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CLINICORADIOLOGICAL ANALYSIS OF HISTOLOGICAL VARIANTS OF CENTRAL AMELOBLASTOMA- A RETROSPECTIVE STUDY

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ABSTRACT

Context: Ameloblastoma is an epithelial benign odontogenic neoplasm with varied histological variants exhibiting different clinical behaviour and radiological appearance.

Aim: To evaluate and correlate the clinical and radiological findings of histological variants of different clinicoradiological types of ameloblastoma.

Settings and design: Retrospective study.

Materials and Methods: A computerised English literature search using pubmed and google search was conducted for open access articles on the case reports of central ameloblastoma from the year 2011-2016. A total of 179 cases were retrieved and 93 case reports were included in the present study with all clinical and radiological relevant data.

Statistical analysis: Descriptive analysis was conducted for the data and the results were obtained.

Results: Study revealed among all the histological variants of different clinicoradiological types the variants of solid type were found to be of highest frequency (58.09%) followed by variants of unicystic (17.09%), desmoplastic (13.97%) and mixed variant (10.75%) respectively. In the variants of the solid type granular variant was found to be commonly occurring (31.48%). Mean age of occurrence is 34.8 years. There was equal preponderance in gender predilection. Radiologically common appearance was multilocular with large locules in variants of solid type and mixed variants, multilocular with sclerosis in desmoplastic variants and unilocular in variants of unicystic type.

Conclusion: This study provides an insight of different histological variants of ameloblastoma. The clinical and radiological characteristics of these variants brought forth the differences to the extent of implication in the appropriate diagnosis and treatment planning.

KEYWORDS : Ameloblastoma, desmoplastic,multilocular,unilocular.

Indroduction

Ameloblastoma is a true neoplasm of enamel organ type tissue and is one of the most commonly occurring, benign, but locally invasive neoplasm of odontogenic origin involving both maxilla and mandible occurring in different age groups. [1,2,3]Robinson defined it as Unicentric, Non-functional, Intermittent in growth, Anatomically benign and Clinically persistent tumour. [1]

According to WHO classification 2005 the different clinicoradiological types of Ameloblastoma included a) Solid/ Multicystic/ Conventional; b) Unicystic; c) Desmoplastic and d) Peripheral types. [4,5,6] The Solid, Desmoplastic and Unicystic types are together coined as Central or Intraosseous Ameloblastoma arising from bony component and the peripheral or extraosseous Ameloblastoma arises from soft tissue component. [7,8,9,10]

There are different histological variants of each clinicoradiological type. Solid type comprises of 5 different histological variants which include Follicular, Plexiform, Acanthomatous, Basal cell and Granular variants. Unicystic type included intraluminal, luminal and mural variants.[1]Desmoplastic type included only Desmoplastic variant and the Mixed variant is included in the present study which is a combination of either Solid, Unicystic or Desmoplastic variants. Since there are different histopathological variants an extensive insight into clinical and radiological behaviour was needed.

The need of the present study is to evaluate clinicoradiological characteristics of different histological variants of Central ameloblastoma and to correlate clinicoradiographic types with histological variants of Central Ameloblastoma.

Materials and Methods:

`A computerised English literature search using pubmed and google search was conducted for open access articles on case reports of Central Ameloblastoma based on histological variants from 2011-2016. A total of 171 cases were retrieved. Among them 93 cases were included in the study.

A retrospective study of 93 case reports was done extracting the clinical parameters of age, gender; duration of swelling (0-1year/1-2years/2years); site of occurrence which included right, left and anterior regions ; presence or absence of trismus, paraesthesia , extra and intra oral swelling and discharge.

The radiological characteristics which were evaluated in the case reports included- [Table 1].

[Table 1] Radiological inclusion criteria.

Location	Anterior, Posterior (included involvement into ramus, body of mandible, condylar and coronoid process for mandible and extension into nasal fossa and maxillary sinus in the maxilla), Anterior+ posterior and Bilateral involvement crossing the midline.
Appearance	Multilocular with large locules, small locules and sclerosis and Unilocular
Margins	Well defined / Ill defined
Shape of margins	Smooth/Scalloped/ Irregular
Borders	Corticated/ Sclerotic
Septae	Absent/ Thin and curved/ Coarse and curved
Buccal cortex, lingual cortex and Inferior border of mandible	Expansion, Thinning and Discontinuity
Associations with	Impacted tooth/Missing tooth
Affecting the tooth	Causing Root resorption / Displacement of tooth

