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Review

A Case Study:

AMELOBLASTOMA – A RADIOGRAPHIC SERIES

DR. Subash B.V, MDS; [1] DR.Asha R Iyengar, MDS; [2] DR. K.Aparna, BDS, (MDS); [3] DR.Seema Patil, MDS. [4]

1, Associate Professor, 2, Head of Department,
3, Post graduate Student,
4, Associate Professor, 1, 2, 3, and 4, Department of Oral Medicine and Radiology,

D.A Pandu Memorial Dental College and Hospital, Bengaluru, Karnataka, INDIA

Correspondence: Dr K. Aparna, Post graduate student, Department of Oral Medicine and Radiology, D.A Pandu Memorial Dental College and Hospital, Bengaluru, Karnataka, INDIA. aparna.oralmedicine@gmail.com. Tel no: +91- 9491334231

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ABSTRACT

Background and Setting: Ameloblastomas are uncommon, locally aggressive odontogenic tumors that comprise 1% of radiolucent lesions of the jaws. On panoramic radiographs, these lesions may appear either unilocular or multilocular; and, histologically, as unicystic or multicystic. Both unilocular and multilocular variants have been shown to recur, particularly following inadequate surgical treatment. The site, margin and influence of the lesion on adjacent structures can be assessed radiographically and can aid in diagnosis and further treatment planning.

Objective: A series of 6 random cases of ameloblastomas which, reported to the out- patient department of the institution, and that presented with varied clinical and radiographic features were studied.

Materials and Methods: A hospital-based retrospective study was conducted by reviewing the clinical and radiographic records of archived ameloblastoma cases from 2016 to 2017; available in the department. The total of 6 patients' records that fulfilled the criteria of the study was selected.

Results: The ameloblastoma patients in this case series, were in the age group 14-41years. The male: female ratio was 2:1. The mandible (100%) was more commonly affected than the maxilla (0) of which the posterior mandible was involved in 83.3% cases. Off the posterior mandibular cases, 80% cases showed multilocularity, and 20% of the cases exhibited unilocular radiolucencies. In the anterior mandible, all the cases were 100% unilocular. Root resorption of variable degree was distinctly observed in only 33.3% cases, whereas was absent in 66.7% cases; as opposed to the tooth displacement, which was seen in 4/6 (66.7%) cases (and absent in 33.3% cases. The cortical expansion on the buccal and lingual side was 66.7% whereas no cortical expansion was noted in 33.3% of the cases.

Conclusion: Radiographic findings are not always expected to provide a specific diagnosis, but aids in narrowing down the differential diagnosis; assisting the clinicians in determining the patient's treatment planning. It is important for a surgeon to know the vital radiologic features of ameloblastomas, which are prevalent in their local population

INTRODUCTION

Ameloblastoma is a locally aggressive and slow growing; odontogenic epithelial neoplasm may form from the enamel organ, remnants of dental lamina, epithelium of dentigerous cysts, or from the basal layer of the oral mucous epithelium; and the term was coined by Ivey and Churchill in 1930. [1] This lesion was first described by Cusack in 1827 and etymologically, the name is derived from an old French word 'amel', meaning enamel, and Greek 'blastos', meaning germ or bud. Over time, this tumor has been

addressed by many names including ‘adamantine epithelioma’, ‘cystosarcoma’, ‘adamantinoma’, and finally ‘ameloblastoma'.Ameloblastomas exhibit a variable geographic prevalence, and is the most common benign odontogenic tumor in Africa and China, and the second most prevalent in the United States and Canada (most common being the odontoma). On the other hand, the Afro-Americans, present with an overall fivefold increased risk of disease as compared to Caucasians. The incidence of this tumor in the world has been estimated to be 0.5 cases/ million persons, and most of the cases are seen in patients 30–60 years of age. [2]There is high recurrence rate and accounts for 1% of cysts and tumors of the jaws and 10% of odontogenic tumors. [3]A malignant variant
with distant metastasis has also been reported in the literature. [4]

Radiographs play an important role in the diagnosis of these lesions. Plain X-rays have been found to lack sensitivity and specificity in understanding the extent of bone and soft tissue invasion. Based on the patterns created by the septae, various multilocular appearances of ameloblastomas are noted namely: ‘soap bubble’, ‘honey comb’ and ‘tennis racket’.

