

Ameloblastoma in Jamaica – Predominantly Unicystic: Analysis of 47 Patients over a 16-Year Period and A Case Report on Re-entry Cryosurgery as a New Modality of Treatment for the Prevention of Recurrence

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ABSTRACT

Objective: To analyse all the cases of ameloblastoma seen in the two major public Hospitals in Jamaica over a 16-year period, and to compare the results with what has been previously documented by other authors in the literature. A new treatment modality for prevention of recurrence of ameloblastoma is described.

Subjects and Methods: The case files of patients histologically diagnosed to have ameloblastoma at both the Kingston Public Hospital and Cornwall Regional Hospital in Jamaica from 1980 to 1995 were retrieved and information about this odontogenic tumour was documented. The analysis revealed that unicystic ameloblastoma was predominant (95%) and solid ameloblastoma was about 5%. It was not therefore surprising that the average age in the study was 29.1 years, with a peak in the 10–19-year age group. No peripheral ameloblastoma was diagnosed.

Results: A total of 47 new cases of ameloblastoma was recorded between 1980 and 1995. This number accounted for 16.03% of all jaw bone lesions in Jamaica and 38.2% of jaw bone lesions of odontogenic origin during this period. These cases of ameloblastoma accounted for 67% of odontogenic tumours with the exception of the odontomas. The mean age was 29.1 (range 13–67) years. The male: female ratio was 1: 1.14). Only 3 cases involved the maxilla. Of significance, is the case of bilateral ameloblastoma with no continuity to the anterior region, and also the case of unilateral involvement of the mandible and the maxilla in the same patient. Re-entry cryosurgery (a procedure in which cryosurgery is done after a specified period from the primary surgical procedure) after excisional biopsy of a relatively small cystic ameloblastoma of the maxilla was negative for recurrence.

Conclusion: The clinicopathological presentation of ameloblastoma in Jamaica is not different from what has been documented by other authors throughout the world; however, we report two unique cases – a case of bilateral ameloblastoma of the mandible and a case of unilateral involvement of the mandible and maxilla in the same patient. We suggest that re-entry cryosurgery before recurrence be considered in the management of ameloblastoma for the prevention of recurrence.

Keywords: Ameloblastoma, cryosurgery

El Ameloblastoma en Jamaica – Predominantemente Unicístico: Análisis de 47 Pacientes por un Período de más de 16 Años y un Reporte de Caso de Criocirugía de Reentrada Como Nueva Modalidad de Tratamiento Para Prevenir la Recurrencia

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RESUMEN

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Objetivo: Analizar todos los casos de ameloblastoma vistos en los dos principales hospitales públicos en Jamaica durante un periodo de 16 años, y comparar los resultados con la información previamente documentada por otros autores en la literatura. Se describe una nueva modalidad de tratamiento para prevenir la recurrencia del ameloblastoma.

Sujetos y método: Se recuperaron los expedientes de los pacientes diagnosticados histológicamente con ameloblastoma en el Hospital Público de Kingston y el Hospital Regional de Cornwall en Jamaica desde 1980 a 1995, y se obtuvo información documentada acerca de este tumor odontogénico. El análisis reveló que el ameloblastoma unicístico era predominante (95%), en tanto que el ameloblastoma sólido fue de alrededor del 5%. Por lo tanto, no fue sorprendente que la edad promedio en el estudio fuera 29.1 años, con un pico en el grupo etario de 10 a 19 años. No se diagnosticó ningún ameloblastoma periférico.

