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CASE REPORT

Maxillary ameloblastoma extending into the maxillary sinus

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ABSTRACT

Ameloblastoma is a benign but locally aggressive odontogenic tumor. Worldwide, maxillary ameloblastoma is rare, but its late detection renders adequate treatment difficult. Majority occur in the mandible with about 5-20% occurring in the maxillary bone. Here we report a case of plexiform ameloblastoma of the left maxilla in a 30-year-old male. The tumor was presented as a radiographically solid mass filling the left maxillary sinus and clinically as a maxillary swelling. The radio-pathological features of this tumor and the possibility of its sinonasal epithelium origin are discussed.

Key words
Ameloblastoma, maxillary sinus, odontogenic epithelium

INTRODUCTION

Ameloblastoma is a benign, locally aggressive jaw tumors with a high propensity of recurrence derived from intra-osseous remnants of odontogenic epithelium, lining of odontogenic cysts and the basal layer of overlying mucosa. Suggested sources for the odontogenic epithelium include rests of dental lamina, a developing enamel organ, the lining of odontogenic cyst, basal cells of oral mucosa, or heterotrophic embryonic organ epithelium. It is the second most common odontogenic neoplasm accounting for 9% to 20% of all odontogenic tumors.[1]

The first detailed description of this lesion was by Falkson in 1879, but the term 'ameloblastoma' was coined by Churchill in 1933.[2]

The estimated incidence of ameloblastomas is approximately 0.5 per million populations per year. There is no distinct gender predilection. Most cases are diagnosed between 30 and 60 years of age.[3] The maxillary ameloblastoma occur 12 years later than that of its mandibular counterpart.[4]

Approximately 15-20% of ameloblastomas have been reported to originate in the maxilla, with just 2% arising anterior to premolars.[5-7]

The literature showed that solid ameloblastoma occurred as the least frequent in maxillary bone.[8-10]

Based on the recent Classification of Odontogenic Tumors, by World Health Organization (WHO), benign ameloblastomas are recognized in four sub-types: The solid/multicystic, the desmoplastic, the unicystic and the extra-osseous/peripheral type. Solid ameloblastomas affect the mandible preferably, especially the posterior region with a proportion between the gnathic bones of 1:5.[9,11]

Ameloblastomas may present on conventional radiographs as a unilocular or multilocular corticated radiolucency resembling a cyst. Bony septa may result in a honeycomb appearance. The lesion may remain asymptomatic before a facial swelling develops. Computed Tomography (CT) and Magnetic Resonance Image (MRI) may be helpful in establishing the extent of the lesion, particularly when located in the maxilla.[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. Long term follow-up is necessary because this lesion has been shown to recur 25 and 30 years following primary treatment.[12]

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A 30 year old male patient reported with a chief complain of pain in upper left back region and swelling in left middle region of face, since one and a half months and pus discharge while pressing the swollen parts since last 2 days.

The onset of swelling was insidious and the progression was gradual. There was dull and intermittent pain along with pus discharge. The patient reported associated symptoms of metallic taste and foul odor from mouth since one month. There was also mild pain on chewing food. He also gave a history of recurrent cold since one month. The clinical appearance of the lesion on extra oral examination revealed that there was asymmetry seen on left side of face in the malar region. Intra-orally, there was a hard swelling (2 × 3 cm) extending from left first premolar to mesial of second molar, obliterating the buccal vestibule and extending onto the palatal side adjacent to first and second molar. The color of the overlying mucosa was normal and the margins of the swelling were ill defined. The swelling was indurated and non tender but there was presence of discharge [Figure 1].

The Orthopantomograph (OPG) revealed a unilocular radioluscency extending from 24 to 27 regions with resorption of roots [Figure 2]. The para nasal sinus (PNS) view showed hypertrophy of nasal turbinates and bilateral antral mucosal thickening was seen. There was partial obliteration of left maxillary sinus [Figure 3]. CT scan was performed which showed evidences of bulging of the floor of the left maxillary sinus with erosion of dental sockets of the left maxilla associated with mucosal thickening in left maxillary sinus. Bilateral nasal turbinates showed hypertrophy associated with deviation of nasal septum towards right [Figure 4].

A provisional diagnosis of tumor with respect to left maxillary sinus, cysts arising from sinus lining and mucocele was made. Fine needle aspiration cytology (FNAC) was performed, but the aspiration yielded negative results. Incisional biopsy was performed and the biopsied specimen was sent to the Department of Oral and Maxillofacial Pathology, Saraswati Dental College and Hospital, Lucknow.

