Follicular Ameloblastoma of maxilla: A case report and review

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Abstract:

A 62-year-old male patient had been reported with complaint of swelling on the left side of the face for last 10 years. Provisional diagnosis of ameloblastoma was given based on clinical, radiographic findings and FNAC. Post operative histopathology confirmed the diagnosis as follicular ameloblastoma. Enucleaiton of cyst with surgical obturator was performed under general anesthesia. Patient has been kept under periodic follow-up. No recurrence had been reported till date.

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Introduction:

Ameloblastoma is a rare tumour occurring in the maxilla. The first detailed description of this lesion was by Falkson in 1879, but the term 'ameloblastoma' was coined by Churchill in 1933.[1] It represents approximately 1% of oral tumours and develops from the odontogenic epithelium and its derivatives or remnants.[3] Sometimes it arises from a dentigerous cyst.[4] It occurs with a wide range of ages; a mean age in the third or fourth decade, and equal frequency between male and female,[5] although a higher frequency in females than in males has been described.[6,7] It has a slightly higher incidence in black races and in the Japanese.[8] Ameloblastoma has a persistent and slow growth, spreading into marrow spaces with pseudopods without concomitant resorption of the trabecular bone. As a result, the margins of the tumour are not clearly evident radiographically or grossly during operation, and the lesion frequently recurs after inadequate surgical removal, showing a locally malignant pattern.[9] Long term follow-up is necessary because this lesion has been shown to recur 25 and 30 years following primary treatment.[5,8] Resorption of the adjacent tooth roots is not uncommon. Several histopathologic types of ameloblastoma are described and include plexiform, follicular, unicystic, basal cell, granular cell, clear cell, acanthomatous, vascular and desmoplastic patterns.[1,3,10] Ameloblastomas rarely metastasize.[11] The most common sites of metastases are the lungs followed by regional lymph nodes, pleura, vertebrae, skull, diaphragm, liver, parotid and small intestine.[13]

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Case report:

A 62 year old man was referred to the Oral and Maxillofacial Surgery Department of The Anwer Khan Modern Hospital, Dhaka, Bangladesh, in 30th March 2015. The chief complaint was a painless and progressive swelling of the left maxilla present for ten years. The patient was apparently asymptomatic ten years back. Then he noticed a swelling on the left side of the face, which was small and painless initially and gradually increased in size. The patient was consulted at another medical college for the same complaint and an FNAC was performed from the left maxillary swelling in 17th May 2007. Histopathological report of the specimen was suggestive of odontogenic cystic lesion. There was no significant past medical history. He gave history of taking anti-hypertensive medication for last twenty years. His physical examination revealed no abnormality other than those related to the chief complaints.

On extra-oral examination, there was facial asymmetry on the left side. On palpation there was a well circumscribed, nontender, smooth surfaced swelling of hard consistency, spherical in shape and approximately 5 × 4 cm in size, present in the left maxillary region extending from midline to 5 cm anterior to the tragus. Superiorly it extended up to the infraorbital rim. Obliteration of nasolabial fold was present along with slight elevation of alar base on the left side. On superficial examination of the nostrils, the nasal floor was found to be elevated in the left nostril.

On intraoral examination (Fig. 1), expansion was present in the left maxillary vestibular area extending from midline to third molar, causing complete obliteration of vestibular fold and also extending into the palatal region from the left maxillary central incisor region to the greater palatine foramen region. Clinical examination revealed diffuse, smooth–surfaced, firm, nontender swelling on the left side of the face. It extended from the zygomatic region to the inferior border of mandible superoinferiorly, and from the corner of the mouth to the angle of mandible anteroposteriorly. It was covered with inflamed mucosa. Intraorally, the swelling extends from distal of first molar posteriorly. Swelling resulted in obliteration of the buccal vestibule.