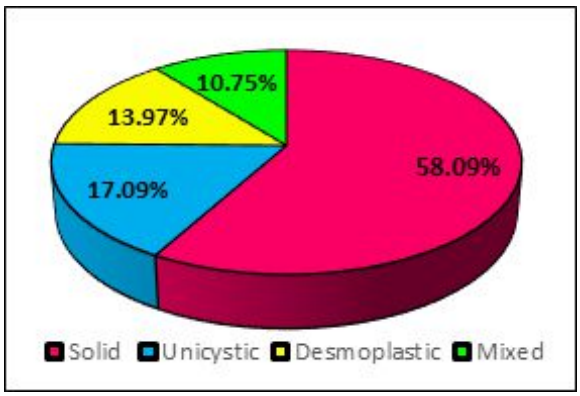
Ameloblastomas associated with other malignancies; Case reports before the year 2011; case reports without relevant clinical and radiological data (8); No adequate information available related to the study (25); case reports on animals (3); Peripheral Ameloblastomas (9); Associated with other tumours and cysts(4); Case reports without proper information on the histological variant (5); Recurrent cases (5); Only abstracts available(20); Case reports in other languages (4) were all excluded in the present study.

Statistical analysis: Descriptive analysis were conducted to calculate the frequency and percentage of the included variables and the results were obtained.

Results:
Over the period of 5 years the different case reports on Ameloblastoma were studied and the corresponding results were analysed.

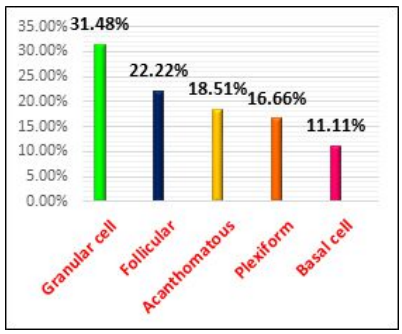
Histological Types-
Among all variants, the variants of Solid Ameloblastoma were frequently seen 54 cases (58.09%) followed by Unicystic 16 cases (17.09%), Desmoplastic 13 cases (13.97%) and Mixed variant 10 cases (10.75%) [Figure 1].

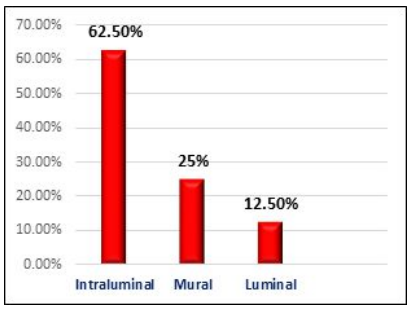
[Figure 1] Distribution of different histological types.



The Granular cell variant were 17 of 54 cases (31.4%) followed by follicular 12 (22.22%) ,Acanthomatous 10 (18.51%), Plexiform 9 (16.66%),Basal cell 6 (11.11%) variants among the solid types [Figure 2] and Intraluminal were of highest frequency 10 of 16 cases (62.5%) followed by mural 4 of 16 cases(25%) and luminal 2 of 16 cases(12.5%) respectively among the unicystic types. [Figure 3].

[Figure 2] Frequency of different histological variants of solid type
[Figure 3]-Frequency of different histological variants of Unicystic type





Age and Gender distribution-
Out of total 93 cases, 69 of 83 cases of Solid, Mixed and Desmoplastic type occurred in 2nd -5th decades of life (73.11%) and Unicystic type was found to be common in the 2nddecade i.e.,7 0f 10 cases (43.75%).

Out of 93 cases 49 cases occurred in females with a male:female ratio of 1:0.89. Results revealed that there was no significant association of gender with type of Ameloblastoma with a P value of 0.725.

Site of Occurrence-
Study revealed 81 of 93 cases (87%) occurred predominantly in mandible and the remaining 12 cases (13%) occurred in maxilla with a mandible: maxilla is 6.75:1. Maxillary involvement was predominatly noted in Desmoplastic variant (41.6%).

In both maxilla and mandible it was found that there was equal preponderance on right and left sides with very few cases occurring in the anterior region [Table 2].

[Table 2]: Distribution of site of occurrence

	Frequency	Percentage
Right	42	45.2%
left	43	46.2%
Anterior	08	8.6%

Duration of Swelling-
The duration of the swelling most frequently noted was <1 year [Table 3].

[Table 3] Distribution of Duration of Swelling

	Frequency	Percentage
0-1 year	48	51.6%
1-2 years	33	35.5%
>2 years	12	12.9%

Symptoms-
Pain was present in 22 cases of 93(23.7%), Trismus in only 2 cases (2.2%) paraesthesia in 5 cases (5.4%) and discharge in 3 cases of 93 (3.2%) were noted.