Most of the unicystic ameloblastomas appear as unilocular radiolucencies, [5] while the solid forms generally appear multilocular with the ‘soap bubble’ pattern being the most common. The unicystic form is clinically, radiographically and pathologically a different entity with a more favourable prognosis as compared to the multicystic form. The multicystic form has more chances of recurrence and is usually treated radically. Radio graphically; the ameloblastoma is a unilocular or multilocular lesion. The margin of the lesion may be either scalloped or smooth. (Figure 4)

There may be a thinning, expansion or even perforation of cortical plates if the lesion in its advanced stages. The occlusal radiograph may exhibit a cyst-like expansion, with thinning of the adjacent cortical plates that would leave behind only a thin 'eggshell' of bone. [6]

Computed tomography (CT) image shows a well-defined radiolucent uni/multilocular expansile lesion and is useful for the estimation of the soft tissue involvement and the cortical destruction (revealing a window for biopsy) to support surgical planning.

MRI, on the other hand, provides a complete information than the CT, about the marrow involvement and the soft tissue extension beyond the lytic bone cavity. It is especially useful in cases of the ameloblastomas arising from the maxilla, as it helps to earmark the extent of the lesion to the sinuses, orbit, and skull base. It is also useful in diagnosing desmoplastic ameloblastomas, because of the poorly defined soft tissue borders and the likelihood of it, being diagnosed as a fibro-osseous lesion. PET-CT is useful in the diagnosis of the metastatic ameloblastomas, where it may be useful in staging the distant metastasis. [2]

Notwithstanding the benefits of CT, MRI and the PET-CT, the structure of the lesion is best detected on panoramic radiographs. However, the less-sharp image and ghost images could be its drawbacks. [6]

Radiographically, the ameloblastomas may be mistaken for a fibroma, keratocystic odontogenic tumor, fibromyxoma, hemangioma, fibrosarcoma, giant cell tumor or an aneurysmal bone cyst. The desmoplastic ameloblastomas may mimic a globulomaxillary cyst or a fibro-osseous lesion. [6]

H. M. Worth has described four radiographic patterns of ameloblastoma:

Unicystic type: This lesion appears cystic, but unlike cyst, it causes a discontinuity or breaks in the cortex and may even show trabeculae traversing the lumen.

Spider-web pattern: Most common appearance and the lesion are seen as a large radiolucent area with scalloped borders. The center of the lumen exhibits coarse strands of trabeculae that radiate peripherally, resembling a gross caricature of a spider.

Soap-bubble pattern: This lesion is seen as a multilocular radiolucency with large compartments of varying sizes, giving rise to the soap-bubble appearance or a multi-chambered or multicystic 'bunch of grapes' appearance.

Honey-comb pattern: This is also called a beehive pattern. These are tumors that have not undergone cystic degeneration. Hence, multiple small radiolucencies are seen surrounded by hexagonal or polygonal thick-walled bony cortices, giving rise to a honeycomb appearance. [7]

This case series highlights the various aspects of ameloblastomas and its

radiographic appearances in 6 randomly chosen archival cases.

MATERIALS AND METHODS

The complete records of the patients with the diagnosis of ameloblastoma were retrieved from the archival records of the institution and then reviewed. Six records were selected which had detailed information about the case. These details were noted and then tabulated on a spread sheet.

