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Recurrent ameloblastoma of mandible - A case report

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Abstract

Ameloblastoma is a benign odontogenic tumor of epithelial origin. It is locally aggressive with unlimited growth capacity and has a high potential for malignant transformation as well as metastasis. Ameloblastoma has no established preventive measures although majority of patients are between ages 30 and 60 years. Among all types of ameloblastomas, multicystic ameloblastoma is believed to be a locally aggressive lesion that has the tendency for recurrence. In this report we present a case of a large multicystic ameloblastoma in the right parasymphysis-body region of the mandible in a 31-year-old man. The large lesion was diagnosed with the help of cone beam computed tomography and was successfully managed by hemi mandibulectomy with simultaneous reconstruction. The need for extending the surgical margins beyond the radiographic extension to prevent recurrence is highlighted.

Keywords: Ameloblastoma, aggressive, recurrence, mandible, odontogenic tumor

Introduction

Ameloblastoma is the most common benign odontogenic tumor and accounts for 1% of tumors of jaws and 11% of odontogenic tumors^[1, 2]. It occurs as a painless, slow growing, locally aggressive tumor, causing expansion and perforation of the cortical plate causing infiltration of the soft tissues. Most of the tumors have a peak incidence during the 3rd and 4th decade of life, but can be found in any age group with equal gender predilection (1:1)^[3-7].

Radiographically, ameloblastoma can present as either unilocular or multilocular radiolucency, in honeycomb, soap bubble, tennis racket pattern, spider-like or mother and daughter cell appearances^[8, 9]. In some cases, cortical plates are spared and expanded whereas in other region they are destroyed. Root resorption is a common finding along with tipping of teeth. There are several histopathological subtypes-follicular, acanthomatous, plexiform, desmoplastic, granular cell and basal cell pattern, which may exist singly or as a combination of two or more types^[10]. Ameloblastoma is treated by enucleation, curettage or surgical resection depending on size and type of the lesion^[8]. Complete excision of large tumors and reconstruction of the defect have always posed a challenge. Radical treatment can leave major cosmetic and functional sequel and may require free flap reconstruction. Although conservative treatment preserves integrity of the bone, there are high chances of recurrence, the rate ranging from 55% to 90%. The recurrence rate of ameloblastomas after segmental resection was 4.5% and after marginal resection was 11.6%^[2, 11]. To prevent local recurrence, wide resection along with healthy bone margin is preferred. This article reports a case of recurrent follicular ameloblastoma along with comprehensive knowledge review regarding the epidemiology, classification, occurrence, and diagnosis.

Case Report

A 31-year-old male patient reported to the department of oral medicine and radiology in July 2021, with a chief complaint of pain and swelling in the lower right posterior region of the jaw since 6 months accompanied with numbness of the lip on the affected side. He gave the previous history of a similar swelling on the same site, 15 years back for which, he was operated under general anesthesia, the record for which was not available. However, the swelling reappeared in the last 6 months causing difficulty in chewing food with that side, due to mobile teeth. On extra-oral examination there was gross facial asymmetry, due to a single diffuse swelling in the right posterior region of the jaw, extending from a line joining corner of

The lip and the tragus up to the inferior border of the mandible. (Fig-1) It measured approximately 3 X 3cm² in size and the overlying skin appeared stretched with no sign of inflammation. Swelling was tender, uniformly hard to firm in consistency and there was no local rise of temperature. On intra-oral examination 45, 46 and 47 were missing. The swelling extended medio-laterally from distal aspect of 31, crossing the midline to distal aspect of 47, (Fig-2) obliterating the labial and buccal vestibule with no signs of pus discharge or inflammation. The swelling was uniformly hard and non-tender on palpation. The lingual cortical plate was expanded anteriorly from 31 to 44 and buccal cortical plate from 45 to 47. There was perforation of the buccal cortical plate in relation to 46. (Fig-3) There was grade I mobility associated with 32, 31, 41, 42, 43, 44 and midline drifting of 31, 41, 42, 43, 44 towards distal aspect. The patient gave history of intentional endodontic treatment done with 31, 41, 42, 43, 44 during previous surgery.

A provisional diagnosis of recurrent benign odontogenic tumor was made with differential diagnosis of recurrent ameloblastoma and central giant cell granuloma. The patient was advised radiographic investigations including panoramic radiograph, mandibular occlusal and CBCT. A large well defined multilocular radiolucency was seen in the panoramic radiograph, extending from distal aspect of 31 to mesial aspect of 47 which was of 2 X 5 cm² in size and supero-inferiorly from the alveolar crest till the inferior border of the mandible without expanding it. (Fig-4) Honeycomb appearance could be appreciated i.r.t. 44, 45 region and soap bubble appearance I.R.T. 31 to 43 and 46 to 47 region. There was external root desorption of 31, 42, 43 showing knife edged pattern and the inferior alveolar nerve canal was pushed downwards. The multilocular radiolucent lesion expanding the bucco-lingual cortical plate from 31 to 47 was observed in the mandibular occlusal radiograph. (Fig-5) Axial CBCT section showed expansion and perforation of buccal and lingual cortical plates and coronal sections revealed involvement of inferior alveolar nerve canal. (Fig-6, 7, 8) The radiographic appearance was suggestive of Ameloblastoma. The histopathological report after incisional biopsy of the lesion showed islands of ameloblastic epithelial cells with long columnar and stellate reticulum cells with reverse polarity in the nest suggestive of follicular ameloblastoma. Surgical resection with right hemi mandibulectomy under general anaesthesia was performed followed by prosthetic rehabilitation.