Extraoral swelling and intraoral swelling were found be present in >85% of cases (82 cases of extraoral and 92 cases of intraoral).

Location Radiographically-
Out of 93 cases occuring in maxilla and mandible 83 cases (89.2%) occurred in posterior region with 21 cases extending anteriorly (22.5%) and 12 cases crossing the midline (12.9%) common in granular variant and 10 cases occurred only in anterior region predominantly in desmoplastic variant.

Study revealed that appearance was predominantly multilocular with large locules in all variants of solid and mixed types, unilocular in unicystic and multilocular with sclerosis in desmoplastic variant. Margins are well-defined in variants of solid, unicystic and mixed, Ill-defined in desmoplastic variant. Shape of the margins was scalloped predominantly in varaints of solid and mixed with smooth margins in unicystic and irregular in desmoplastic variant. Borders are corticated in all variants except in desmoplastic where 100% sclerotic borders were noted. Septae were present in all variants except in variants of unicystic type. [Table 4]

Table 4: Radiographic parameter results

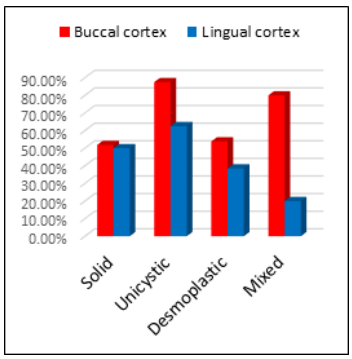
FEATURES	SOLID	UNICYSTIC	DESMOPLASTIC	MIXED
Appearance	Multilocular with Large locules (68.5%)	Unilocular (75%)	Multilocular with sclerosis (76.9%)	Mutilocular with large locules (70%)
Margins	Well-defined (72.2%)	well-defined (100%)	Ill-defined (76.9%)	Well-defined (70%)
Shape of margins	Scalloped (79.6%)	Smooth (62.5%)	Irregular (76.9%)	Scalloped (80%)
Borders	Sclerotic (51.8%) Corticated (48.1%)	Corticated (62.5%)	Sclerotic (100%)	Corticated (90%)
Septae	Thin and curved (57.4%)	Absent (68.75%)	Coarse and curved (76.9%)	Thin and curved (50%)

Association with an impacted tooth was present in 13 cases of 93

(14%).Tumour causing displacement of teeth was seen in 32 of 93 cases (34.4%) predominantly seen in desmoplastic variant with 12 of 13 cases (92.3%). Resorption of teeth was noted in 37 cases of 93 (39.8%) predominantly seen in mixed variant about 5 of 10 cases (50%).

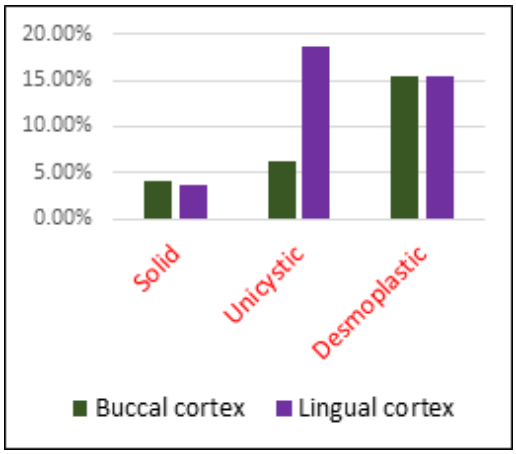
Expansion and thinning of buccal cortex was seen frequently than lingual cortex with the significant difference noted commonly in mixed variant (Buccal cortex-80%; lingual cortex-20%) [Figure 4] while solid variant did not show significant difference.

[Figure 4]- Expansion and thinning of buccal and lingual cortex



Study revealed discontinuity of buccal and lingual cortex was equally seen in variants of solid, desmoplastic types. Discontinuity of lingual cortex was predominantly noted in variants of unicystic type (Lingual-18.75%, Buccal-6.25%) and it is not reported in mixed variant cases. [Figure 5]

[Figure 5]- Discontinuity of buccal and lingual cortex



Expansion and thinning of inferior border of mandible was most frequently seen in variants of solid type which is about 17 of 50 cases occurring in mandible (34%). Discontinuity was predominantly seen in variants of unicystic type 4 of 12 cases in mandible (33.33%). [Table 5]

