INTRODUCTION

Ameloblastomas are benign, locally invasive tumors of the jaws. They are relatively rare, accounting for about 1% of all oral tumors. Ameloblastomas usually present as a slow-growing painless swelling causing expansion of the cortical bone, perforation of the lingual or buccal plates and soft tissue infiltration. Peripheral ameloblastomas are extrasosseous versions of the lesion and constitute 1-5% of all ameloblastomas.

They originate from odontogenic epithelium and may arise from enamel organ, remnants of dental lamina, the lining of odontogenic cysts (dentigerous) or possibly from the basal epithelial cells of the oral mucosa.

In the jaw bones, ameloblastomas have a high predilection for the posterior mandible. Radiographically, 60% of ameloblastomas are multilocular. Published data differ considerably regarding the association between ameloblastomas and impacted teeth (15-40%).

ABSTRACT

Context: Ameloblastomas are benign tumors of the jaws with locally invasive capacity. Aim: The aim of this study was to review 112 cases of ameloblastoma seen over an 18-year period (1992-2009) at the Pernambuco Dental School, University of Pernambuco and at Federal University of Sergipe, in the northeast region of Brazil. Materials and Methods: The following data were selected for analysis: age, gender, race, site distribution, radiographic appearance, association with an impacted tooth, size, presence of symptoms, clinicopathologic subtypes and recurrence. Settings and Design: In this retrospective study, Pearson’s χ² test and t-test were employed. The critical level of significance was set at P < 0.05. Results: The mean age of the patients at presentation was 35.1 ± 16.8 years with a slight female preference. The peak prevalence was in the 11- to 20-year age group and declined with increasing age. Total 75 patients were black and 37 were white, for a 2:1 black: white ratio. The location of the ameloblastomas showed a marked predominance in the mandible (84.8%) and 69% of the cases presented with a multilocular radiographic appearance. The tumor was associated with an embedded tooth in 14 cases (12.7%): nine unilocular and five multilocular ameloblastomas. The maximum radiological extension of the lesions on panoramic radiographs was 0.5-20 cm (mean ± SD: 5.2 ± 3.3 cm) and most cases were symptom-free (75.9%). Solid/multicystic ameloblastoma was the most common clinicopathologic subtype. There was an association between the clinicopathologic subtypes and radiographic appearance (P < 0.001). Recurrence was observed in 13.3% of cases. Conclusion: We propose that racial factors may have strong influence on the incidence of ameloblastomas in the northeast region of Brazil, since most people have African descent. Data related to gender, location, radiographic appearance, size, symptoms, clinicopathologic subtypes and recurrence were similar to previous studies conducted in various parts of the world.

Key words: Ameloblastoma, Brazilian, epidemiology, odontogenic tumors
Treatment is primarily surgical and ranges from conservative (enucleation, curettage and cryosurgery) to radical (marginal resection, segmental resection or composite resection) modes. The rate of recurrence depends on the type of surgery and ranges from 15-25% after radical surgery to 75-90% after conservative surgical management. As ameloblastomas of the jaws are rare, it takes considerable time for any center to collect sufficient representative cases. The lesion is the most common odontogenic tumor in African and Asian populations, but not in European and American ones. The aim of this study is to present a review of 112 cases of ameloblastomas seen over a 18-year period (1992-2009) at the Faculty of Dentistry, University of Pernambuco and at Federal University of Sergipe, in the northeast region of Brazil.

MATERIALS AND METHODS

This was a retrospective study of ameloblastomas diagnosed at the Pernambuco Dental School, University of Pernambuco and at Federal University of Sergipe, in northeastern Brazil, from January 1992 to December 2009. For all cases, the clinical records were re-examined by the authors and two oral pathologists confirmed the type of tumor. This work was approved by the ethics committee of the University (nº 85/07).

The following data were analyzed: Age, gender, race, site distribution, radiographic appearance, association with an impacted tooth, size, presence of symptoms, clinicopathologic subtypes and recurrence. Data were analyzed using the SPSS for Windows (version 13; SPSS, Chicago, IL). Pearson’s $\chi^2$ test and t-test were employed. The critical level of significance was set at $P < 0.05$.