Fig 1: Swelling in lower right posterior region



Fig 2: Midline drifting



Fig 3: The swelling extended medio-laterally from distal aspect of 31, crossing the midline to distal aspect of 47, obliterating the labial and buccal vestibule



Fig 4: OPG



Fig 5: Mandibular occlusal radiograph



Fig 6: PAN image

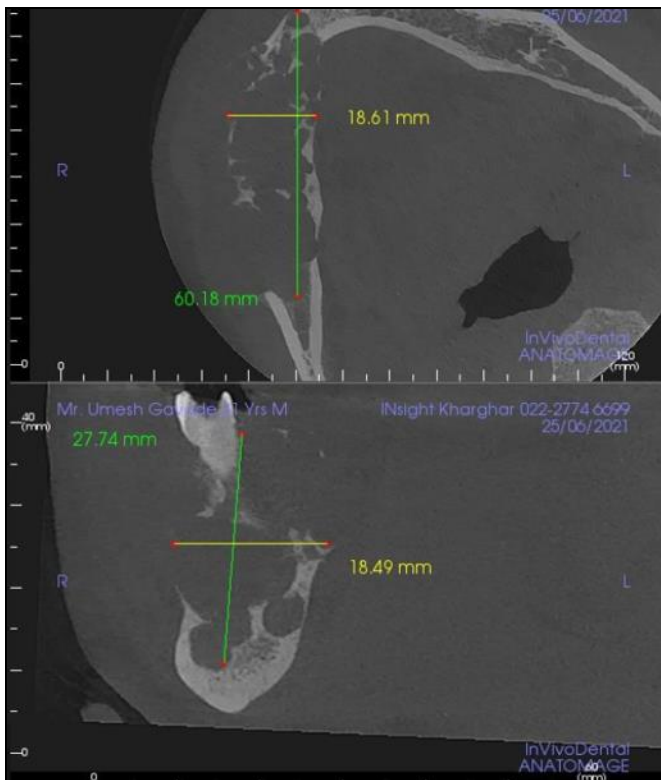


Fig 7: Axial and sagittal CBCT sections

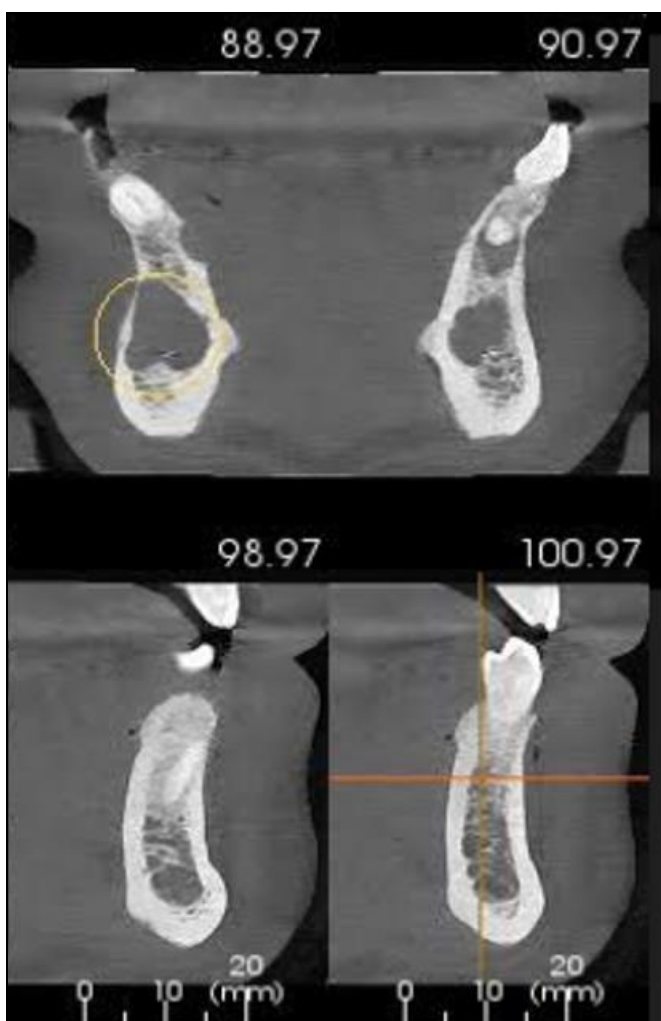


Fig 8: Coronal and sagittal CBCT

Discussion

Ameloblastoma is a locally aggressive, anatomically benign

tumour of the oral cavity which can undergo malignant transformation [12]. Etiological factors associated with ameloblastoma have evolved over the years and are yet to be conclusively established. As development of odontogenic tumours are associated with remnants of the migrating epithelium at the cervical loop of the enamel organ, it is not surprising that development of ameloblastoma is also linked to the enamel organ, remnants of odontogenic epithelium and lining of odontogenic cyst. Studies have also put forward that the absence of stratum intermedium hinders the differentiation of pre ameloblasts to ameloblasts because the stratum intermedium produces alkaline phosphatase needed to breakdown nutritional elements that will be passed on to ameloblasts during the bell stage. It should also be taken into account that the stellate reticulum within the tumour nests of columnar epithelium can degenerate to form microscopic cysts. The coalescing of these microcysts to form larger cystic spaces gives the multicystic features of ameloblastoma [13, 14]. At the molecular level, the genetic factors involved in tooth development, morphogenesis, cytodifferentiation and tooth patterning have been associated with development of ameloblastoma because some of these are altered significantly in ameloblastic tissues [13]. The molecular pathogenesis of ameloblastoma is now attributed to dysregulation of the mitogen-activated protein kinase (MAPK) pathway based on studies using ameloblastoma tissues, cell lines and transgenic mice [13, 15]. The mutation causes constitutive activation of BRAF protein downstream of MEK/ ERK that ultimately results in neoplastic transformation [16]. To further strengthen the association of MAPK signalling with ameloblastoma, the mutations in the RAS gene that acts upstream of BRAF and fibroblast growth factor receptor 2 (FGFR2) have also been identified in ameloblasts [17, 18]. Taken together, these more recent molecular data strongly indicate the existence of unique genetic abnormalities that eventually lead to development of ameloblastoma.

Varkhede *et al.* [6], Chukweneke *et al.* [23] and Ranchod *et al.* [25] reported that ameloblastoma appears in equal frequency between sexes, although higher frequency in females than in males was reported by Mahos *et al.* [26]. In our case, the site of occurrence of ameloblastoma was the mandibular posterior region. Agbaje *et al.* [19], Ruslin *et al.* [20], Siar *et al.* [21] and Reichardt *et al.* [22] indicate that the mandibular posterior region is the most common site affected by ameloblastoma. However, Chukweneke *et al.* [23] and Adekeye *et al.* [24] in their study showed that the anterior region is more commonly involved. The difference in location in the various population groups is largely unknown and the histological and molecular characteristics of the tumour can be related to geographic and ethnic differences [25].

