Case Report

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Dentistry Section

Conservative Management for Acanthomatous Ameloblastoma of Anterior Maxilla: A Rare Case Report

SANJEEV KUMAR[1], JITENDER KUMAR[2], AASIMAH HAMEED[3], ALOK BHATNAGAR[4], KEERAT PREET KOUR[5]

ABSTRACT

First described by Broca in 1886, Ameloblastoma is one of the most frequently encountered epithelial odontogenic benign tumour. The literature reports Mandible as the utmost favourable area of this tumour with approximately 80% of the cases involving this area. Histologically, an ameloblastoma can have six variations i.e, follicular, plexiform, basal, granular, desmoplastic and/or acanthomatous form. According to the 2005 World Health Organisation (WHO) classification the incidence of acanthomatous ameloblastoma is 7.06%. The acanthomatous type of ameloblastoma is deemed to be a rare and uncommon variation that mostly involves the mandible in 81% of the cases and maxilla in only 19% of the cases. In the maxilla, it is most commonly seen in the molar area, antral area and nasal floor with reported rate of occurrence being 47%, 33% and 9% respectively. The incidence in the canine are is only 9%. On the basis of previous studies, acanthomatous ameloblastoma is usually found in the geriatric population than in younger ones and is considered as an aggressive variant of ameloblastoma. Authors hereby present a case of acanthomatous ameloblastoma occurring in the anterior maxilla in a 36-year-old male patient presenting with painless swelling involving the left side of the anterior upper jaw managed with a conservative type of treatment. This type of presentation of the already rare acanthomatous ameloblastoma is extremely sporadic. Also, the occurrence of such a tumour in maxilla can significantly affect the progression and prognosis of such tumours because of the anatomical and histopathological differences in the maxilla and mandible and their densities, thereby further influencing the management and follow-up.

Keywords: Adamantioma maxilla, Jaw, Neoplasms, Odontogenic, Tumour

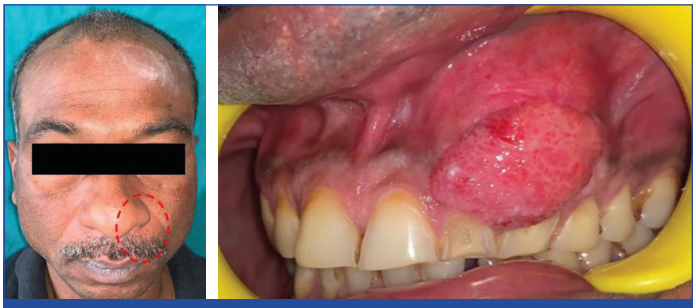
CASE REPORT

A 36-year-old male reported to Department of Oral and Maxillofacial Surgery with the complaint of swelling on his left upper front jaw region evolving for four months, small at the beginning then gradually expanding in size. Patient gave no significant past dental or medical history. Also, no history of pain, trauma or discharge from swelling was elucidated.

On extraoral examination, a large swelling with diffused borders of the left superior labium was seen that obliterated the nasolabial fold [Table/Fig-1]. It was approximately 2.5×2.5 cm in greatest dimension and extended from the left philtral ridge to the left nasolabial fold not crossing the midline anteroposteriorly and from the nasal floor to the vermilion on left side superiorinferiorly. The edges appeared indistinct; skin over the swelling appeared tense and glossy, with no change in colour. On palpation, the swelling was non tender, non compressible, and firm in consistency.

On intraoral examination single localised, round, sessile swelling approximately 2×2 cm in size extending from 22-24 anteroposteriorly and mucobuccal fold to cervical margin of gingiva superioinferiorly was seen. On palpation, the swelling was non tender, non fluctuant, fixed and firm in consistency. There was slight rise in temperature and bleeding on probing. Vestibular obliteration with expansion of left buccal cortical plates in relation to 22-24 were noted [Table/Fig-2]. Attrition in relation to 21-24 was also seen.