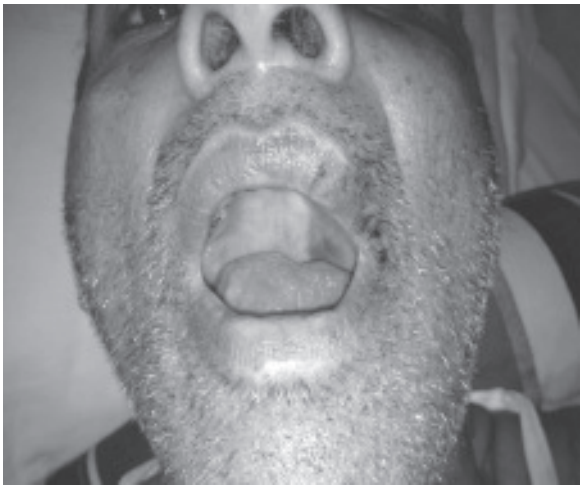

Fig.-1: *Pre-operative intra-oral photograph.*

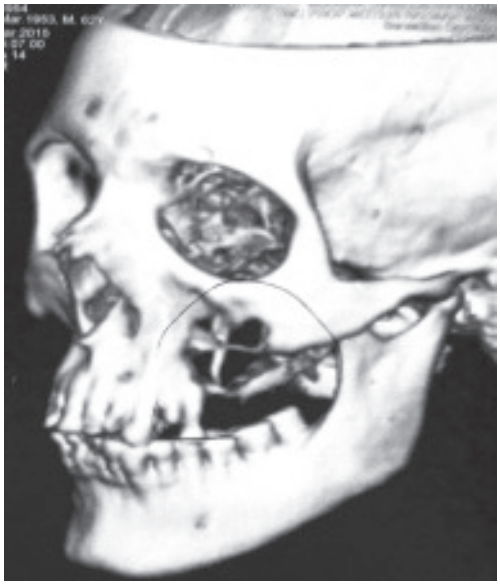

Fig.-2: *Pre-operative 3D radiographic image of affected maxilla.*

Computerized tomography (contrast enhanced) (Fig. 3, 4) showed a well-defined cystic area measuring 6.09cmX4.21cm in coronal scan and in axial scan measuring 4.46cmX 3.91cm within the maxillary antrum. The wall of the maxillary antrum are almost intact except near the alveolar process of the left maxilla where cystic lesion shows some septation suggestive of odontogenic cyst. Pre-operative histological examination showed the features of an odontogenic cyst.

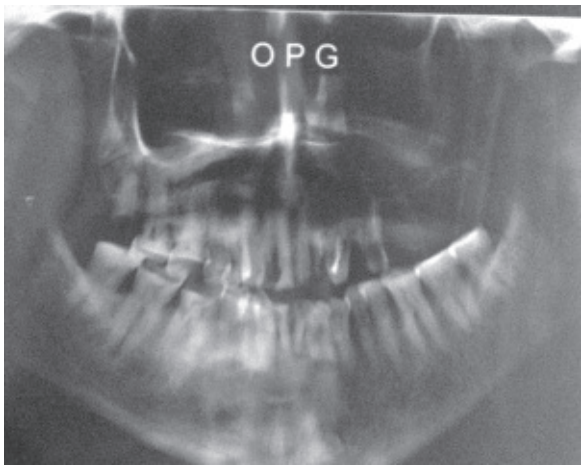

Fig.-3: *Pre-operative radiograph.*

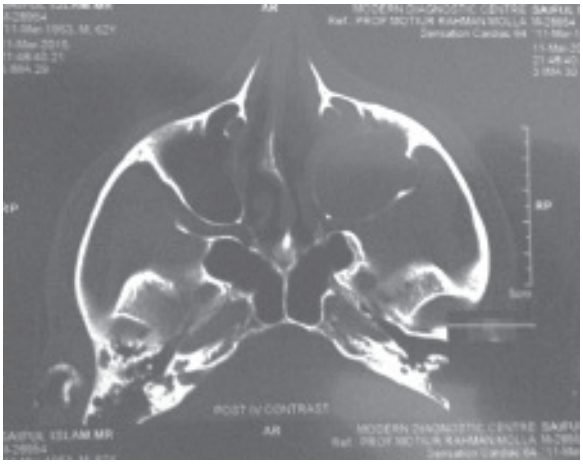

Fig.-4: *Pre-operative CT scan.*

Under general anaesthesia enucleation of cyst with surgical obturator was performed with left sided maxillary circular incision with posterior extension. Cystic lining was dissected out from buccal and palatal periosteum and whole mass was taken out in toto. Irrigation was done with betadine and normal saline. An antibiotic soaked gauze

