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Case Report

Use of cone beam computed tomography in the diagnosis and treatment planning of follicular ameloblastoma: A case report with review of literature

Ananya Madiyal, Subhas G Babu, Renita Castelino, Vidya Ajila, Supriya Bhat, Sonika Achalli, Medhini Madi, Kumuda Rao

Department of Oral Medicine and Radiology. A. B. Shetty Memorial Institute of Dental Sciences, Nitte University,, Deralakatte, Mangalore

Abstract

Ameloblastoma is a benign but locally invasive odontogenic tumor. It is found most often in the posterior mandible and occurs as a slow growing painless mass. Diagnosis is made based on the clinical, radiographic and histopathologic features. Newer imaging modalities such as cone beam computed tomography offer massive advantage over traditional radiography thereby helping in better diagnosis and treatment planning. The goal of treatment is complete elimination of the tumor with reconstruction of the defect. This can be achieved with precise radiographic images concerning the size and extension of the lesion as is obtained by cone beam computed tomography. We present here a case of follicular ameloblastoma with a detailed description of its various radiographic features thereby improving the body of knowledge regarding the use of this new development in the field of imaging.

Keywords: Ameloblastoma; CBCT; Radiography; Follicular.

INTRODUCTION

Ameloblastoma is a rare, benign but locally aggressive neoplasm that represents half of all odontogenic tumors and 1% of oral ectodermal tumors.(1) The WHO defines ameloblastoma as locally invasive polymorphic neoplasm consisting of proliferating odontogenic epithelium which usually has a follicular or plexiform pattern lying in fibrous stroma.(2) It was first described by Cusak in 1827 and was named adamantinoma by the French physician Louis-Charles Malassez in 1885. The first detailed description was given by Falkson in 1879. Over time, the lesion was referred to by many names such as adamantine epithelioma, cystosarcoma and was renamed amelobalstoma only in 1930 by Ivey and Churchill for the French word 'amel' meaning enamel and the Greek word 'blastos' for germ (1,3).

Ameloblastomas occur in the third to fifth decades of life and show a propensity for occurring in the mandible with 70% of the tumors occurring in the molar-ramus area. They show an increased prevalence in males with the male-to-female ratio of 1.7:1(1,2,4).

Although the etiology is not confirmed in its entirety, it is known that the tumor can arise from the enamel organ, epithelial cell rests of Malassez, cell rests of Serre, remnants of dental lamina, epithelium of odontogenic cysts or basal epithelial cells of the oral mucosa (2,4). Clinically and radiographically ameloblastoma can be differentiated into three forms viz. unicystic form- with a radiographic feature of a unilocular radiolucency, solid/multi-cystic form- with a radiographic feature of multilocular honeycombed or soap bubble appearance and an extraosseous peripheral form (4,5).

The solid variant was first described in 1977 by Robinson and Martinez and is found to be more aggressive and recurrent than the unicystic variant. The neoplasm is well known for its tendency to recur, especially in the presence of cortical perforation or soft tissue invasion(5).

Since excision is curative while enucleation causes recurrence, treatment for all types of ameloblastoma centres around total excision with wide margin of 2 cm of healthy bone(4).

CASE REPORT

A 50 years old male patient reported to the dental hospital with a complaint of a slowly progressive swelling in the

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Corresponding Author: Ananya Madiyal, Department of Oral Medicine and Radiology. A. B. Shetty Memorial Institute of Dental Sciences, Nitte University, Deralakatte, Mangalore E-mail: ananyamadiyal@gmail.com

left lower jaw since the last 6 months with pain during mastication that radiated to the entire jaw. The patient also noticed occasional discharge of a yellow-white fluid from the same region. He reported that the impacted posterior teeth in the region were extracted 7 months ago although they were asymptomatic at the time of surgery. His medical history was non-contributory. A soft, palpable but non-tender solitary left submandibular lymph node was present which measured 1 X 1 cm in dimension. Extra-oral examination showed no gross asymmetry of the face. Intra-oral examination revealed a diffuse, slightly tender swelling of the left mandibular alveolus extending anteriorly from the distal aspect of the mandibular left second premolar up to the retro molar pad posteriorly. The lesion was seen to extend up to the buccal and lingual sulci and cause an expansion of the buccal and lingual cortices with a fluctuance on the lingual aspect. The overlying mucosa appeared normal with no sign of any discharge (Figure 1).