According to Ranchod *et al.* [25], 68.32% of ameloblastoma predominantly showed soap bubble pattern, 15.84% showed a honeycomb pattern and spider like pattern was present only in a small percentage of individuals (10.67%). The case reported here presented a hybrid pattern with both soap bubble and honeycomb appearance. Becelli *et al.* inferred that, in mandibular ameloblastoma half of the patients presented with swelling of the affected region (38.3%), paraesthesia of the innervated region of the mandibular nerve (13.3%) and altered occlusion of teeth in 10% of cases. The patient reported here also presented with swelling in the right mandibular region along with paraesthesia.

It seems acceptable to group the treatment regimens of ameloblastoma into three modalities-conservative, that includes enucleation and curettage, marsupialisation and

radical surgery which includes resection with or without continuity defect^[8]. Radical surgical sectioning is customarily the treatment of choice for biologically aggressive subtype of primary and recurrent ameloblastomas. It involves en bloc tumour resection with wide bone margin followed by an immediate or delayed bone reconstruction of the surgical defect with tissue grafts and prosthetic rehabilitation. The inter-relationship between clinical and histological properties of the ameloblastoma determines its aggressiveness which in turn dictates the treatment approach and recurrence^[13]. Radical approach is indicated for large ameloblastomas involving the inferior alveolar nerve canal or below or far more aggressive variants like intramural ameloblastoma or multicystic type. It involves segmental or marginal resection with 1.5 to 2 cm normal bony margin beyond the radiologic margin^[12]. Possible factors for recurrence after radical resection may be the histologic type and location of the tumour and solid type particularly, follicular variety being the most aggressive type. Ranchod *et al.* reported the follicular variant to be the known variety to cause recurrence^[25]. In the mandibular posterior region, which is the common site of occurrence, the tumour can invade the cancellous portion beyond radiological margin to cause cortical perforation. Gradually invasion of the periosteum can lead to spread of the tumour cells to the soft tissue.

Conclusion

A common reason for recurrence of ameloblastoma is inadequate surgical resection. At least 1cm of healthy bone should be removed during surgical procedure beyond radiographically visible margins. The same reason would have led to recurrence of the tumour in the case reported here. Hence it is highlighted that the best treatment of ameloblastoma is aggressive embolic resection with simultaneous reconstruction.

Conflict of interests

The authors declare that they have no conflict of interests.

Authors' contribution

Dr. Simran Verma and Dr. Deepa Das equally contributed to this article.

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