Review of Six Cases of Maxillary Ameloblastoma from the West Indies
Re-entry Cryosurgery as Prophylactic Surgical Intervention
C Ogunsalu1, E Scipio1, N Williams2

ABSTRACT

Maxillary ameloblastoma is a rare histopathological entity. A total of six cases of histologically confirmed maxillary ameloblastoma from the West Indies is reviewed. Three of the cases were taken from a total of 47 histologically confirmed ameloblastoma over a 15-year period (1980−1995) from two major maxillofacial units in Jamaica. Two other cases were from documentation in Jamaica between 2000 and 2002, one of which occurred in a 13-year old girl (these two patients have been followed-up periodically to 2006). The sixth case was from the records of the maxillofacial department of the University of the West Indies in Trinidad and Tobago. This last patient, at a recent review, has inoperable recurrence.

These cases were reviewed with respect to demographics (patient’s age and gender), location and extent of tumour, radiological features, concurrent involvement of the mandible, treatment with special emphasis on current treatment modality and follow-up. The findings do not differ from what has been documented by other authors from other parts of the world.

Because of the radiographic anatomy of the maxilla, recurrence may be detected late despite such occurring earlier following initial surgical management. It is for this reason that we suggest re-entry cryosurgery for prevention of recurrence for maxillary ameloblastoma. The only case of maxillary ameloblastoma that had re-entry cryosurgery continues to benefit from absence of recurrence at periodic follow-ups at four years post-primary surgical management (which was enucleation).

Revisión de seis Casos de Ameloblastoma Maxilar de Criocirugía de Reentrada de West Indies como Intervención Quirúrgica Profiláctica
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RESUMEN


Estos casos se examinaron con respecto a los datos demográficos (la edad y el género del paciente), la situación y magnitud de tumor, rasgos radiológicos, compromiso concurrente de la mandíbula, tratamiento con énfasis especial en la modalidad del tratamiento actual y el seguimiento. Los hallazgos no difieren de lo documentado por otros autores de otras partes del mundo.

Debido a la anatomía radiográfica del maxilar superior, la recurrencia puede ser detectada tarde, a pesar de que ocurra temprano tras el tratamiento quirúrgico inicial.

Es por esta razón que sugerimos la criocirugía de reentrada para prevenir la recurrencia del ameloblastoma maxilar. El único caso de ameloblastoma maxilar que tuvo criocirugía de reentrada...

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INTRODUCTION
Maxillary ameloblastoma is a rare histopathological entity, although ameloblastoma (mandible and maxilla) is the most common odontogenic tumour (1–4). This tumour arises from odontogenic epithelium of embryonal toothelement and is most frequent in the mandibular molar region, with 20% occurring in the maxilla (1–10).

While ameloblastomas, in general, are considered benign but locally invasive neoplasms, the maxillary ameloblastoma is a more aggressive and persistent lesion, presumably because the thin and fragile bone of the maxilla, unlike the thick cortical plates of the mandible, allows a relatively unimpeded spread of the tumour to the surrounding structures, such as the maxillary sinus, nasal cavity, orbit and occasionally the cranial base (11–13).

The rich blood supply to the maxilla relative to the mandible may also contribute to the accelerated spread of this locally aggressive neoplasm. The maxillary ameloblastoma is more cellular than the mandibular counterpart and is further characterized by less distinct peripheral palisading, fewer columnar cells, frequent focal acanthomatous metaplasia and more cellular stroma.

Ameloblastoma, a benign but locally invasive and persistent polymorphic neoplasm of odontogenic origin, usually has a follicular or plexiform pattern, lying in a fibrous stroma (14). Broca is credited for the first scientific description of ameloblastoma in 1868. Subsequently, Falkson in 1897 coined the term adamantinoma with the current term ameloblastoma introduced by Churchill in 1929 (15).

It is more common in the third to fifth decades and shows no gender predilection. While the numbers in this report are small, except for one paediatric case, the cases are demographically consistent with those reported in the literature. Eighty per cent of ameloblastomas occur in the mandible and the rest in the maxilla. In the maxilla, the expansion can be quite extensive before diagnosis since the tumour can spread into the maxillary sinus, without detection. Occasionally, pterygomaxillary spread may be present and this may make surgical management very technical, with total clearance of tumour almost impossible without damage to vital anatomical structures. None of the cases in this series involved the orbital floor, presumably as a result of relatively early detection.

In this paper, six cases from the West Indies are reported. The report emphasizes detection and a new treatment modality called re-entry surgery with liquid nitrogen, for prevention of recurrence, and long term radiological follow-up with advanced imaging technique in the management of maxillary ameloblastoma.

SUBJECTS AND METHODS
Only three cases of maxillary ameloblastoma out of a total of 47 cases of ameloblastoma were seen over a 16-year period (1980–1995). Additionally we included two cases of Maxillary Ameloblastoma seen between 2000 and 2002. The sixth case was from the maxillofacial surgery department of the University of the West Indies at the Eric Williams Medical Complex in Trinidad and Tobago.

The clinical and radiographic features, including the outcome of treatment from long-term follow-up are reviewed and analysed with respect to demographics (patient’s age and gender), clinical signs and symptoms, location and extent of tumour, radiographic features (including CT-scan findings), treatment modality and outcome of long-term follow-up post treatment.

RESULTS
There were three males and three females. Five of six patients were adults, mean age of 44.8 years (range 31–58 years). The sixth patient was a 13-year-old girl (Table 1).