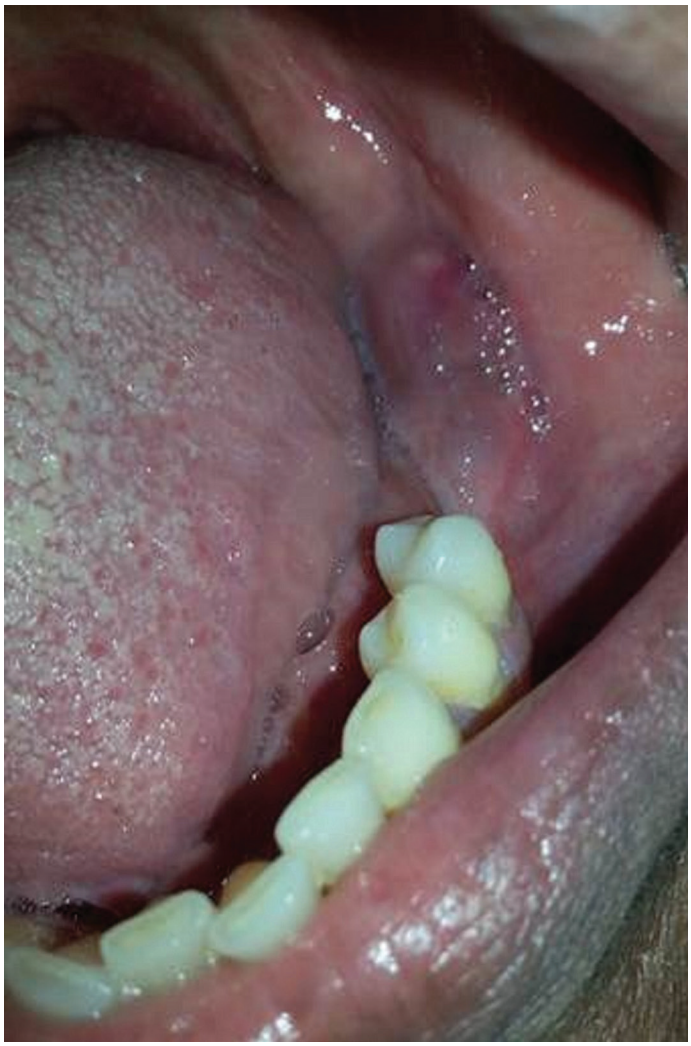

Figure 1. Intra-oral view showing a diffuse swelling of the left mandibular alveolus

Hard tissue examination revealed that the maxillary and mandibular right first molars, maxillary left third molar and all three molars on the left side of the mandible were missing. Fine needle aspiration at the site of fluctuance yielded a straw colored fluid with a bloody tinge. Based on the history, clinical findings and chairside investigations, a provisional diagnosis of residual cyst in the left mandible was made and differential diagnoses such as benign odontogenic tumor and odontogenic cyst were considered. Radiographic investigations were then carried out.

Intra-oral periapical radiograph of the left posterior mandible showed a well-defined multilocular radiolucency in the molar region measuring approximately 3X2 cm in size along with a soap bubble appearance of the internal structure (Figure 2).

Figure 2. Intra-oral periapical radiograph showing well-defined multilocular lesion with soap bubble appearance

Orthopantamograph revealed a well-defined multilocular radiolucency measuring 4 X 2 cm in size extending anteriorly from the region of the second mandibular premolar, posteriorly up to the middle third of the ramus. Superiorly the radiolucency extended from the crest of the alveolus, inferiorly up to the inferior alveolar canal. There was no displacement of teeth or resorption of roots. The inferior border was found to be intact. Internal structure showed locules with a soap bubble appearance suggestive of residual bone (Figure 3).