Resultados: Entre 1980 y 1995, se registró un total de 47 nuevos casos de ameloblastoma. Este número representaba el 16.03% de todas las lesiones óseas de mandíbula en Jamaica y 38,2% de las lesiones de hueso de la mandíbula de origen odontogénico durante este período. Estos casos de ameloblastoma representaron el 67% de los tumores odontogénicos con la excepción de los odontomas. La edad promedio fue 29.1 años (rango 13–67). La proporción hombre:mujer fue de 1: 1.14. Sólo tres casos implicaron el maxilar. Resulta particularmente importante el caso del ameloblastoma bilateral sin ninguna continuidad a la región anterior; así como el caso de la participación unilateral de la mandíbula y el maxilar en el mismo paciente. La criocirugía de reentrada (un procedimiento en el cual la criocirugía se realiza después de un periodo especificado a partir del procedimiento quirúrgico primario) después de la biopsia por escisión de un ameloblastoma quístico del maxilar relativamente pequeño, fue negativa para la recurrencia.

Conclusión: La manifestación clínico-patológica del ameloblastoma en Jamaica no difiere de lo que ha sido documentado por otros autores en todo el mundo. Sin embargo se reportan dos casos únicos: un caso de ameloblastoma bilateral de la mandíbula y un caso de compromiso unilateral de la mandíbula y el maxilar en el mismo paciente. Sugerimos que se tenga en cuenta la criocirugía de reentrada a la hora de tratar el ameloblastoma para prevenir la recurrencia.

Palabras claves: Ameloblastoma, criocirugía

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INTRODUCTION

The preponderance of ameloblastoma in West Africa has been documented in previous reports (1–3). This odontogenic tumour is said to be uncommon in the Caucasian population (4, 5). Anand *et al* (6) concluded that the apparent high incidence of ameloblastoma in West Africa, could be the result of a harvesting phenomenon (that is a collection of cases). Recent studies on jaw-bone tumour in Jamaica revealed that ameloblastoma is the most common odontogenic tumour (7), and this is similar to the outcome of other studies (8–13) emanating from West Africa.

Robinson and Martinez in 1977 (14) documented their findings on a subtype of ameloblastoma which they designated as unicystic ameloblastoma. They suggested that the unicystic variant is less aggressive than its solid counterpart and as such favoured simple enucleation as the treatment modality. They further documented that unicystic ameloblastoma occurs more in the 3rd decade of life whilst its solid counterpart occurred more in the 4th decade of life. In their paper, they mentioned that unicystic ameloblastoma is usually associated with an unerupted tooth and that unicystic ameloblastoma are more commonly found in the posterior

mandible followed by the parasympheseal region, anterior maxilla and posterior maxilla.

This study which was conducted to establish the incidence and other clinicopathologic aspects of ameloblastoma in Jamaica is the only such study in the English-speaking Caribbean.

SUBJECTS AND METHODS

The case file of patients histologically diagnosed to have ameloblastoma of the jaw bone in Jamaica were retrieved from the records department of both the Cornwall Regional Hospital and Kingston Public Hospital in Jamaica, and data collected of patient age, gender, radiographic appearance and clinical behaviour of the lesion.

At the time of study (April 1995), records were only readily available from 1980, hence a 16-year study period was conducted. The radiographic appearance and clinical findings were based mainly on what was documented in the patient's case file by the radiologist or the attending maxillofacial surgeon. Radiographs for about 35 patients were actually reviewed.

The existing legislation of the Ministry of Health of Jamaica on the retention of medical records and radiographs allows an active storage period of 5 years for radiographs; this is the reason for not viewing all the radiographs for the 47 patients analysed, however, the radiographic information in the patients case files was adequate. No record was duplicated. A separate case of unicystic ameloblastoma of the maxilla treated with re-entry cryosurgery is reported.