RESULTS

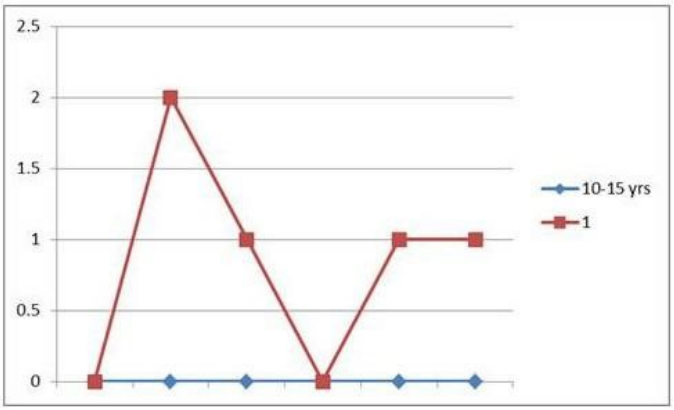
Out of 6 cases in the present case series, 4 cases were in males (66.7%), and 2 in females, with a male to female ratio, is 2:1. The youngest patient was 14 years of age and the oldest patient; 41 years. (Table 1)

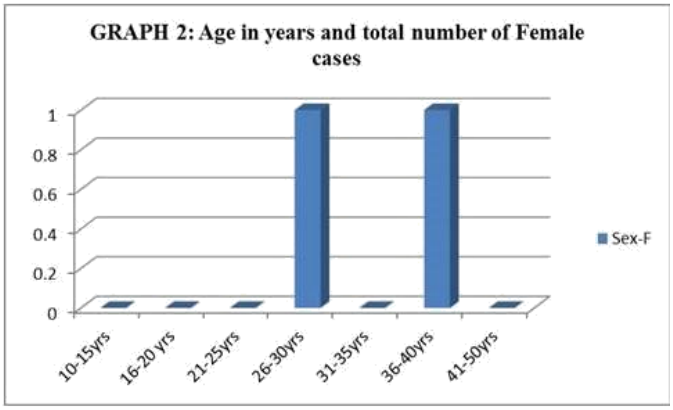
TABLE1: Age, Gender, Clinical details of cases.

Case	Age in Years	Gender	Location	Extra-Oral swelling	Intra-oral swelling	Duration	Pain	Tooth displacement	Cortical Expansion
1	14	Male	Posterior mandible	Present	Present	4 months	Present	Absent	Present
2	25	Male	Posterior mandible	Present	Present	5 months	Present	Absent	Present
3	25	Male	Posterior mandible	Present	Present	3 months	Present	Present	Absent
4	26	Female	Posterior mandible	Present	Present	2 years	Absent	Present	Present
5	39	Female	Posterior mandible	Present	Present	2 months	Absent	Present	Present
6	41	Male	Anterior mandible	Present	Present	12 years	Absent	Absent	Present

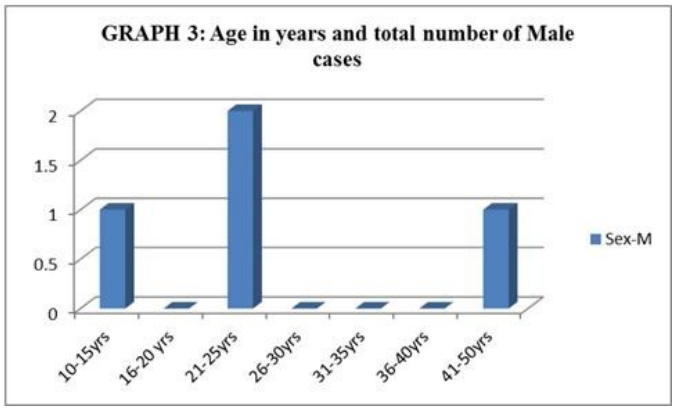
Ameloblastoma cases were seen in 66.7% males and remainder in females; the ages ranged from 14- 41 years in males and 26-39 years for females. The average male age was 26.3 years and in the case of the females averaged 32.5 years. The posterior mandible was involved in 83.3% cases and 16.7% in the anterior mandible. Both extraoral and intraoral swellings were present in all cases; Pain was seen in 50% of cases and absent in 50%; Tooth displacement occurred in 50% cases and was absent in the remainder 50%: Cortical expansion was present in 83.3% cases and absent in 16.7%