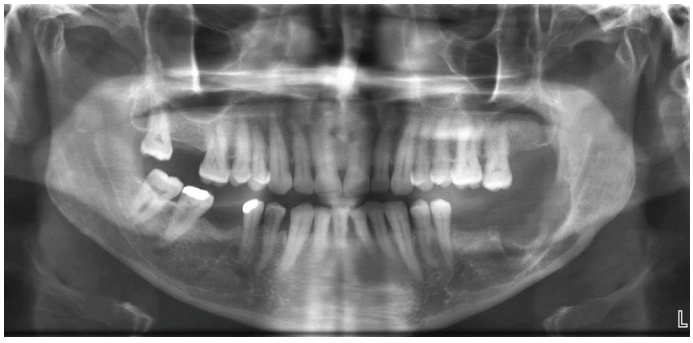

Figure 3. Orthopantamograph showing well-defined multilocular radiolucency with the classic soap-bubble appearance

Cone beam computed tomography was made with a small field of view. The axial section showed perforation of the

lingual cortex in the region of left mandibular molars (Figure 4)

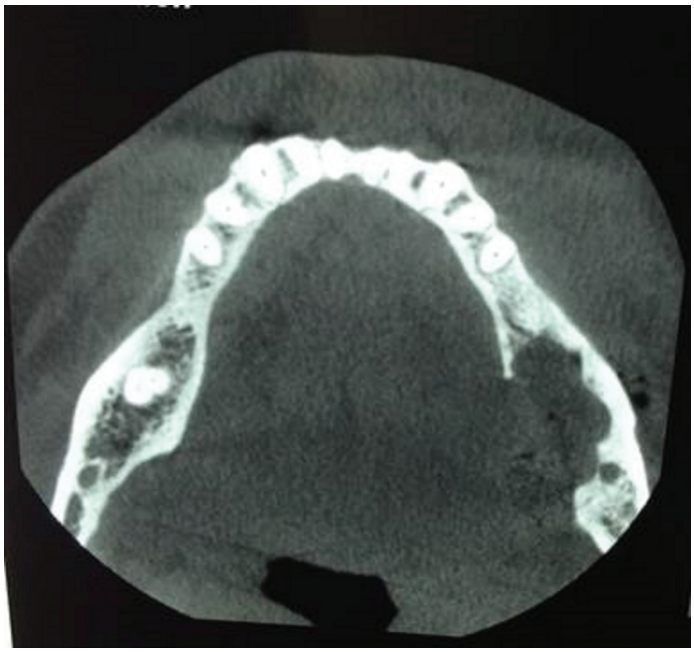

Figure 4. Axial section of C,BCT showing perforation of the lingual cortex

Reconstructed panoramic view showed a well-defined multilocular radiolucency extending anteriorly from the region of the left mandibular second premolar up to the middle third of the ramus posteriorly. The lesion extended from the crest of the alveolus up to the superior border of the inferior alveolar canal (Figure 5).

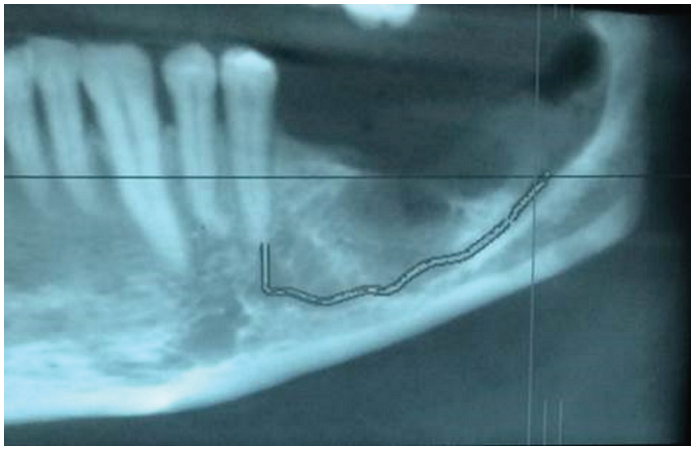

Figure 5. Panoramic reconstruction of CBCT showing proximity of the tumor to the inferior alveolar canal (Traced in the radiograph).