pack was kept in antral cavity and a surgical obturator was placed in situ. The healing was uneventful. Histological examination of the excised lesion showed cystic ameloblastoma with plexiform pattern. Hemostasis was achieved; vacuum drain was secured and closure was done in layers (Fig. 5). Antibiotics, analgesics and anti-inflammatory drugs were given postoperatively. Healing was uneventful, and sutures were removed on 7th postoperative day. Patient has been kept under periodic follow-up. No recurrence had been reported till date.

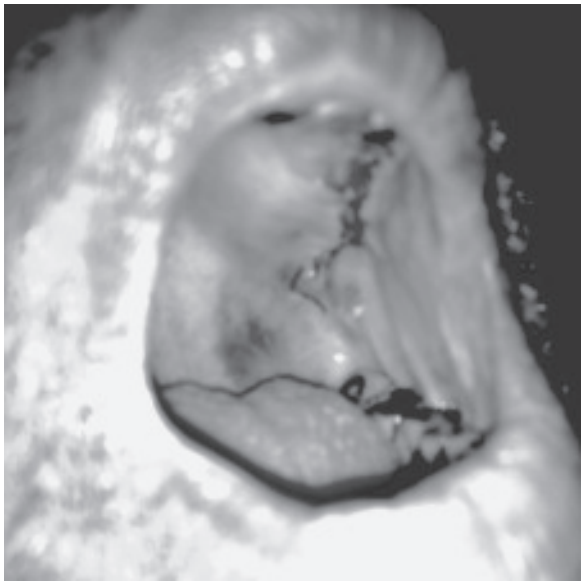

Fig.-5: *Post-operative photograph (intra-oral).*

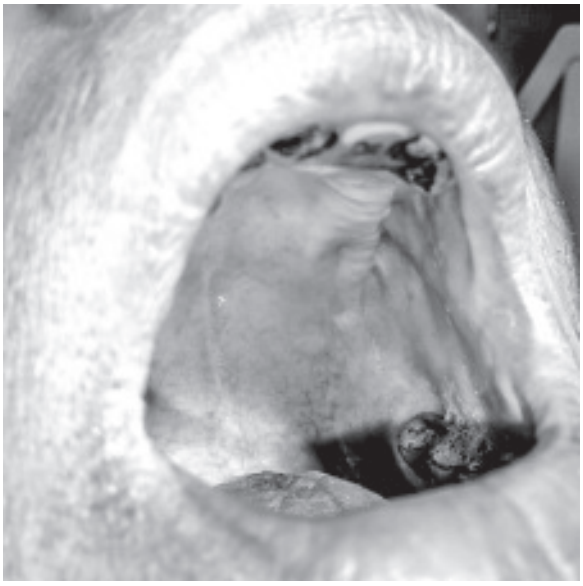

Fig.-6: *Post-operative photograph (intra-oral).*

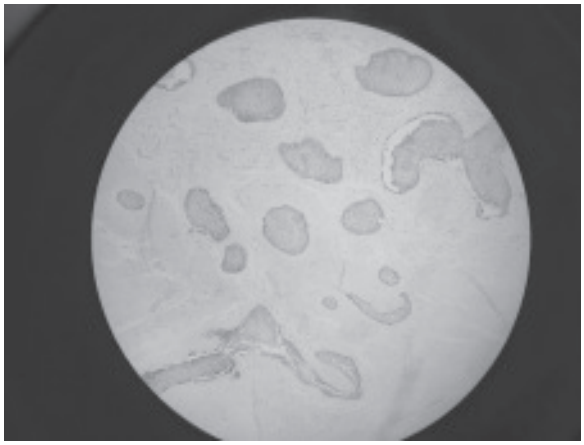

Fig.-7: *10x modification.*

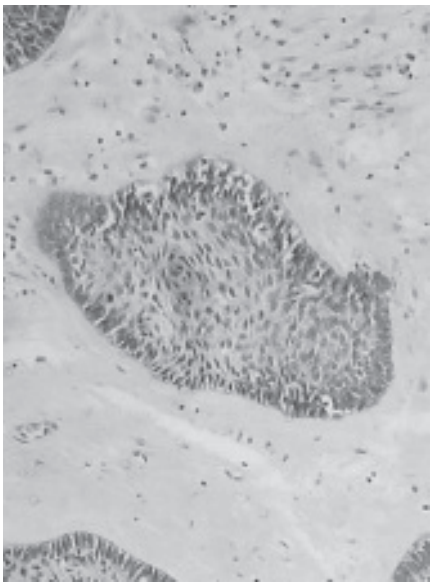

Fig.-8: *100x Modification.*

Discussion:

It is generally accepted that only 20 percent of ameloblastomas occur in the maxilla,[2] although some reports indicate an incidence as low as one percent in the maxilla,[14] and of those 47 percent occur in the molar region, 15 percent in the antrum and the floor of the nose, 9 percent in the premolar areas, 9 percent in the canine regions and 2 percent in the palate.[2] Since maxillary ameloblastoma has a predominantly painless and slow growth because of the lack of a thick cortical plate, the plentiful cancellous bone and the proximity of the maxilla to the nasal cavity, nasopharynx, paranasal sinuses, orbits and skull base, there is commonly a delay in the recognition of the maxillary ameloblastoma extending into these structures and this itself may provide useful diagnostic evidence.[15] In this case the ameloblastoma extended into the left maxillary

antrum up to the lower concha of the nose and the inferior orbital wall without penetrating these structures. In addition, the more abundant blood supply of the maxilla provides another possible mode of spread. Sometimes invasive maxillary ameloblastomas with extension into the orbit, frontal sinus, skull base, middle cranial fossa and petrous apex have resulted in the death of the patient.