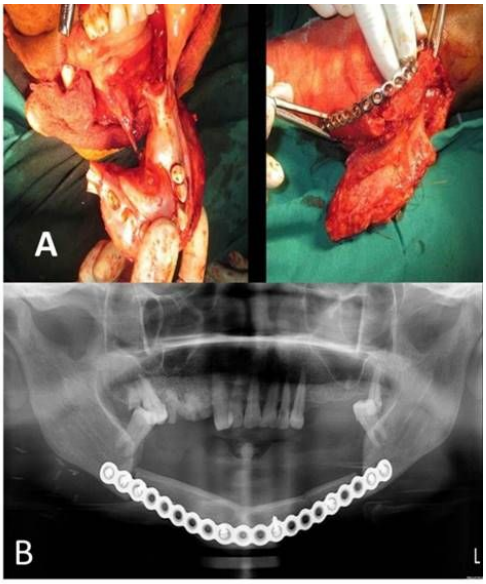


**Figure 3A. Surgical photograph showing wide excision followed by reconstruction with free fibular graft. 3B Post- operative panoramic radiograph of the patient.**

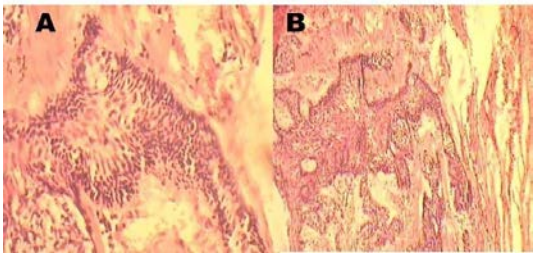


**Figure 4(A)40x and (B)10x showing peripheral columnar cells with hyperchromatic nuclei with reverse polarity and central cells with squamous metaplasia, keratin formation and few keratin pearls. Features were suggestive of acanthomatous ameloblastoma.**

Ameloblastoma arises from dental epithelium and is characterized by its histological resemblance to the enamel organ of the developing tooth [6].The tumour is believed to arise from either the cell rests of the enamel organ, epithelium of odontogenic cysts, disturbances of the developing enamel organ, basal cells of the surface epithelium or from heterotropic epithelium in other parts of the body [7].

Ameloblastoma of the jaws is the most commonly seen odontogenic tumour in Africa and Asia and the second most common in North and South America [3].Ameloblastomas account for 1% of all jaw tumors encountered in an individual during 3rd to 5th decade. Eighty percent lesions occur in the mandible of which 70% are seen in the molar region, ascending ramus and angle of the mandible; 20% are seen in the canine region and only 10% occur in the incisor or anterior region. Present case occurred in the anterior mandible which is a less common site for an ameloblastoma [6].

Ameloblastomas are classified as unicystic, multicystic, peripheral and malignant subtypes. Conventional or multicystic variant constitutes about 86% of cases, unicystic type comprises 13% of cases and peripheral or extra-osseous type is the rarest with only about 1% of cases [8].Clinical presentation includes a slow growing, painless swelling causing cortical bone expansion. This may lead to perforation of the cortical plates and infiltration into the soft tissue. Mandibular lesions may present with paresthesia [9]. In our case, the patient presented with a slow growing, non-tender swelling causing expansion of bone and displacement of the adjacent teeth.

Radiographic features of a multicystic ameloblastoma include a radiolucent, expansile, multiloculated cystic lesion with scalloped borders. The locules are described as characteristic "soap bubble" or "honey comb" appearance. Unicystic ameloblastoma typically presents as a unilocular radiolucency usually associated with an impacted tooth. Resorption of the roots of adjacent teeth is a common feature.[9]Soap bubble appearance, expanded cortices with labial and lingual perforation and displacement of teeth were seen in our case. The chief histopathological variants of ameloblastoma are the follicular and plexiform types. The uncommon variants include acanthomatous, desmoplastic, basal cell, clear cell and granular cell types. Follicular ameloblastoma is the commonest accounting for 64.9% cases. The acanthomatous variant is the rarest with a prevalence of only 3.9%. Our case was diagnosed as acanthomatous ameloblastoma. In acanthomatous ameloblastoma squamous cell metaplasia of the odontogenic epithelium can simulate the appearance of both squamous cell carcinoma and basal cell carcinoma [6].

Management includes segmental resection including the periosteum and overlying soft tissues in case of extensive lesions. Bone grafts can be used to repair the defect[3]. Few authors have advocated different surgical modalities based on the histological type of ameloblastoma [4]. The follicular, granular cell and acanthomatous types have a relatively high recurrence [4]. A recurrence rate of 60-80 % is frequently seen after simple treatment with enucleation or curettage. In the present case, the recurrence occurred following surgical excision and curettage. Marginal resection is still widely used for treating small multicystic ameloblastoma but due to a recurrence rate of around 15 %, it is recommended to keep the safety margin at least 1 cm beyond the radiographic margin. Despite this, studies have reported an overall recurrence rate of around 22% [8].Among these, more than 50 % of recurrences happen within the first five years following treatment [4].Therefore a long term follow up is recommended.

To conclude, ameloblastoma is an odontogenic tumour with high recurrence rate in conservatively managed cases. Recurrences are highest for treatment with curettage and in solid or multicystic lesions. This implies that such cases should be treated with adequate margin of normal bone. Conservative management should be restricted to unicystic ameloblastomas. Also, due to its slow growth, recurrences can occur many years after treatment.

Thus, patients should be followed up for long term.

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