Cross sectional images of the lesion showed that the loss of bone was more pronounced on the lingual aspect while compared to the buccal (Figure 6 a,b,c).

The three dimensional reconstruction showed destruction of the buccal and lingual cortical plates with the lingual erosion extending into the inferior border of the mandible (Figure 7).

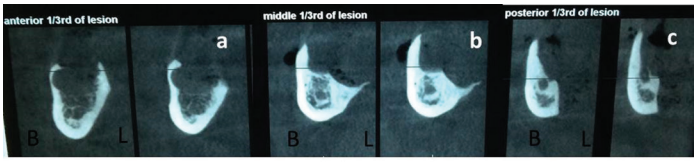

Figure 6 (a,b,c). Cross sectional images of CBCT showing loss of bone in the lingual aspect of the mandible

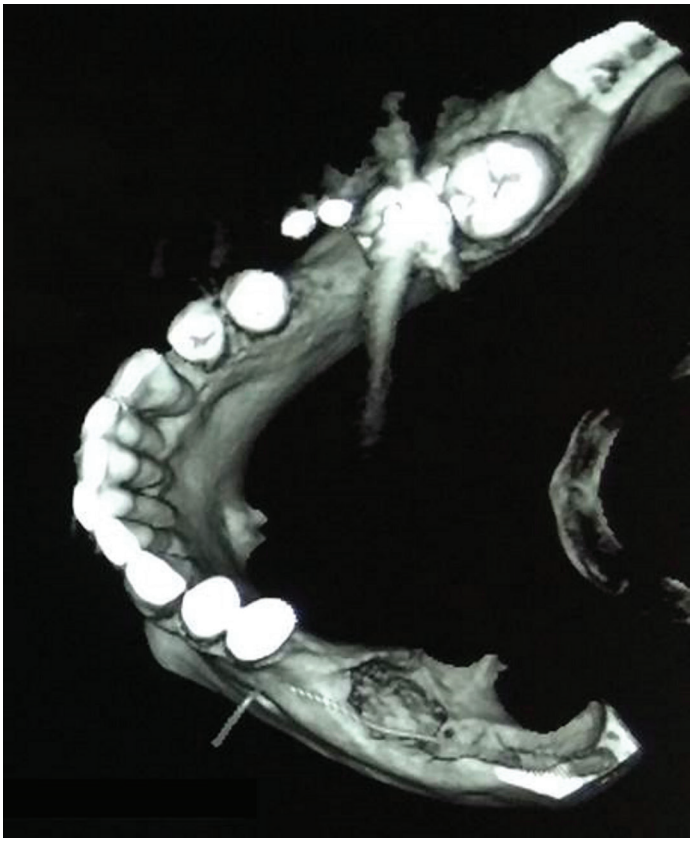

Figure 7. Three dimensional reconstruction showing perforation of the lingual cortex and inferior border of the mandible.

Considering these features, radiographic differential diagnoses of benign odontogenic tumor such as ameloblastoma as well as odontogenic cyst like keratocystic odontogenic tumor were considered.

Incisional biopsy was done. Histopathological examination of the H & E stained section showed surface epithelium, odontogenic epithelium and connective tissue. Tumour islands consisted of odontogenic cells present in islands and sheets. Tumour islands had peripherally placed tall columnar cells and hyperchromatic cells with palisaded nuclei. Centrally placed cells were star shaped stellate reticulum like cells. Few tumour islands showed squamous metaplastic changes in their centre. Connective tissue capsule was composed of dense bundles of collagen fibres, blood vessels and fibroblasts with inflammatory infiltrate consisting mainly of lymphocytes. Blood vessels, extravasated RBCs and salivary gland mucous acini and ducts could also be seen. Histologic impression had

features suggestive of follicular ameloblastoma (Figure 8 a,b).