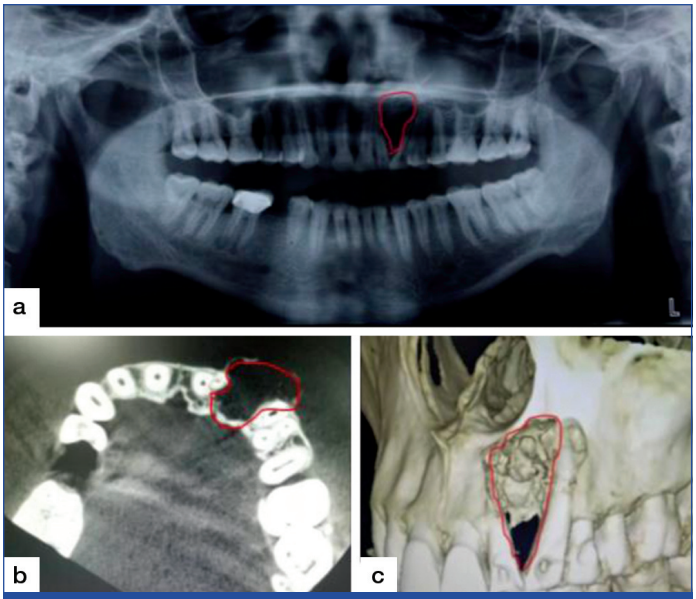
Radiological investigations, Orthopantomogram (OPG) and Cone Beam Computed Tomography were done. The OPG [Table/Fig-3a] revealed a multilocular radiolucency with a honey comb pattern in the anterior region with displaced roots of 22 and 23. A cone beam computed tomography [Table/Fig-3b,c] was done and it showed an osteolytic lesion that extended from distal side of 22 to mesial side of 23, displacing their roots. Expansion and break in continuity of buccal cortical plate and thinning of palatal cortical plate was noted. Based on the history, clinical and radiological features, a central giant cell granuloma was provisionally diagnosed. Ameloblastoma and odontogenic keratocyst were considered as differential diagnosis.



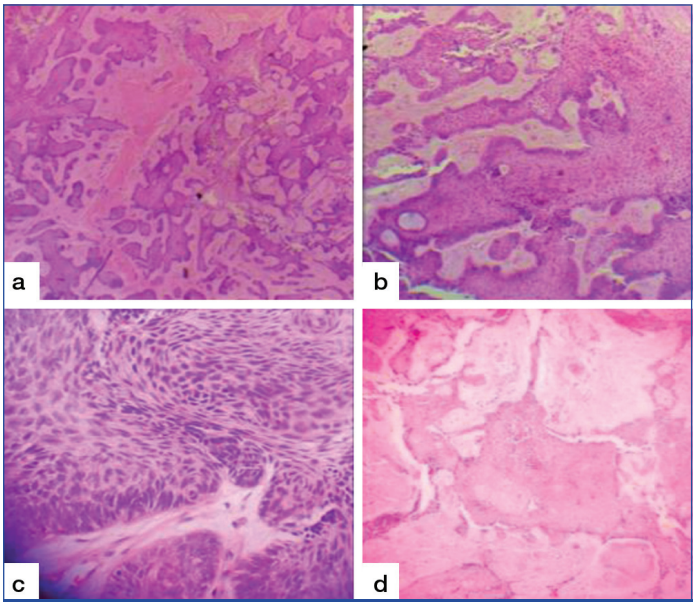
[Table/Fig-1]: Extraoral photograph showing swelling of the left side of superior labium obliterating the nasolabial fold. **[Table/Fig-2]:** Intraoral photograph showing localised, round, sessile swelling extending from 22 to 24. (Images from left to right)

Incisional biopsy sample taken and sent for histopathological examination revealed periphery of the follicle lined by a layer of tall columnar ameloblast like cells with polarity of the nucleus away from the basement membrane. Loosely arranged polygonal or angular cells resembling stellate reticulum were seen in centre region. Ameloblastic epithelium surrounded large keratin filled cavity and many solid epithelial cell nests also showed squamous differentiation suggestive of acanthomatous ameloblastoma [Table/Fig-4a-d].

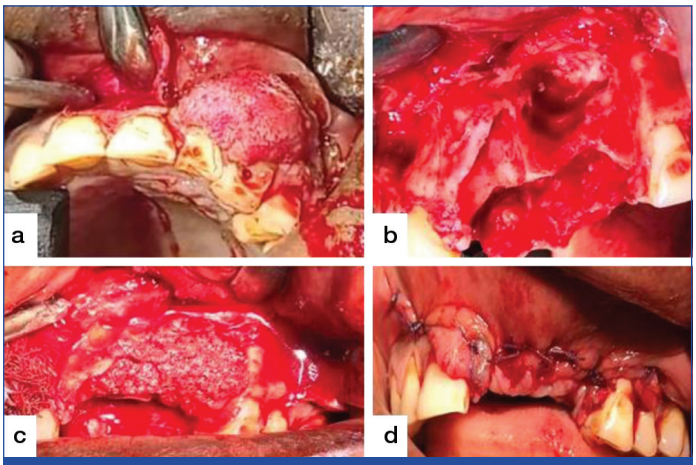
After obtaining the consent from the patient, patient was taken for surgery under general anaesthesia. A conservative approach was followed. Crevicular incision extending from 11-24 was made. A full thickness mucoperiosteal flap was raised. Extraction of 22, 23 and 24 was done and lesion was enucleated along with curettage of the site [Table/Fig-5a-d]. Peripheral ostectomy was done to ensure complete removal. Reconstruction was done with iliac cancellous bone. Antibiotics, analgesics and anti-inflammatory drugs were given postoperatively. Histopathological examination of the excised



[Table/Fig-3]: a) Orthopantomagram (OPG) showing honey comb pattern radiolucency extending between roots of 22 and 23; b) Cone Beam Computed Tomography (CBCT) showing expansion and thinning of buccal cortical plates between 22 and 23; c) 3D reconstruction showing displacement of roots.