CASE REPORT: Re-entry cryosurgery

A 51-year old Jamaican black female presented to the Cornwall Dental Centre in Montego Bay with mild pain in the anterior region on the right side of the maxilla. On examination, no clinical swelling was present. An orthopantomogram revealed a well-defined radiolucent lesion (Fig. 1)

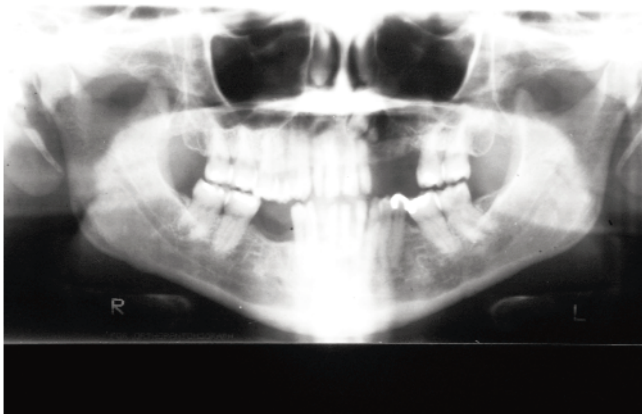


Fig. 1: Orthopantomogram preoperatively showing unilocular radiolucency of the anterior maxilla.

surrounding a tooth or a denticle, apical to the upper left lateral incisor. The cyst outline was moderately corticated with evidence of root resorption of the upper lateral incisor on the left side. The cyst measured about 7 mm x 6.5 mm on radiograph. The differential diagnosis included dentigerous cyst, ameloblastoma and odontogenic keratocyst. Because of the small size of the lesion an excisional biopsy with adequate margin (definitive treatment) was done without an initial incisional biopsy, bearing in mind that the lesion, may be an ameloblastoma or odontogenic keratocyst rather than a dentigerous cyst.

The pathologists report was as follows:

Gross appearance: *The specimen consists of three fragmented and friable haemorrhagic tissue, which is admixed with a 1.7 x 0.6 x 0.4 cm tooth.* **Histology:** *The submitted tissue showed a small multiloculated cystic structure amidst a mixture of fragmented bone, cartilage and mucosal soft tissues representing the wall of the maxilla. The cyst is lined by ameloblastic epithelium and surrounded by a densely fibrous stroma containing scattered islets of epithelium. The overall picture is that of an ameloblastoma, so-called unicystic type (follicular variant) as shown in Fig. 2.*

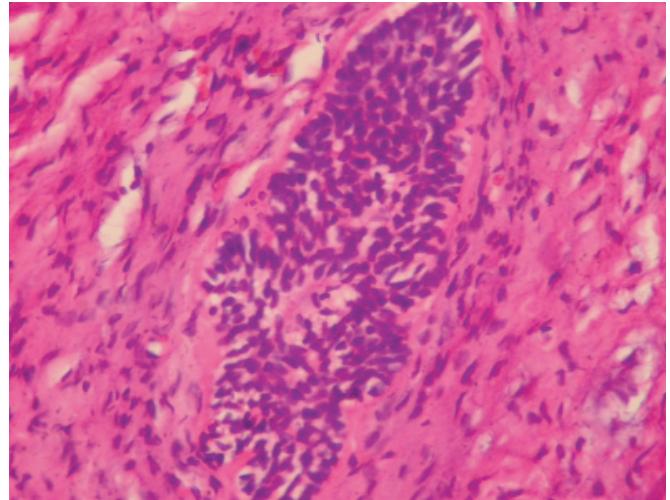


Fig. 2: Histology of case shown in figure 1 demonstrating follicular ameloblastoma.

The pathologist further commented on the fact that these so-called unicystic ameloblastomas are reported to be less aggressive in growth, and that enucleation rather than resection is the recommended treatment.

Based on the histological diagnosis of ameloblastoma (of a lesion which was initially diagnosed on clinical and radiological grounds to be a dentigerous cyst prior to complete excision) it was decided to do an early radiological follow-up with CT-scan at 6-months post surgery. Although the CT-scan at 6-months could not confirm the presence of recurrence (Fig. 3), as one could not exclude the possibility



Fig. 3: Post-operative follow-up CT-scan of patient at 6 months.

of pathologic tissue in the post surgical defect, it was suggested to the patient to opt for re-entry cryosurgery and biopsy of any residual pathological tissue. This re-entry cryosurgery was done one year after the excisional biopsy. At re-entry surgery, prior to the application of liquid nitrogen, the upper lateral incisor on the left side (which was mobile) was extracted and all the soft tissue (Fig. 4) in the healing



Fig. 4. Picture of the enucleated soft tissue and tooth removed during re-entry surgery.

cavity was curetted and preserved for histopathology.