GRAPH 1: Age in Years and total number of cases

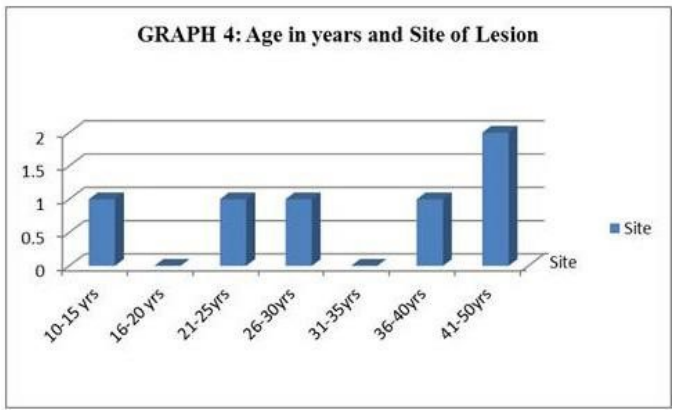




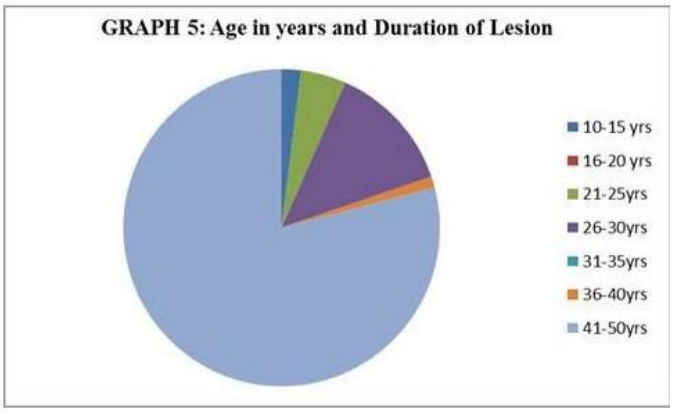
16.7% females in the age groups 26-30 years and 36-40 years each

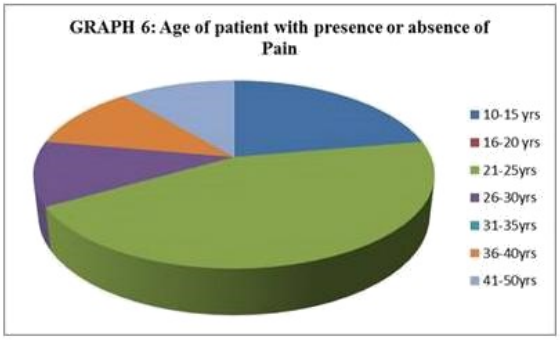


16.7% males in 10-15 age group and 41-50 age group and 33.3% in age group 21-25 years



Age in years and Posterior and anterior mandibular involvement (Post-1, Ant-2)





Age and presence or absence of pain where absent=1 and present=2

TABLE 2: Radiographic features of 6 cases

Case	**Location**	**Appearance**	**Internal structure**	**Septae**	**Tooth resorption**	**Tooth displacement**	**Cortical expansion**
1	**Posterior mandible**	Unilocular	Radiolucent	Absent	Present irt 46	Present irt 47,48	Buccal, lingual
2	**Posterior mandible**	Multilocular	Radiolucent	Present	Absent	Present	Buccal, lingual
3	**Posterior mandible**	Multilocular	Radiolucent	Present	Present irt 37,38	Absent	No expansion
4	**Posterior mandible**	Multilocular	Radiolucent	Present	Absent	Present irt 34,35	Buccal, lingual
5	**Posterior mandible**	Multilocular	Mixed (honey comb)	Present	Absent	Absent	Buccal , lingual
6	**Anterior mandible**	Unilocular	Radiolucent	Absent	Absent	Present irt 32,33	No expansion

Locularity= Unilocular-33.3%: Multilocular-66.7%; Radiolucency= Radiolucent-83.3%: Mixed-16.7%; Septae=Present-83.3%: Absent-16.7%; Root resorption=Present-33.3%: Absent-66.7%; Tooth displacement=Present-66.7%: Absent-33.3%; Cortical expansion=Buccal+Lingual expansion-66.7%: No expansion-33.3%