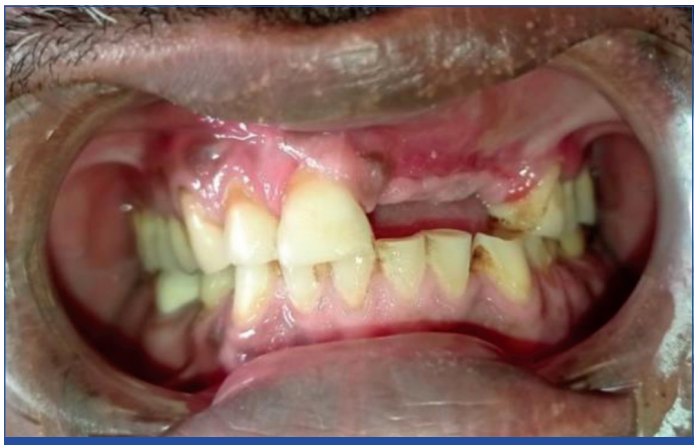


[Table/Fig-4]: Haematoxylin and Eosin stained slides under magnification 10X show: a) numerous tumour cells; b) tall columnar tumour cells with polarisation of nucleus away from the basement membrane along with places of squamous metaplasia; c) tumour cells show loss of polarisation, hyperchromatic nucleus and eosinophilic cytoplasm; d) Squamous metaplasia and keratinisation.

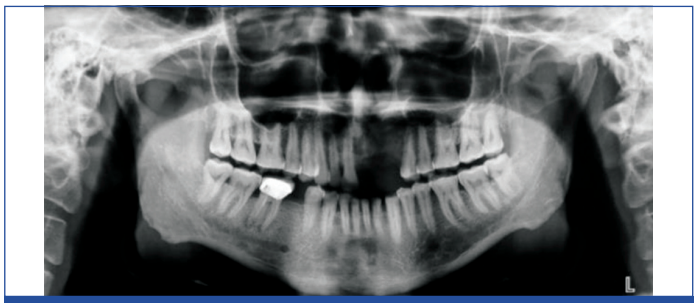


[Table/Fig-5]: Intraoperative photographs: a) Crevicular incision; b) Enucleation and curettage; c) Illiac grafting; d) Closure.

specimen confirmed acanthomatous ameloblastoma. Wound healing was uneventful. Patient was reviewed one week and one month postoperatively and there after every six months for clinical and radiographic assessment. During follow-up, graft uptake was satisfactory and there were no signs of inflammation, infection or recurrence of the tumour clinically [Table/Fig-6]. Radiographically, there were signs of graft maturation and new bone formation [Table/Fig-7].



[Table/Fig-6]: One year follow-up.



[Table/Fig-7]: One year follow-up OPG showing bone formation.

DISCUSSION

Ameloblastoma is a benign epithelial tumour of odontogenic origin. It is locally aggressive and destructive tumour and has a tendency to erode bone and invade adjacent structures. The term ameloblastoma was devised by Churchill in 1933, but Falkson in 1879 made the first detailed description [1,2]. It is the second most common odontogenic tumour after odontoma [3] and makes for approximately 1% of all tumours of the jaw, that's mostly encountered between 3rd to 5th decade of life. About 80% of all cases occur in mandible with a predilection towards the molar and ascending ramus with the incidence of approximately 70% [2,4].

There are various etiological factors associated with the incidence of ameloblastoma. Trauma, nutritional deficiencies, inflammation, irritation from extractions and dental caries were earlier thought as the aetiological factors associated with ameloblastoma. However the development of tumours of odontogenic origin was later associated with remnants of migrating epithelium of the enamel organ, remenants of odontogenic epithelium and lining of odontogenic cysts [5].