The pathologists report is as follows:

Gross appearances: *The specimen consists of 4 irregular portions of friable to haemorrhagic tissue ranging in size from 0.4 x 0.2 x 0.2 cm. Separately submitted in the same container is a 2 x 0.5 x 0.5 cm incisor tooth.*

Histology: *The biopsy has been examined in toto and at multiple deeper levels. It showed portions of fibrovascular tissues with skeletal muscle and fragments of bone, which are significant only for scarring and aggregates of chronic inflammatory cells, inclusive of multinucleated giant cells disposed focally within the fibrous tissue. There is no evidence of recurrence of tumour.*

RESULTS

Over the 16-year period, ameloblastoma accounted for 16.03% of all jaw- bone tumours in Jamaica. Out of a total of 123 cases of jawbone lesion of odontogenic origin, ameloblastoma accounted for 38.2%. It accounted for 67% out of 70 cases of odontogenic tumours, except odontoma's.

The histopathological documentation revealed that 95% of cases of ameloblastoma in Jamaica are unicystic. Twenty-seven (57%) of the total cases of ameloblastoma presented as multilocular radiolucencies with root resorption in the majority of cases (Fig. 5). Others (43%) were either unilocular with root resorption or large and extensive with no internal septa, but radiographically well defined. To be more

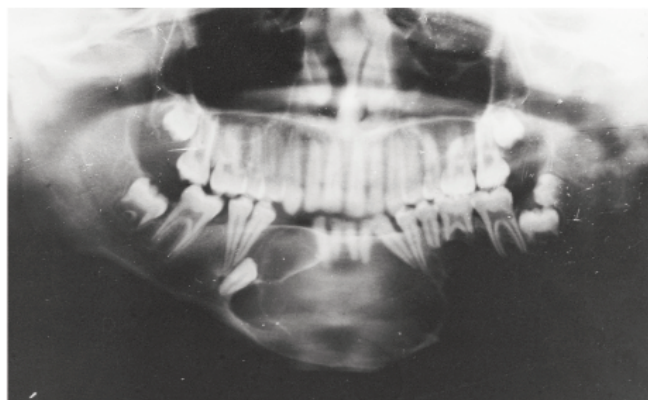


Fig. 5: Orthopantomogram of a 14-year old boy showing a well demarcated, partially corticated, expansive multilocular radiolucency of the mandible.

precise, of the 43%, 27% were large and expansive and in most cases affected the ascending ramus of the mandible. Three cases involved the maxilla of which one case (case 13) also involved the mandible on the same side – a case of unilateral involvement of both maxilla and mandible by ameloblastoma in the same patient. Forty-four cases (93.6%) involved the mandible variably thus consistent with what has been previously documented by other authors. Twenty-one cases (44.7%) presented in both the posterior and anterior mandible. For all these presentations we would prefer to use the term “radiographic site of ameloblastoma” which we have consistently found to be more extensive than the clinical site.

One case (case 8) presented bilaterally and almost symmetrically in the posterior mandible and without any continuity of the separate posterior lesions to the anterior mandible, as can be seen in the oblique lateral view of the mandible (Fig. 6). From these two radiographic views it was