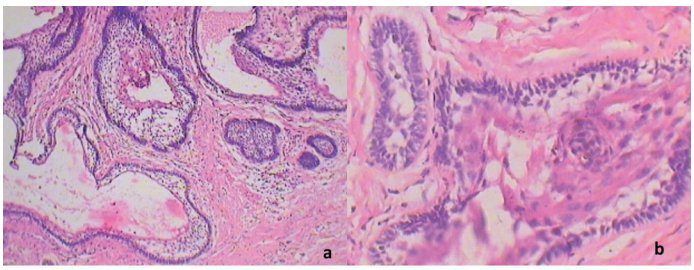

Figure 8 (a,b): Photomicrograph showing features of follicular ameloblastoma.

Given the size, extent and the aggressive nature of the lesion, resection was undertaken via an extra-oral approach. Enbloc resection with continuity defect was made, condyle was retained and the ramus and body were reconstructed using standard reconstruction plate. On gross examination the resected mandible showed resorption by the tumour involving the inferior border of the mandible. No ameloblastoma component was seen in the slides. This was attributed to the fact that a major portion of tumour was included in the incisional biopsy and the remaining portion may have been lost due to secondary inflammation. The patient was kept on a long term clinical and radiographic follow-up and is disease free 18 months post-surgery (Figure 9).

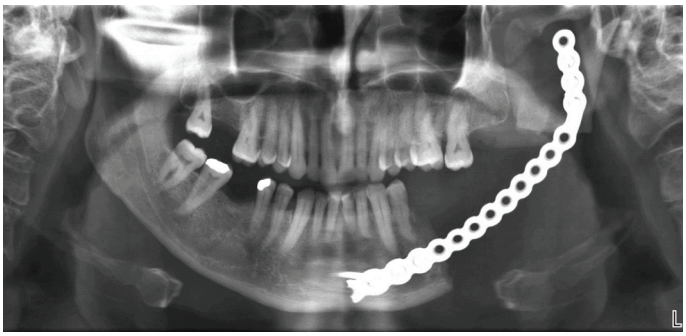

Figure 9. 18 months post-operative orthopantamograph showing the resected mandible and reconstruction plate.

DISCUSSION

Ameloblastoma is a neoplasm of the odontogenic epithelium, especially of enamel organ-type tissue that has not undergone differentiation to the point of hard tissue formation (6). It persists as a slow growing painless mass that often shows expansion of the buccal and lingual cortical plates with perforation and extension into the soft tissue (5). WHO histologically classified ameloblastoma into 4 subtypes: unicycstic ameloblastoma, classic solid/ multicystic ameloblastoma, desmoplastic ameloblstoma and extraosseous ameloblastoma (7). Other histologic variants include follicular and plexiform patterns which are the most common as well as the acanthomatous and granular cell types. Less common cellular variants are the papilliferous keratoameloblastoma, keratoameloblastoma, basal cell ameloblastoma and clear cell ameloblastoma (8). The various types do not vary greatly in their treatment except the unicystic and peripheral types which can be managed with simple enucleation and curettage.

The follicular pattern simulates the developing enamel organ and dental follicle by arranging the epithelial cells to resemble stellate reticulum. It consists of discrete follicles with similarity to the stellate reticulum of enamel organ and with the varying quantity of connective tissue stroma (9). The present case also conformed to the literature in these aspects. Ameloblastoma has a global incidence rate of 0.5 cases per million patient years but is varied in its geographic prevalence. It is seen with increased incidence in Asians and is the most common odontogenic tumor in China and Africa. However, in Canada and the United States, its incidence is second to odontomas.(3) The mandible is five times more likely to develop ameloblastoma and the posterior region of both the maxilla and mandible show an increased prevalence for the lesion (10). The present case also showed the presence of a slowly growing mass in the posterior mandible in an Indian male.

