AMELOBLASTIC CARCINOMA. A CASE REPORT
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ABSTRACT: Ameloblastoma of the jaw is a rare tumor that stems from dental embryonic remnants in the mandible or maxilla. It grows slowly, without clinical signs in the early stages. Although it is considered as a benign tumor, it does invade locally and sometimes metastasizes. Lungs, liver and kidneys are the common sites for metastasis. Metastasizing ameloblastoma is designated as malignant ameloblastoma. Malignant behaviour of ameloblastoma attributed to the malignant transformation of the epithelial component, called ameloblastic carcinoma, has also been reported. The differentiation between the entities malignant ameloblastoma and ameloblastic carcinoma is controversial. A case of ameloblastoma exhibiting aggressive behaviour with possible metastasis to lungs is reported along with discussion of its features.

INTRODUCTION
Ameloblastoma is a rare benign odontogenic tumor, occurring with an increased incidence in the third and fourth decades. It accounts for 1% of all oral tumors. About 80% of the cases are reported in mandible with a definite predilection for molar-ramus area. Clinically ameloblastoma is locally invasive and persistent. Although rare, ameloblastoma do undergo malignant transformation as well as metastasis. Malignant ameloblastoma and ameloblastic carcinoma, the two malignant types of ameloblastoma must be differentiated from each other. Former refers to tumors that have metastasized retaining histologic appearance of a benign lesion in both primary and metastatic sites whereas latter term is used to designate those lesions exhibiting histologic features of malignancy within a pattern of ameloblastoma. Corio et. al. defined ameloblastic carcinoma as any ameloblastoma in which there is histologic evidence of malignancy in the primary tumor or the recurrent tumor, regardless of whether it has metastasized. These lesions are usually aggressive with perforation of cortical bone and extensions into floor of the mouth and adjacent soft tissues.

CASE REPORT
A 52 year old male patient was referred to the Dept. of Oral Medicine and Radiology, College of Dental Surgery, Manipal, for evaluation of the lesion in the oral cavity. The lesion was noticed as a small nodule six years prior to the visit and it gradually increased to the present size. Growth was reported to be faster in the last six months. He was taking some indigenous medicine for the same. Patient was a chronic smoker with a history of bronchial asthma for the past five years. Family history was negative for any similar condition.

On general examination, the patient looked chronically ill and weak. Lips were incompetent and a swelling could be noticed near
the right angle of the mouth (Fig 1). Psoriasiform skin lesions were found on the hands and legs.

Intraoral examination showed poor oral hygiene with few of the upper and lower teeth remaining. A large mass of 7 x 8 cm size was observed in the lower part of the oral cavity (Fig. 2). It was extending from right vallecula and tonsils to the left lower canine region. The growth occupied the whole of right floor of the mouth, alveolar ridge, buccal sulcus and the anterior part of the oral cavity. The lesion extended into the labial sulcus pushing the lower lip anteriorly. The tongue was elevated due to the tumor mass. Overlying mucosa showed areas of ulceration due to impingement of the opposing teeth. The mass, on palpation, was firm and non-tender. Bleeding was not seen from the growth. None of the lymph nodes, in the head and neck region, were palpable. From the history and clinical examination, a provisional diagnosis of ameloblastoma was made.

Patient was subjected to roentgenographic examination which included orthopantomograph OPG and right lateral oblique radiographs. Considering the extensive nature and large size of the tumor, a chest x-ray was also advised. OPG showed a multilocular radiolucency extending from right molar region to left canine region. It also showed two embedded teeth in the radiolucent area. The thickness of the mandible was reduced considerably due to the destructive lesion, however, no pathologic fracture was noticed (Fig. 3). Lateral oblique radiograph of the right side revealed similar findings. Chest x-ray showed a large homogeneous opacity over the upper lobe of right lung (Fig 4).

Bone scan with 99Tc radioisotope was done to rule out other metastatic bony lesions. It did not show any abnormal trace concentration except over right mandible.

A biopsy was taken from the oral lesion and sent for histopathological examination. But patient refused to undergo biopsy of the lung lesion. Microscopically the lesion showed tumor islands in dense connective tissue stroma arranged in the form of follicles (Fig 5). Periphery of follicles were lined by columnar cells with palisading nuclei and central cells exhibited stellate reticulum like arrangement. The central cells in some follicles showed squamous metaplasia and keratin pearl formation. Peripheral as well as central follicular cells showed marked pleomorphism, altered nuclear cytoplasmic ratio, prominent nucleoli and few mitotic figures (Fig 6). Over lying epithelium was hyperparakeratinized stratified squamous atrophic to acanthotic. The features were suggestive of ameloblastic carcinoma. Patient was then referred to Dept. of Oncology, Kasturba Hospital, Manipal for further management.

DISCUSSION

Ameloblastoma, the term was coined by Ivy and Churchill in 19307, is considered to be arising from the odontogenic apparatus in the jaws. Dental epithelium, enamel organ, remnants of dental lamina, the rests of malassez and epithelial lining of odontogenic cysts are all implicated with the origin of this tumor8. The ameloblastoma is characteristically slow growing, seldom with a complaint of pain or discomfort other than the presence of a swelling. This results in the patient approaching for treatment years after noticing the lesion. In the present case also, the patient had the swelling for six years. Although there was a gradual increase in size of the swelling, absence of any discomfort led to the neglect, allowing the growth to reach the present size. Ameloblastoma, in mandible, is quite often associated with impacted teeth2, as in our case where two impacted premolars were present within the tumor.

Both malignant ameloblastoma and ameloblastic carcinoma show metastasis and the most frequent site is lungs, followed by liver and kidney9. Some cases of single deposits in lungs can be explained by local extension and
Fig. 1: Lateral profile view showing protrusive lips

Fig. 2: Intra-oral view showing the lesion

Fig. 3: OPG revealing impacted premolars in a multilocular radiolucency

Fig. 4: Chest x-ray with a large opacity over right lung

Fig. 5: Photomicrograph of H&E stained section showing follicles characteristic of ameloblastoma (x10)

Fig. 6: Areas showing cellular pleomorphism, prominent nucleoli and altered nuclear cytoplasmic ratio (x 40)
aspiration. In this case, the patient had a lesion in the upper lobe of right lung, which could not be confirmed histologically as a metastatic lesion. However the clinical features and extent of the primary tumor along with the position and characteristics of the lung lesion point towards a possible metastatic lesion.

The microscopic pattern of tumor islands arising as follicles in a dense connective tissue stroma is typical of follicular ameloblastoma. The features like pleomorphism, altered nuclear cytoplasmic ratio, vesicular nuclei with prominent nucleoli seen among the peripheral columnar and central stellate cells are histologic evidence of malignancy. The above picture correlates with that of ameloblastic carcinoma, where a combination of features of ameloblastoma occur with a less-differentiated morphology.

Treatment depends on the extent and location of the lesion, type of ameloblastoma, soft tissue involvement, age and condition of the patient. The various treatment modalities used are surgery, radiotherapy, cauterity, cryotherapy etc. The ability to predict behaviour, based on clinical, radiographic and microscopic features is paramount to appropriate treatment for avoiding recurrence and undue morbidity. However, regardless of the type of treatment, long term periodic follow-up for recurrence is of utmost importance.

REFERENCES