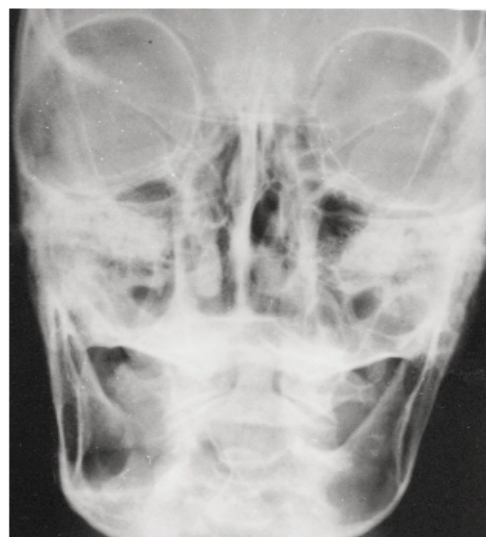


Fig. 6: (a) Posterior anterior view of the mandible showing bilateral ameloblastoma in a 65-year old black Jamaican female.



Fig. 6: (b) the oblique lateral view shows that the right and left lesions are not really communicating.

difficult to ascertain if these lesions were actually multilocular.

Twelve cases, however, presented bilaterally and with continuity radiographically within the anterior mandible. For these set of cases, it would appear that these lesions actually started from one side and extended anteriorly to involve the anterior mandible and in some cases the posterior mandible on the other side (Fig. 5).

Six cases (12.8%) were associated with an unerupted permanent tooth (Fig. 5), thus stressing the significance of a dentigerous lesion (lesions associated with an unerupted tooth). One of these six cases presented with an unerupted lower central incisor in association with a lesion found to have extended from 36 to 46 radiographically. Case 11 was seen to have a denticle (simple odontoma) associated with the base of the lesion (Fig. 7) which further makes one appreciate the possible cystic degeneration and ameloblastoma formation in association with an odontome.

It is a very common finding that the multilocularity of a lesion is absent on an extra – oral radiograph, such as an oblique lateral of the mandible, but can be confirmed by an occlusal radiograph (Fig. 8). In this particular patient, a calcified matter simulating the appearance of a simple odontome can be seen associated with the lesion.

Of the total cases of ameloblastoma, (47) 38.3% occurred in teenagers and of these 15% presented in children between the ages of 13 and 15 years.

DISCUSSION

The most significant clinicopathologic finding of ameloblastoma of the jaw bones in Jamaica is its enormous extent, both clinically and radiographically (7). This enormous extent is however, to be found in the majority of African countries such as Nigeria (8–13).

In most large series (18, 19), about 80% of the tumours occur in the posterior mandible with extension to the ramus.



Fig. 7: Oblique lateral view of the mandible of a 24-year old black Jamaican female showing a well-defined extensive radiolucent lesion with evidence of root resorption of 44, 45 and 46. The radiolucency is associated with a buried denticle or simple odontome.

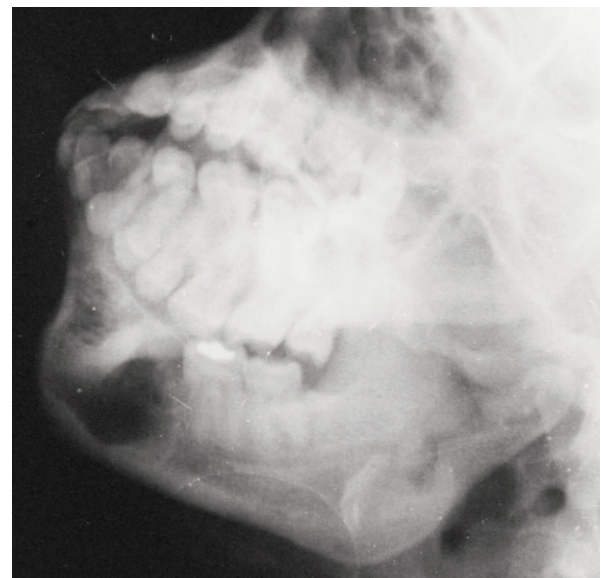


Fig. 8: (a) The multilocularity of this lesion was not seen on a lateral oblique view.