The histopathological examination revealed odontogenic epithelium proliferating in the form of sheets and follicles.The odontogenic epithelium revealed peripheral layer of tall columnar cells showing pallisaded, hyperchromatic nuclei with reversal of polarity and central cells resembling stellate reticum like tissues. The background stroma was scant and showed cystic areas of degeneration and foci of juxta epithelial hyalinization. The hard tissue examination revealed mature cancellous bone invaded by tumor cells [Figure 5].

A diagnosis of ameloblastoma of maxilla predominantly plexiform pattern extending into the maxillary sinus was given. A radical left maxillectomy was performed. A total maxillectomy of the left maxilla was done under general anaesthesia.

The resected maxillary specimen along with the mucosal

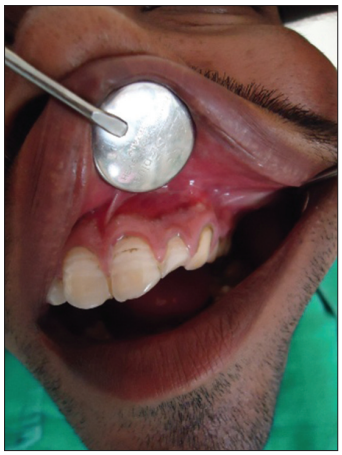

Figure 1: Shows a hard swelling (2 × 3 cm) extending from left first premolar to mesial of second molar, obliterating the buccal vestibule and extending onto the palatal side adjacent to first and second molar

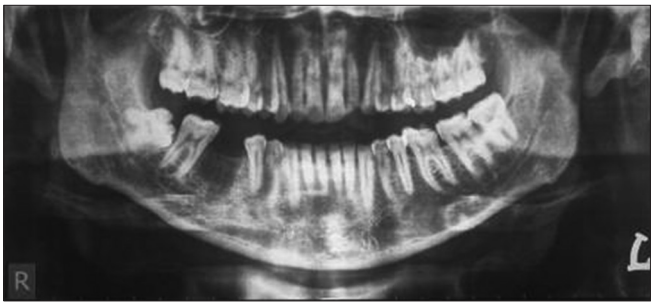

Figure 2: The OPG revealed a unilocular radioluscency extending from 24 to 27 region with resorption of roots

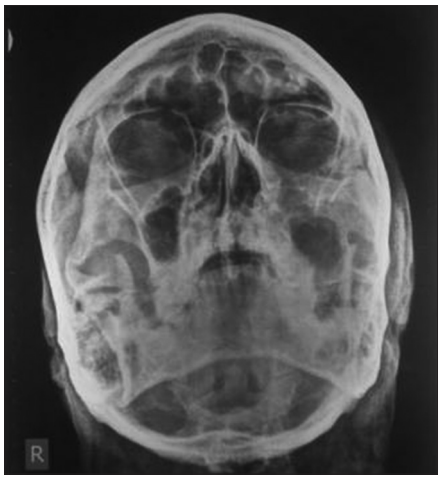

Figure 3: The PNS (Occipitometal view) showed hypertrophy of nasal turbinates and bilateral antral mucosal thickening is seen. There was partial obliteration of left maxillary sinus

lining was sent for histopathological examination which confirmed the provisional diagnosis of ameloblastoma. Tumor cells were seen close to the bony margins of maxilla [Figure 6]. These findings confirmed the provisional diagnosis of Ameloblastoma mainly plexiform type extending into the maxillary sinus.

DISCUSSION

Globally, maxillary ameloblastoma is rarer than mandibular lesions. It is generally accepted that only 20% of ameloblastomas occur in the maxilla, although some reports indicate an incidence as low as 1% in the