[16] The most common clinical symptom of the maxillary ameloblastoma is a painless swelling of the involved part of the jaw. Pain is an uncommon finding,[1] referred in some cases,[7] but it is not clear whether the pain is caused by the tumour itself or by a secondary infection. Ameloblastoma is an osteolytic lesion and does not produce mineralized components except in rare cases.[10,17] As far as etiology is concerned, it may arise from the enamel organ, remnants of dental lamina, the lining of an odontogenic (dentigerous) cyst, or possibly from the basal epithelial cells of the oral mucosa.[18] A few studies also showed that the human papillomavirus might have a role in the etiology of ameloblastoma.[19] About 10-15% of ameloblastomas are associated with an unerupted tooth.[20] In the present case, a large follicular ameloblastoma was found in left maxilla. The diagnosis of follicular ameloblastoma was confirmed by excisional biopsy. Out of all the histologic variants of ameloblastoma, the incidence of plexiform variety is one-third. The term "plexiform" depicts the appearance of anastomosing islands of odontogenic epithelium in contrast to a follicular pattern.[21] A number of modalities have been proposed in the treatment of ameloblastoma, like wide excision, curettage, enucleation, cryotherapy, cautery, laser usage, radiotherapy and chemotherapy.[22] The best surgical method for the treatment of a maxillary ameloblastoma is a limited or wide excision of the tumour with a 10-15 mm margin of normal bone if available.[9] Enucleation of the cyst with surgical obturator with was the method of choice in this case. Ameloblastoma is generally considered to be a radioresistant tumour and may be performed in cases when surgery is not considered to be the method of choice.[23] Chemotherapy, when used independently, does not seem to be effective at the present time, notwithstanding the variety of agents, schedules and routes of administration that have been reported.[24]

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