Site and locularity: - Posterior mandible and locularity=80% Multilocular-20% Unilocular; Anterior mandible=0-Multilocular: 100% -Unilocular;

Site and radiodensity: - Posterior mandible and Radio-80% Radiolucent: 20% Mixed; Anterior mandible=100% Radiolucent;

Site and septae: - Posterior mandibule and septae=80% Present: 20%- Absent; Anterior mandible=100% absent

Site and presence/ absence of Tooth resorption:- Postereior mandible= 60%-Absent: 40% Present; Anterior mandible=100% Absent

Site and Tooth displacement: -- Posterior mandible-60% Present: 60% Absent-40%; anterior mandible-60%-100% Present

Cortical expansion: -- Posterior mandible-80%-Buccal+Lingual: No expansion-20%; anterior mandible-100%-No expansion

The ages of males in the group ranged from 14-41 years, and the females between 26-39 years; Average male age was 26.3 years, and in the case of females it was 32.5 years. Among the 4 male cases, 3 of them had a lesion in the posterior mandible, and only 1 case was seen in the anterior mandible. Both the female cases had posterior mandibular involvement. The average duration of the lesion in males' was13years and females it was 4.2years

All the cases involved the mandible; and of these, 5 were present in the posterior mandibular region while 1 was in the anterior portion of the mandible. Body and ramus were involved in three cases (Table 2).

In the present case series; 4/ 6 (66.7%) cases presented with multilocular appearance and 2/6 cases with unilocular appearance. -83.3% cases were radiolucent whereas 16.7% had a mixed radiodensity. The radiolucent lesions showed septae in

83.3% cases whereas they were absent-in 16.7% of them. In 33.3% root resorption was observed whereas tooth displacement was seen in 66.7% cases. Cortical bone expansion on the buccal+lingual side was seen in 66.7% cases, and there was no expansion present in 33.3% cases. (Figure 4)

In 80% cases within the posterior mandible, the lesion was multilocular, and 20% cases were unilocular; whereas in the anterior mandible, the cases were 100%-unilocular. In 80% cases of the ameloblastoma affecting the posterior mandible were radiolucent and 20% cases had a mixed radiolucency and radiopacity whereas the lesions in the anterior mandible were 100% radiolucent. In 3/4th of the cases, the septae were present, and anterior mandible showed a 100% involvement of the septae. Root resorption was seen in 60% cases in posterior mandible; it was absent 40% cases, and in the anterior mandible there was no resorption in relation to the lesion. In 60% cases of the posterior mandible, there was tooth displacement, and the anterior mandible cases showed definite tooth displacement. The cortical expansion on the buccal and lingual side of the posterior mandible was seen in 4/5 cases, and there was no expansion in one case. The case that involved the anterior mandible showed absolutely no cortical expansion.

DISCUSSION

Robinson has defined Ameloblastoma as a unicentric, non-functional tumor, intermittent in growth, anatomically benign and clinically persistent.

Ameloblastomas are usually seen in the 3rd to 5th decades of life. In the present case series, 3 cases were seen below 30 years of age and 3 cases above 30 years. In a retrospective study, it was stated that prevalence of ameloblastoma was found to be highest in the 2nd decade amongst the younger Thai population. [9]However in the Burmese population it was highly prevalent in the 3rd decade. There is slight male predilection with the male: female ratio as 1.6:1. In the present case series, 4 cases were reported in males, and 2 were in females; with a male to female ratio of 2:1. 80% of the ameloblastomas [10] affect the mandible, especially the molar-ramus area [1]whereas the maxilla is less frequently affected. [4]In the present series, all the cases were reported in the mandible. Out of 6 cases, 5 were reported in the posterior mandible and 1in the anterior mandible.