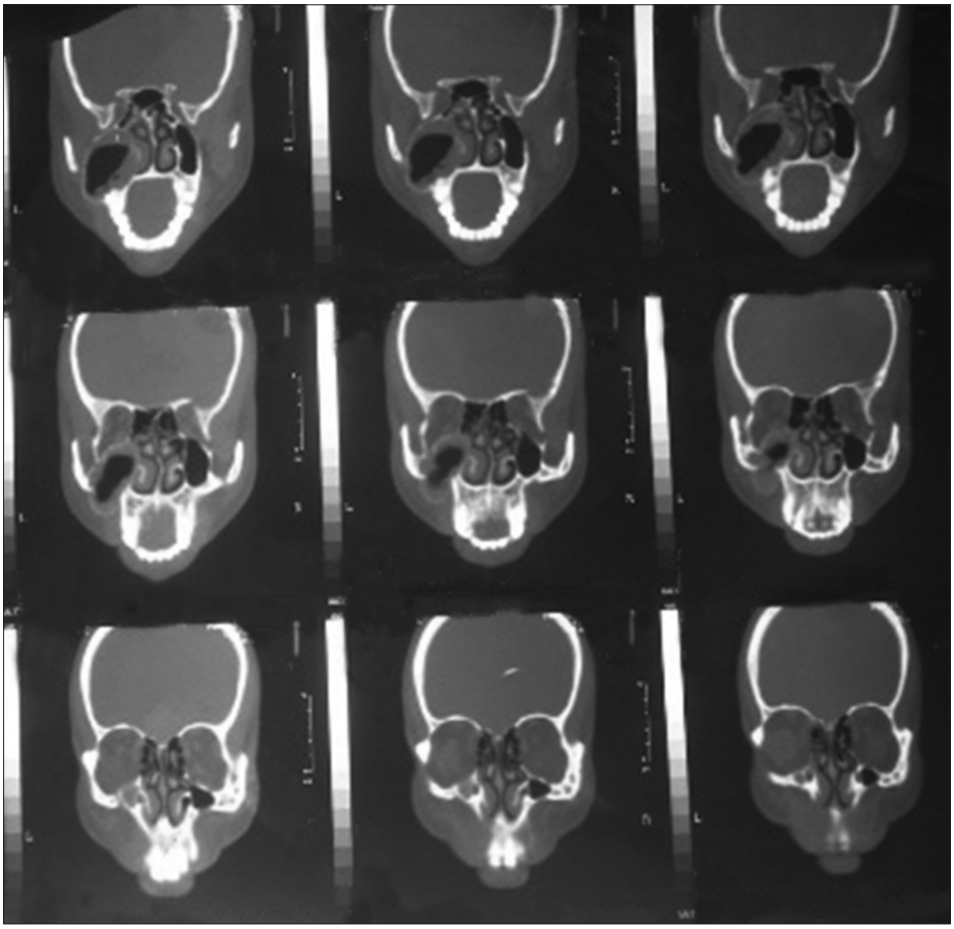

Figure 4: CT showed bulging of the floor of the left maxillary sinus with erosion of dental sockets of the left maxilla associated with mucosal thickening in left maxillary sinus

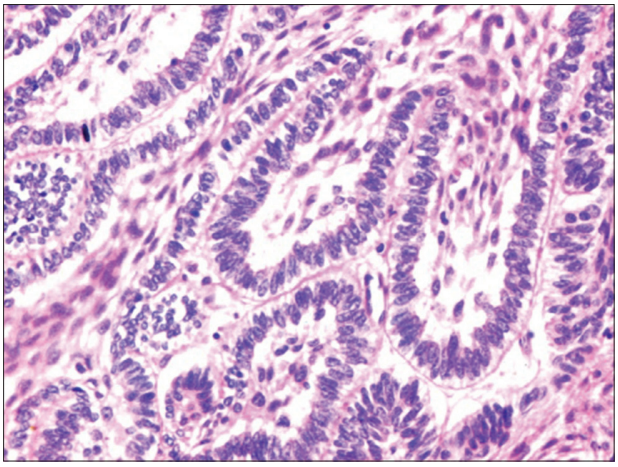

Figure 5: The odontogenic epithelium revealed peripheral layer of tall columnar cells showing pallisaded, hyperchromatic nuclei with reversal of polarity and central cells resembling stellate reticum like tissues

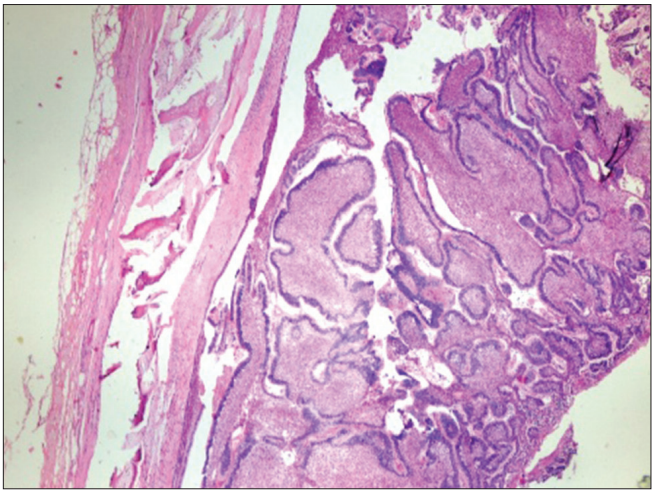

Figure 6: Tumor cells were seen close to the bony margins of maxilla with odontogenic epithelium proliferating in the form of islands and cords

maxilla and of those 47% occur in the molar region, 15% in the antrum and the floor of the nose, 9% in the premolar areas, 9% in the canine regions and 2% in the palate.[4]

In a study, 379 cases of ameloblastoma of jaw 15% had an antral localization.[13]