RESULTS

During the study period, 112 ameloblastomas were diagnosed. The patient group comprised 60 females and 52 males, for a female: Male ratio of 1.15:1. The mean age ($\pm$ standard deviation) of the patients at presentation was 35.1 $\pm$ 16.8 (range, 13 to 99) years. There was no statistical difference in the mean age between genders (females: 36.8 $\pm$ 18.6 years; males: 33.0 $\pm$ 14.2 years; t-test, $P = 0.262$). The peak prevalence was in the 11- to 20-year age group and declined with increasing age [Figure 1]. Total 75 patients were black and 37 were white, for a black: White ratio of 2:1.

Two cases (1.8%) were peripheral types. The location of the intraosseous ameloblastomas (98.2%) showed a marked predominance for the mandible (84.8%), while the maxilla was affected in 13.4% of the cases. In both jaws, the posterior region was the most affected site [Figure 2]. Radiographically, most cases showed a multilocular appearance (69%), whereas 31% were unilocular. An associate embedded tooth was observed in 14 cases (12.7%): Nine unilocular and five multilocular ameloblastomas ($\chi^2$ test, $P = 0.01$) [Figure 3].

The maximum radiological extension of the lesions on panoramic radiographs was 0.5-20 (mean $\pm$ SD: 5.2 $\pm$ 3.3) cm. Most patients had lesions from 3.0-6.9 cm in size (63.3%). The unilocular ameloblastomas were smaller than the multilocular ameloblastomas, but the difference was not statistically significant (unilocular: 4.5 $\pm$ 2.1 cm; multilocular: 5.6 $\pm$ 3.7 cm; t-test, $P = 0.171$). Most cases were symptom-free (75.9%).

Three clinicopathologic subtypes of intraosseous ameloblastomas were identified: Solid/multicystic ameloblastoma (SMA) ($n = 92$, 83.8%), unicystic ameloblastoma (UA) ($n = 17$, 15.3%) and desmoplastic ameloblastoma (DA) ($n = 1$, 0.9%). In SMA, the most common histologic pattern encountered was follicular [Figure 4], followed by plexiform [Figure 5], acanthomatous [Figure 6] and granular cell [Figure 7]. Of the UA, most cases were diagnosed histologically as luminal [Figure 8]. Total 64.5% of SMA had a multilocular appearance ($\chi^2$ test, $P < 0.001$) [Table 1].

Of the 112 patients, 15 (13.3%) experienced a recurrence of their tumor. The recurrence had developed from 3 months to 11 years (median 2 years; mean 3.2 $\pm$ 3.5 years) after initial treatment. Most cases of recurrent ameloblastomas were located on the posterior mandible (60%), had a multilocular radiographic appearance (93.3%) and were diagnosed as SMA (93.3%). Patients’ data are presented in Table 2.

DISCUSSION

Studies of diverse ethnic groups from various parts of the world have reported differences in the clinical features of ameloblastomas. In this study, the wide age range (13-99 years) was similar to reports worldwide, except in sub-Saharan African countries, where this lesion tends to occur in relatively young patients when compared with reports from white Africa, Asia, America and Europe. However, our experience concurs with a recent Brazilian study.
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which found the peak age of prevalence in the second decade of life. The higher prevalence of ameloblastomas in young people in reports from sub-Sahara Africa is either a true racial predilection or a reflection of the relatively younger population in developing countries. Studies using mitochondrial DNA showed that about 30% of Brazilian self-classified as white and 80% of Brazilian blacks carry maternal lineages typical of sub-Saharan Africa. In the northeast Brazil i.e. Pernambuco State, 92% of the people are Afro-descendants. These data suggest a strong influence of racial factors on the incidence of ameloblastoma. In fact, in our study, the occurrence of ameloblastomas was twice as high in blacks as in whites.