Ameloblastomas present as an asymptomatic swelling [9] which grows slowly, and few, if any, symptoms, occur in the early stages. [11]Because of the poor socio-economic status of the patients the ameloblastoma was diagnosed in the late stages. [8]All the patients in the present series presented with extraoral swelling. However, a published case series found around 35% of the ameloblastomas; identified on routine radiographic examination. [10]Pain is a rare feature of ameloblastomas. [10]In the present series, 50% of subjects presented with pain and swelling and the remaining presented with only swellings.

Of the total cases, there were 66.7% males, and rest females and the involvement of the posterior mandible was seen in 83.3% cases, and the remaining cases involved the anterior portion of the lower jaw. All the cases showed both an extraoral and intraoral swelling. There was a pain in 50% cases, and tooth displacement was also seen in ½ of the cases selected. The cortical expansion was seen in 83.3% cases and was not visible in the remainder cases

The ages of males in the group ranged from 14-41 years, and the females between 26-39 years; Average male age was 26.3 years, and in the case of females it was 32.5 years. Among the males, 3 cases had a lesion in the posterior mandible, and only 1 case was seen in the anterior mandible. Both the female cases had posterior mandibular involvement. The average duration of the lesion in males' was 13yrs and in females it was 4.2yrs.

Pain can be associated with ameloblastomas, and this could be due to a chair-side investigative procedure such as FNAC which may cause haemorrhage leading to pain. Also, pain may be a sign of malignant ameloblastoma. [9]In the present case series, ulceration was seen in 1 case due to the investigative procedure.

Radiographically, ameloblastomas can present as unilocular or multilocular radiolucency. Based on the patterns created by septae; various multilocular appearances of ameloblastomas are described: which include the 'soap bubble', 'honey comb', 'spiderweb' and 'tennis racket' forms. According to Worth, the most common radiographic appearance of ameloblastoma is a multilocular radiolucency with a corticated border, and

margins, which usually show irregular scalloping. [7]

In the present case series 4 out 6 cases presented with multilocular appearances and 2 cases with unilocular appearance. Locularity= Unilocular-33.3%: Multilocular-66.7%; Radiolucency= Radiolucent-83.3%: Mixed-16.7%; Septae=Present-83.3%: Absent-16.7%; Tooth resorption= Present-33.3%: Absent-66.7%; Tooth displacement=Present-66.7%: Absent-33.3%; Cortical expansion=Buccal+Lingual expansion-66.7%: No expansion-33.3%

The locularity in the posterior mandible in 80% cases was multilocular and 20% unilocular; whereas in the anterior mandible the cases were 100%- unilocular. In 80% cases of the posterior mandibular ameloblastomas, there were radiolucent, and 20% cases had a mixed radiolucency and radiopacity; whereas the lesions in the anterior mandible were 100% radiolucent. In 3/4th of the cases affecting the posterior mandible, the septae were present, and anterior mandible radiolucency showed a 100% presence of the septae. Root resorption was seen in 60% cases in posterior mandible; it was absent 40% cases, and in the anterior mandible there was no resorption about the lesion. In 60% cases of the posterior mandible, there was tooth displacement, and the anterior mandible tooth displacement was observed. The cortical expansion on the buccal and lingual sides of the posterior mandible was seen in 4/5 cases, and there was no expansion in 1case. The case that involved the anterior mandible showed absolutely no cortical expansion

Unicystic ameloblastomas are most frequently seen in young adults, and they are remarkedly noted in the posterior region. [10]Unilocular radiolucency predominates over multilocular radiolucency in children and young adolescents. [12] In the present case series unilocular appearance was seen in two cases (Figures 1 and 6).