The premolar and symphyseal areas may also be affected. These previous findings can be compared with those of the current series of 47 cases, with 93.6% of the tumour being present in the posterior mandible, 12.3% in the anterior

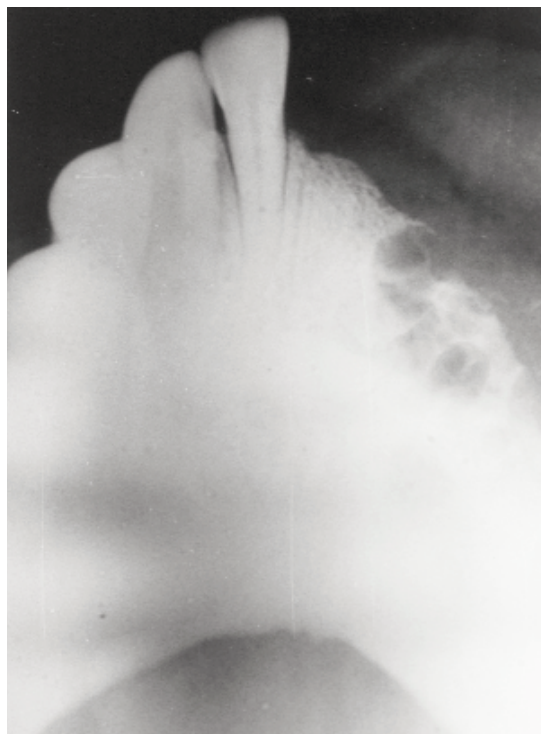


Fig. 8: (b) Occlusal radiograph showing multilocularity of the lesion.

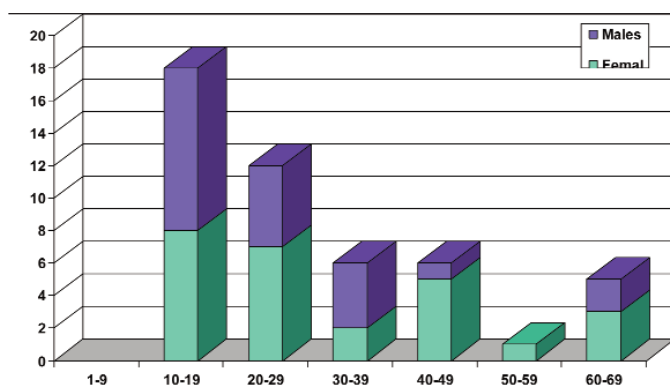


Fig. 9: Histogram showing gender and age distribution of ameloblastoma in Jamaica.

mandible and 29.8% in both the anterior and posterior aspects of the mandible.

From this study, ameloblastoma seems to be the most frequent of all jaw bone tumours and odontogenic tumours in Jamaica, and this may be due to the larger percentage of blacks of African origin in Jamaica relative to other ethnic groups. It is very important to recognize the fact that this tumour is more frequent in Africans than in Caucasians (8–13), but this may not necessarily be due to any genetically determined difference. It may be due to environmental factors.

The report of the survey of the literature in 1937 of 379 cases of ameloblastoma (15) have contributed significantly to

current knowledge of this tumour. Later, a survey by Small and Waldron of 984 published cases (16) and 62 unpublished ones showed results very similar to those of Robinson (15) in the clinical features of ameloblastoma. The relatively more recent series, such as the 126 cases reported by Mehlich and his co-workers (17), the 92 cases reported by Sehdev and his associates (18), the 78 cases reported by Regezi and his colleagues have all exhibited data essentially identical with those of previously reported series.

In the present series, we recorded an average age of 29.1 years with a peak between the age of 10 and 19-years (Fig. 8). Other clinical features, apart from the age seems to be consistent with what has been previously documented (15–18). It is our opinion that the occurrence of ameloblastoma is approximately evenly divided between the genders, and the slight difference noted is insignificant. The occurrence of ameloblastoma in a larger number between the ages of 10 and 19 years in our series would appear to be significant (Fig. 9).