Most studies have reported an equal gender distribution of ameloblastomas, but a male preponderance was reported in Nigerian, Egyptian, and Indian.
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Figure 8: Histological section showing a unicystic ameloblastoma with luminal pattern (H&E stain, ×200 original magnification)

Table 1: Relationship between radiographic appearance and clinicopathologic subtypes of intraosseous ameloblastomas

| Clinicopathologic subtypes | Radiographic appearance (%) | n (%) |
|----------------------------|-----------------------------|-------|
|                            | Unilocular                  | Multilocular |
| SMA                        |                             |       |
| Follicular                 | 08 (7.3)                    | 26 (23.6) | 33 (30.0) |
| Plexiform                  | 08 (7.3)                    | 14 (12.7) | 23 (21.0) |
| Follicular and plexiform   | 04 (3.6)                    | 21 (19.1) | 25 (22.7) |
| Acanthomatous              | 01 (0.9)                    | 06 (5.5)  | 07 (6.5)  |
| Granular cell              | -                           | 02 (1.8)  | 02 (1.8)  |
| Plexiform and acanthomatous| -                           | 02 (1.8)  | 02 (1.8)  |
| UA                         |                             |       |
| Luminal                    | 07 (6.5)                    | 03 (2.7)  | 10 (9.0)  |
| Intraluminal               | 04 (3.6)                    | 01 (0.9)  | 05 (4.5)  |
| Mural                      | 02 (1.8)                    | -        | 02 (1.8)  |
| DA                         | -                           | 01 (0.9)  | 01 (0.9)  |
| Total                      | 34 (31.0)                   | 76 (69.0) | 110 (100) |

SMA: Solid/multicystic ameloblastoma; UA: Unicystic ameloblastoma; DA: Desmoplastic ameloblastoma

Chinese populations. A remarkable female predominance was observed in the West of Scotland. In our series, there was an almost equal distribution between the genders, with a slight female preponderance. Although the age difference between genders was not statistically significant, males were slightly younger than females, which corroborates the findings of Adeline et al. In contrast, in another study, women were 4 years younger than men when their ameloblastomas were diagnosed.

Regarding the anatomic site of occurrence, ameloblastoma has a high predilection for the mandible. The posterior region of both jaws is the area most affected. Interestingly, anterior lesions are seen more frequently in blacks compared to Caucasians and Asians. Sawyer et al. also noted this predisposition among blacks, although it was not seen in our series. Ameloblastomas of the maxilla present special difficulties in terms of treatment. Indeed, most patients in some countries present with large tumors and surgery often involves resection of the tumor with healthy bone, leaving the patients with large anatomical defects and marked disfigurement.

Radiographically, an ameloblastoma can be a multilocular or unicystic cyst-like lesion. The observation that the most radiographic appearance was the multilocular type (69%) is consistent with other reports, but it is debatable whether this is an indication of more aggressive behavior since a unicystic radiographic appearance is not synonymous of a UA. By contrast, Kim et al. founded that approximately 60% of ameloblastomas had a unicystic radiographic appearance and indicated that cyst-like lesions may be treated conservatively with enucleation and/or curettage whenever all areas of the cyst lumen can be controlled intraoperatively.

According to Kim et al., postoperative follow-up is important in the management of ameloblastoma because more than 50% of all recurrence occur within 5 years of surgery. However, based on our findings, the recurrence of ameloblastomas can occur as early as 2 years after initial treatment. Most cases of recurrent lesions observed in our study were multilocular SMA subtype and diagnosed histologically as follicular and/or plexiform pattern, similar to the findings by Nakamura et al. and Kim et al. Therefore, the present results indicate the need of radical surgery when treating ameloblastomas with these features.

In addition, a higher proportion of unicystic lesions were associated with an impacted tooth, mimicking a dentigerous cyst. According to Philipsen and Reichart, the unicystic pattern is more common in the unicystic variant than in the multilocular, especially in cases associated with tooth impaction. UAs and dentigerous cysts have similar clinical and radiographic appearance, and the histological distinction between these lesions can be difficult. Therefore, although some authors have suggested that UAs develop via the cystic degeneration of solid ameloblastomas, there are indications that some ameloblastomas may arise in a dentigerous cyst in which the neoplastic ameloblastic epithelium is preceded temporarily by a non-neoplastic stratified squamous epithelial lining.

Regarding tumor size, both the multilocular and unicystic variants may become very large, resulting in airway compromise and potentially life-threatening metabolic abnormalities. However, few patients present with symptoms other than a slow-growing swelling.
in this study. Consequently, patients generally tend to seek medical advice only when a deformity is evident.

**CONCLUSION**

In summary, we proposed that racial factors strongly influence the incidence of ameloblastomas in northeast Brazil, since most people in this area are of African descent. The findings related to gender, location, radiographic appearance, size, symptoms, clinicopathologic subtypes and recurrence showed similarities with studies conducted in other parts of the world.

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