Ameloblastomas occur at all ages, however majority present during 3rd or 4th decades of life. However, in the maxilla, lesions are seen more in patients over a decade older.[14] However, in our case the patient was a young 30-years-old male concurrent with the study Ajike *et al.* who reported a mean age of 38.14 years for maxillary ameloblastoma.[15]

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 prove fatal in some cases.[13]

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]

According to Williams multilocular ameloblastomas have a poorer prognosis than their unilocular counterpart.[17] In our case a unilocular lesion was present which occurred in younger patients as reported earlier by Ajike SO *et al.* and was similar to our case.[15]

Pain is an uncommon finding, referred in some cases, but it is not clear whether the pain is caused by the tumour itself or by a secondary infection.[2,12]

Ameloblastomas are epithelium-derived odontogenic tumors that typically are originated in jaw bones, primarily involving the mandible and less often the maxilla. The presence of ameloblastomas ie sinonasal region is usually secondary to an extension of a tumor of gnathic origin into this area. Although hypothetical, it appears quite likely that this embryological approximation allows the sinonasal tract mucosa either to incorporate odontogenic epithelium or to acquire cells capable of odontogenesis during development.[18]

The epithelial source of origin for gnathic ameloblastomas still is being debated.[1] Although, there are extra-gnathic ameloblastomas that have been shown to arise from misplaced odontogenic rests.[19] When considering the possible histogenesis of sinonasal tract tumors, parallels can be made with peripheral ameloblastomas, which also were believed to originate outside the boundaries of odontogenic epithelium.

In a report of 24 cases of sinonasal ameloblastomas, as was previously noted, direct continuity with the sinonasal surface epithelium was identified. This finding supports a mucosal surface epithelium derivation. Perhaps the development of sinonasal tract ameloblastomas occur after some inductive process on the sinonasal epithelium that results in the neoplastic transformation of the retained or acquired odontogenic cells towards ameloblastomatous differentiation. The presence of chronic sinusitis and squamous metaplasia in the areas adjacent to the ameloblatoma could be the initiating event. Although the surface epithelium appears to represent the likely site of origin, the histogenesis for the primary sinonasal ameloblastomas remain unknown.[20]

It has been suggested that peripheral gnathic and sinonasal ameloblastoma may originate from pluripotential cells of the basal layer of the oral and sinonasal epithelium, respectively.[21] The possibility of sinonasal surface epithelium origin of the present case was considered. This direct continuity with the sinonasal surface epithelium could not be proved because there was maxillary bone involvement in our case. Although, the development of sinonasal tract ameloblastomas may be initiated after some inductive process on the sinonasal epithelium that results in the neoplastic transformation of retained odontogenic cells and leads to differentiation of ameloblastoma.[9]

Given the classical histological features of ameloblastomas, the differential diagnosis is limited. Of primary importance is to exclude extension into the sinonasal tract from a primary gnathic ameloblastomas. The only other consideration might be a craniopharyngiomas. However, the clinical features of craniopharyngiomas markedly contrast with the sinonasal tract ameloblastomas so that the lesion should be readily seperable.[22]

Nasal obstruction localized facial enlargement and swelling of the cheek, gingiva or hard palate is usually described in maxillary ameloblastomas.[23]

MRI and CT offer the best imaging methods for visualization of extensive lesions. In this case with alveolar process involvement, radiographic views were not enough to make the diagnosis of sinonasal ameloblastoma. In some CT scan slices, primary involvement of maxillary sinus can be suspected. So, it is not easy to exclude extension into maxillary bone from a primary sinonasal tract ameloblastoma.

A number of modalities have been proposed in the

treatment of ameloblastoma, like wide excision, curettage, enucleation, cryotherapy, cautery, laser usage, radiotherapy and chemotherapy.[23]

The maxillary ameloblastoma are more difficult to treat because of the combination of the well vascularised, fragile, cancellous maxillary bones, presence of the paranasal sinuses, nasal and orbital cavities which readily facilitates tumor spread to the zygomatic bone, cranial base and paracranial structures and the pterygomaxillary fissure. Radical surgery as defined by Muller and Slootweg is a procedure in which ameloblastoma is removed with a marginal of normal bone by using segmental or marginal resection.[24] However, most investigators have recommended at least between 1 cm and 3 cm of surrounding healthy bone.[25]

Hertog *et al.* proposed annual follow up during five years after radical surgery of solid ameloblastomas.[3] In cases of maxillary involvement, up to a period of at least 10 years follow up has been recommended as these lesions were more dangerous clinically and can invade adjacent sinus and vital structures.[3,26]

The prognosis of the treatment is basically dependent to the extension of the lesion and adjacent structures involvement rather than origin of lesion.

Based on the histological findings there is strong evidence to propose origin of these tumors directly from the sinonasal tract epithelium. However, further studies are needed to substantiate the histogenesis of these tumors.

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