Of them, one case was reported in 41 years old with a unilocular radiolucency in the anterior mandible (Figure 6). A desmoplastic variant of ameloblastoma has a greater likelihood of occurring in the anterior mandible.[6]

FIGURE 1: *A unilocular well-defined radiolucency involving the posterior mandibular region with a displacement of 47, 48 from its normal position.*

FIGURE 2: *A well-defined, multilocular radiolucency in the left mandibular body and ramus surrounding 38 with faint septae and expansion of lower border.*

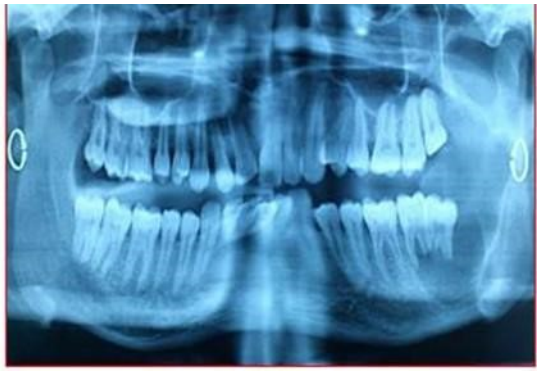


FIGURE 3: *Well defined, irregular, radiolucency involving left molar, angle, and ramus up to the sigmoid notch with a single bony septa and blunt resorption of root apices in relation to 37and 38.*

Ameloblastomas tend to cause extensive root resorption and tooth displacement. [11]In the present case series, 4 cases were presented with tooth displacement and 2 cases with knife edge pattern of root resorption (Figures 1 and 3).

Knife edge pattern is seen in ameloblastomas because roots are cut off in a single linear plane. 11When roots are not resorbed; they tend to extend into the lesion (Figures 2, 5, 6).

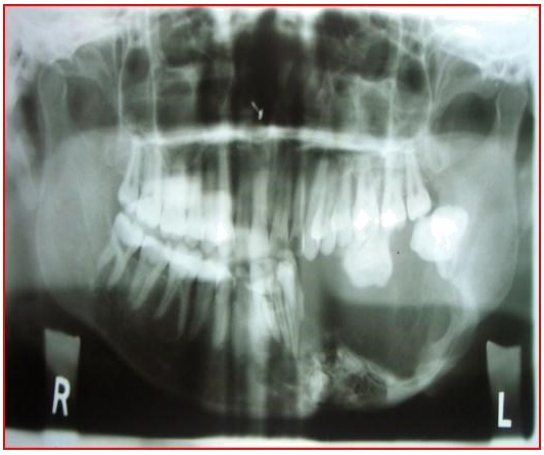


FIGURE 4: *Ill- defined radiolucency in the left body region of the mandible with radio-opaque scalloping is present at the lower border of the mandible and displacement of teeth in relation to 34, 35.*

FIGURE 5: *Well-defined unilocular radiolucency which is continuous with multilocular radiolucency extending from 47 till distal aspect of 43 and also involving inferior corticated border.*

FIGURE 6: *Well defined radiolucency which is surrounded by a sclerotic margin extending from mesial of 43 to distal of 33 is evident with the tilting of 32 and 33.*

Both buccal and lingual cortical expansion is seen in 5 cases, and expansion was not present in one case. Radiographically, this helps in differentiating ameloblastomas from dentigerous cysts, which expands only in one direction, usually to the buccal. [11]

Unicystic appearance and unilocular presentation are predominantly associated with impacted teeth. [10] In the present case series, 2 cases were associated with impacted tooth.

According to Worth, ameloblastomas may cause ballooning of inferior aspect of the mandible with convexity downwards which is of 'egg shell' thickness and intact. [11] In the present case series, one case presented with ballooning of inferior aspect of mandible (Figure 2)

CONCLUSION

Radiographs are an important diagnostic aid for oral lesions of various types, especially those that involve bone. It is important for the practising clinicians to know the salient features of ameloblastoma to strategize the treatment plan. Although very often the diagnosis of ameloblastoma is made by radiographic features, one should never rely on it, alone. All these lesions should be biopsied, and an accurate histologic diagnosis should be obtained before definitive treatment is commenced.

Conventional radiographs are the initial imaging choice for a patient presenting with swelling of the jaws. It is therefore mandatory for the clinician to be aware of the various radiographic appearances of ameloblastoma on conventional radiographs.

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CONFLICT OF INTEREST: No conflicting interests with regard to the study.

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