The unicystic and the multicystic types vary greatly in their radiologic presentation. While the unicystic variant resembles a cyst with its well-defined unilocular radiolucency, the multicystic variant presents as a multilocular radiolucency with the internal spetae arranged in honeycomb or soap bubble appearance. The appearance of septae in the radiographs is not an actual compartmentalization of the tumor by new bone formation but represents the differential resorption of the alveolar bone within the lesion (11). Worth described the internal structure as four major types- unicystic type that resembles a cyst but with break or discontinuity of the cortex, spider-web pattern that has coarse strands of trabeculae radiating from the centre to the periphery, soap bubble/multi-cystic pattern which resembles a bunch of grapes and the honeycomb/solid pattern where the lesion has not undergone cystic degeneration and presents as multiple small radiolucencies surrounded by hexagonal thick-walled cortices (11).

Roentogram usually shows a smooth periphery although large and advanced lesions might show thinning and expansion of the cortical plates.(10) The multicystic variant shows the highest prevalence for recurrence with 50% of the lesions recurring in the first 5 post-operative years. This may be either due to regrowth of the original tumor cells that were not completely resected or due to growth of new tumor cells (5). This makes long term clinical and radiographic follow up essential as is being done in the present case.

The location and its radiographic presentations are key factors in the diagnosis of the lesion. Diagnostic modalities can range from intraoral radiographs to extraoral radiographs like the mandibular lateral oblique views, orthopantomographs, computed tomography (CT) scans and magnetic resonance imaging (MRI) (10). Recently, cone beam computed tomography (CBCT) is rapidly gaining notable status due to its lower radiation exposure to the patient while compared to CT, ease of making the radiograph, relatively lower financial burden and decrease in the amount of radiographic artefacts. CBCT facilitates visualization of the three dimensional architecture of the lesion and provides accurate information regarding the internal structure, relationship with other anatomic structures and expansion of the cortical plates. It also provides a higher isotropic spatial resolution of osseous structures (12).

This helps in a more detailed and precise preoperative treatment planning. However, CBCT only provides information about the hard tissue, thereby necessitating an additional contrast CT with soft tissue windows or MRI for the soft tissue extent of the lesion. CBCT is extremely useful in cases of desmoplastic ameloblastoma (DA) which is usually misdiagnosed radiographically as a fibro-osseous lesion. Presence of both loss of internal bone structure as well as coarse internal calcifications in DA can be seen on a CBCT thereby confirming its radiographic diagnosis.(13)

In the present case, CBCT examination of the lesion was made using Planmeca ProMax 3D Mid (Helsinki, Finland) CBCT machine which helped to visualize the proximity to the inferior border of the mandible and the involvement of the inferior alveolar canal which were not seen in the traditional radiographs. Three dimensional reconstruction showed the perforation of the lingual cortical plate extending up to the inferior border of the mandible which was also not visualized in the other radiographs. This played a major role in planning of the treatment and rehabilitation of the patient.

CONCLUSION

Ameloblastoma is a highly aggressive odontogenic tumor of epithelial origin. Due to the typical radiographic appearance, it is often placed in the differential diagnosis for many cysts and benign tumors of the jaws. The goal of treatment of ameloblastoma is to achieve complete elimination of the lesion and to provide adequate reconstruction of the surgical defect. Diagnosis is usually enabled through the clinical behaviour, radiographic appearance, and confirmatory biopsy of the lesion. Cone beam computed tomography shows the border, internal structure, cortical expansion, erosion and surrounding structures which helps clinicians and radiologists with the preoperative diagnosis and treatment planning. Since CBCT is a recent development, large number of cases with varied features should be studied to form a database in literature and to compare it with traditional radiography to make the most of this new imaging modality.

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