We have noticed the term “cystic ameloblastoma” on the histopathology reports of approximately 95% of the cases of ameloblastoma in Jamaica.

We are uncertain if the remaining 5% are actually all solid ameloblastoma, however, no peripheral ameloblastoma was documented.

The average age of occurrence for unicystic ameloblastoma is generally thought to be earlier than that of the more solid form and tends to occur more in the mid-twenties. There is also a slightly higher incidence in males than females (19). The unicystic ameloblastoma is almost exclusively found in the mandible although there have been a few recorded cases in the maxilla. The molar area in the mandible is the most common site with two-thirds of the lesions occurring in this region (20). It is said to be frequently associated with the impacted third molar. In his classical paper, Eversole further stated that unicystic ameloblastomas have 6 basic patterns seen radiographically. These radiological patterns are all well defined radiolucencies. These patterns can be divided into two groups as thus:

- (I). *Radiologic patterns that are seen associated with mandibular third molar:*
 - a. Unilocular pericoronal radiolucency measuring less than 20 mm.
 - b. Large unilocular expansile radiolucency extending up to the coronoid process and notch.
 - c. Large expansile radiolucency with scalloped margins.
- (II) *Radiological pattern not associated with a third molar.*
 - a. Expansile unilocular periapical radiolucency associated with root resorption.
 - b. An inter-radicular pear-shaped radiolucency with adjacent root divergence.
 - c. A multilocular periapical radiolucency with root resorption.

Without any doubt, the features of root resorption and displacement is a strong radiological indication of a neoplastic process. All these radiological patterns have been seen in our current series of 47 new cases.

Three histological subsets of unicystic ameloblastoma were classified in 1995 by the World Health Organization (21). These histological subsets are as follows:

A relatively innocuous lining which may give way in parts to cell changes specific for ameloblastoma. A nodule may project into the lumen of a cyst and examination of the nodule exhibits a plexiform pattern. The so-called mural type where part of the wall of the cyst is infiltrated with typical plexiform or follicular ameloblastoma.

Because groups 1 and 2 lesions are said to be non-aggressive, enucleation would be a sufficient treatment modality (21), whereas group 3 lesions with mural invasion should be treated aggressively as case 3 which is shown radiologically in Figure 4. Fortunately, the definitive diagnosis was made pre-operatively rather than retrospectively as is usually the case following microscopic evaluation of the entire surgical specimen. As such, the treatment of this case was radical (mandibular resection with reconstruction with corticocancellous autogenous bone graft from the iliac crest). The treatment of ameloblastoma in Jamaica varied from enucleation with or without cryosurgery to block resection with or without reconstruction with autogenous corticocancellous bone graft from the iliac crest. Recently Rosenstein *et al* stated that cystic ameloblastoma are frequently multilocular and exhibited clinical aggressiveness (21). They re-evaluated the so called "unicystic ameloblastoma" through assessment of clinical features, histological patterns and proliferative indices in a series of 21 patients. They further compared the proliferative capacity of the unicystic tumour with the solid ameloblastoma and dentigerous cyst by assessment of immunohistochemical stain for cell-cycle associated protein, k-1 67.

Seven cases recurred (one of these recurred seven times despite its continuous benign nature on histology). The factor responsible for recurrence is variable and is not necessarily dependent on the modality of treatment. This will be explored in a subsequent report.

CONCLUSION

The clinicopathologic findings of ameloblastoma in Jamaica are similar to those previously documented by other authors

throughout the world except for a peak incidence between the ages of 10 and 19 years. Further, studies into the histological types of ameloblastoma need to be conducted and this correlated with recurrence rate irrespective of the modality of the treatment. Finally re-entry cryosurgery *can only be suggested* at this stage as a new modality of treatment as the only documented case so far has too short a period post-cryosurgery to conclusively claim prevention of recurrence.

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