[Table 5]- Total distribution of expansion, thinning and discontinuity of inferior border of mandible

	Frequency	%
Expansion and thinning	29	34.9
Discontinuity	11	13.3

Discussion:

Ameloblastoma is the most common odontogenic tumour accounting for about 1% of all tumours of head and neck region. [11]and about 11 % of all odontogenic tumours. [12,13] The word Amel means enamel and Blastos means germ or bud giving the meaning of Ameloblastoma as a tumour arising from tooth bud. [12] It was first recognised by Cusack in 1827. [12] The first neoplasm of this nature reported in scientific literature is credited to Broca in 1868. [1]

The first detailed description was given by Falkson in 1879. [1,5] The term Ameloblastoma was suggested by Churchill in 1934 to replace the term adamtinoma coined by Malassez in 1885. [1,5]

In the present study variants of solid type (58.09%) were of higher percentage followed by Unicystic (17.09%), desmoplastic (13.97%) and mixed variants (10.75%). This is in correspondence with studies by santanu patsa, Riteshkumar Baldevbhai Jadav et al., where among solid, unicystic and mixed variants solid variants were predominantly seen (66.21%) than unicystic(22.9%) and about 10 % of mixed variants [6] confirming our results. However the desmoplastic variant was not considered in their study. Among the solid types granular cell was of higher percentage (31.48%) followed by follicular (22.22%). This result is in contrast to the previous literature where follicular variant is frequently seen followed by plexiform. This may be due to constrained sample size of the present study or may be due to small chance of bias as some of the case reports have not been included in the present study because of the availability of only abstracts and the present study requires long term follow-up.

In this study solid, mixed and desmoplastic types most frequently occurred in 2nd – 5th decade of life. This corresponded with the analysis of Small and Waldron (1955) and Mehlisch, Dahlin and Masson (1972), of 1036 which asserted that ameloblastoma cases occurred most frequently in 20-50 years old patients. [14]Unicystic type was found to be common in the 2nd decade which is in the consistent with the literature where unicystic occurred in younger age group. [15,16] Gender distribution had inconsistent results in the previous literatures. Present study has no gender predilection which is in accordance with the studies of Keszler et al., and Takahashi et al.,[17] In the site distribution mandible was most commonly involved than maxilla which is in consistency with the retrospective study conducted by santanu patsa , Ritesh Kumar et al., on 148 patients. [6] Most ameloblastomas develop in the mandible (75%) posterior region usually the molar-ramus area and the remaining developed in the maxillary posterior region usually third molar area. 89.2% of the Ameloblastomas involving the mandible and maxilla in the present study occurred in posterior region with only few cases occurred in anterior region which corroborates with the findings obtained by Cosola et al.,[18] and Kim and jang.[19] According to worth, the most common radiographic appearance of ameloblastoma is multilocular radiolucency with corticated margins. [15] Present study is in correspondence with findings of worth. In the study conducted by Nigel.R.Figueiredo, Asit .D.Dinakar et al., in Goa among 11 cases 70% cases were with multilocular radiolucency and 30% cases were with unilocular radiolucency. Present study findings are in consistent with these findings with about 67.7% cases reported with multilocular radiolucency and 32.2% cases with unilocular radiolucency.

Present study reported 61.3% cases causing buccal cortical expansion with a predominance in mixed variant (80%). And about 47.3 % cases causing lingual cortical expansion with a predominance in variants of unicystic type (62.5%)and thinning of inferior border was noted in about 34.9% cases. This is in contrast to the study conducted on 11 cases in goa by Nigel.R.Figueiredo, Asit .D.Dinakar et al. where 100% cases showed buccal cortex expansion, 70 % cases showed lingual cortex with thinning of inferior border of mandible reported in 40% of the cases.[15]However predominance in particular variant has not been reported in this literature work. This contrast may be attributed to the large sample in the present study compared to the literature work.

Ameloblastomas are thought to have pronounced tendency to cause root resorption and displacement of tooth. [15] Root resorption was seen 39.8% cases and displacement of tooth was seen in 34.4% cases. According to white and pharaoh and Kim and Jang, around 10-14% are associated with non-erupted tooth. [15] This study confirms our results where 14% cases were associated with an impacted tooth.

Conclusion:

Since there are different histopathological variants of Ameloblastoma an extensive insight into the clinical behavior and radiological presentations was the need of the study.

From this study it was found that histopathological variants have unique characteristics that explains the importance of evidence based knowledge which gives oral physician an immense capability to diagnose appropriately so that accurate treatment can be advocated.

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