According to the current World Health Organisation (WHO) 2017 classification [6] of odontogenic tumours, ameloblastomas are divided into four main categories; conventional, unicystic, extraosseous and metastasising. Various histological subtypes have been defined, including those of follicular, plexiform, acanthomatous, mural, intraluminal and luminal [7]. The maxillary ameloblastoma makes for about 15% of all ameloblastomas. However, most occur posterior to the premolars and only 2% occur in the region anterior to them [7,8]. The acanthomatous variant is locally benign in most clinical scenarios but can potentially present as an aggressive tumour with invasion in adjacent bone. This can lead to increased chances of reoccurrence after a marginal surgical excision. The preference of location also varies among different variants of ameloblastoma. The plexiform and follicular ameloblastomas have a tendency to develop

more frequently in the molar-ramus region (plexiform 61.2%, follicular 58.5%) than in the incisor-canine region of the jaw (plexiform 10.6%, follicular 20.8%) [9]. The acanthomatous ameloblastoma variant is considered to be rare and uncommon.

According to the 2005 WHO classification the incidence of acanthomatous ameloblastoma is 7.06% [6]. The tumour more commonly appears in the lower jaw (81%) followed by the upper jaw (19%). In the maxilla, the lesion is mostly seen in the molar area (47%). However, in present case, it was seen in anterior maxilla. This type of presentation of the already rare acanthomatous ameloblastoma is extremely sporadic. On the basis of some previous cases, it was reported that acanthomatous ameloblastoma usually occurs in older individual, mostly in the seventh decade of life [10,11] which was not consistent with the present case where it was reported in a young male patient further making present case peculiar. As per the authors knowledge, only two case reports of acanthomatous ameloblastoma occurring in the maxilla have been reported in literature. Bansal M et al., in 2010 reported the case in a 13-year-old female child and Gruica B et al., in their case report published in 2003 reported a case of ameloblastoma in the maxillary sinus that consisted of three types; follicular, plexiform and acanthomatous [10,11].

Most of the lesions remain asymptomatic and are detected incidentally on radiographic studies however, the patients may also present with the history of a slow growing mass, malocclusion, loose teeth or more rarely pain and paresthesia, the lesions usually have a slow and gradual growth pattern but if left unattended can cause cortical plate resorption and extend to adjacent areas [12]. In the present case also the patient reported with a slow and progressively growing swelling.

On a radiography the ameloblastoma appears as a radiolucent, unilocular or multilocular cyst like lesion with a distinctive “soap bubble like” appearance, cortical thinning or perforation with adjacent tissue invasion and root resorption [8]. Similar radiographic features were also evident in our present case. The tumours that are devoid of the above mentioned characteristic features or are predominated by invasive growth and squamous component can be a diagnostic dilemma for a clinician. This can happen in cases of acanthomatous ameloblastoma as squamous metaplasia may also be present [13].

A multivariate analysis by Yang R et al., including 890 craniofacial ameloblastoma patients reported reoccurrence in 72 (9.78%) of case with cancerous recurrence in 15 (1.69%) cases however the recurrence and progression of ameloblastoma still remain unpredictable [14]. The treatment of choice is aggressive surgical resection. If possible, conservative surgery can also be performed if assured comprehensive removal can be achieved [15]. In the present case, conservative approach was taken and surgical enucleation with curettage of the lesion followed by autogenous graft placement was done. The intraosseous location of ameloblastoma along with the low sensitivity of this neoplasm limits the use of radiotherapy as an effective therapeutic modality. Radiation can also induce the development of secondary tumours. Therefore, in all the types of ameloblastomas, a meticulous and detailed long term clinical and radiographic follow-up is always recommended [16]. In the present case, the follow-up for one year was done and clinically and radiographically no recurrence of the disease was evident.

CONCLUSION(S)

Although ameloblastomas are among the commonest of odontogenic benign tumours, the incidence of occurrence of the acanthomataous variant of ameloblastoma in the anterior region of maxilla is extremely rare in human population. As per present knowledge, only a few cases of this type of ameloblastoma have been so far described in the literature. Ameloblastomas are aggressively treated owing to their locally destructive behaviour and propensity of reoccurrence, however, in present case it was managed conservatively and so far no recurrence is reported. However, further study of molecular mechanism, implication in clinical practice, and different treatment options have to be considered.

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PARTICULARS OF CONTRIBUTORS:
1. Professor and Head, Department of Oral and Maxillofacial Surgery, SGT University, Gurugram, Haryana, India.
2. Reader, Department of Oral and Maxillofacial Surgery, SGT University, Gurugram, Haryana, India.
3. Postgraduate Resident, Department of Oral and Maxillofacial Surgery, SGT University, Gurugram, Haryana, India.
4. Professor, Department of Oral and Maxillofacial Surgery, SGT University, Gurugram, Haryana, India.
5. Postgraduate Resident, Department of Oral and Maxillofacial Surgery, SGT University, Gurugram, Haryana, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Aasimah Hameed,
Postgraduate Resident, Department of Oral and Maxillofacial Surgery, SGT University, Gurugram, Haryana, India.
E-mail: aasimahkanth@gmail.com

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