A slowly enlarging painless, disfiguring maxillary mass was the main clinical feature in four of six cases. In Case 5, detection was a chance finding on both periapical radiograph and on orthopantomograph following a complaint of pain only without intraoral or extraoral swelling. Case 4 also presented with nasal obstruction.

Five out of six cases involved both the anterior and posterior maxilla (Fig. 1). One case (Case 5) involved the anterior maxilla only (Fig. 2). The case of maxillary ameloblastoma in the child (Case 4) also resulted in extensive destruction of both the nasal cavity and maxillary sinus (Fig. 1).

The first three cases in Table 1 also caused varying destruction of the maxillary sinus. None of the cases involved the orbital floor. Case 1 also involved the posterior mandible on the same side – a case of unilateral involvement of the maxilla and mandible in one patient.

The radiological presentation included: multilocular radiolucency in Case 1, cystic/unilocular radiolucency in Case 2, obliteration of the nasal cavity in Cases 3 and 4, and unerupted teeth and denticle associated with radiolucent lesions in Cases 4 and 5. Tooth displacement and root resorption were also radiographic findings in all cases where teeth were involved. The radiological differential diagnoses were other locally aggressive jaw lesions such as the odontogenic keratocyst, myxoma and fibroma.

Treatment
The surgical management for the current series of cases of maxillary ameloblastoma is shown in Table 1. Cases 1 to 3...
have not presented with recurrence to date. Case 4 has been followed clinically from the time of surgery (January 2000) up to 2006 with no clinical evidence of recurrence. However, for economic reasons she has not been able to do CT scans which has been recommended for radiological follow-up. To date the cosmetic result of the surgery is satisfactory (Fig. 3).

Case 5 has been followed up both clinically and radiographically by CT scan. The post-operative CT scan at six months post-surgery did not suggest the presence of recur-

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**Table 1: Summary of cases of ameloblastoma of the maxilla in the West Indies**

<table>
<thead>
<tr>
<th>Case</th>
<th>Sex</th>
<th>Age (yrs)</th>
<th>Site</th>
<th>Radiographic appearance</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M</td>
<td>43</td>
<td>Anterior maxilla (also posterior mandible on the same side)</td>
<td>Multilocular radiolucency which is well defined, with evidence of resorption of teeth in both lesions.</td>
<td>Enucleation and removal of affected teeth</td>
</tr>
<tr>
<td>2</td>
<td>M</td>
<td>31</td>
<td>Anterior and posterior maxilla</td>
<td>Cystic radiolucency overlying the maxillary antrum</td>
<td>Enucleation and removal of affected teeth</td>
</tr>
<tr>
<td>3</td>
<td>F</td>
<td>40</td>
<td>Anterior and posterior maxilla</td>
<td>Obliteration of the right maxillary sinus.</td>
<td>Enucleation and removal of affected teeth</td>
</tr>
<tr>
<td>4</td>
<td>F</td>
<td>13</td>
<td>Anterior and posterior maxilla</td>
<td><strong>CT Scan Findings</strong> A 4.7 x 3 x 4.3 cm hypodense mass filling and expanding the left maxillary antrum. The mass also erodes the bony walls inferiorly through the hard palate and superior alveolar wall processes, medially into the nasal processes and anterolaterally into the subcutaneous tissues. The mass does not enhance and no calcification is seen within it.</td>
<td>Enucleation and removal of affected teeth and inferior turbinate</td>
</tr>
<tr>
<td>5</td>
<td>F</td>
<td>52</td>
<td>Anterior maxilla</td>
<td>Well defined radiolucent lesion surrounding a tooth and a denture apical to 12 region.</td>
<td>Excisional biopsy enucleation patient scheduled for secondary cryosurgery although no evidence of recurrence</td>
</tr>
<tr>
<td>6</td>
<td>M</td>
<td>58</td>
<td>Posterior maxilla</td>
<td>See Case Report</td>
<td>Partial maxillectomy after multiple curettage type procedures</td>
</tr>
</tbody>
</table>

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Fig. 1a: CT scan showing both anterior and posterior involvement by ameloblastoma in a 13-year old child (case 4). Note the destruction of the lateral wall of the nasal cavity on the left side.

Fig. 1b: CT scan showing buried tooth within the tumour mass.
rence (Fig. 4). This patient was advised of the need and possible advantage of re-entry surgery with application of liquid nitrogen (cryotherapy), although no recurrence was noted. Excision biopsy had been done for her intraosseous dentigerous lesion without prior histological knowledge that the lesion was an ameloblastoma or a dentigerous cyst histologically. The histopathological examination revealed an apical mass admixed with fragments of bone, cartilage and respiratory-type mucosa from the antrum. The cyst was lined by attenuated odontogenic epithelium which was also evident as epithelial islands within the densely fibrotic wall in keeping with unicystic ameloblastoma (Fig. 5). Central apical degeneration was present in the epithelial nests consistent with the follicular variant. The significance of a regular follow-up has also been stressed to this patient.

Case 6 is a 58-year-old Trinidadian male who was histologically diagnosed with ameloblastoma in the left posterior maxilla, post-extraction of the upper left third molar. The socket apparently did not heal and between 1985 and 1999 he had multiple curettage-type procedures without resolution of the lesion. In 1987, a hemi-maxillectomy was done and obturation was satisfactory post-surgery. The lesion, which was a follicular type ameloblastoma, however recurred in 1999 in the pterygoid region (Fig. 6). Due to various circumstances including management of prostatic cancer, this patient has been unable to access re-entry surgical intervention so far.

The postoperative follow-up CT scan six months after treatment of Case 5 is shown in Fig. 4. Radiologically, 3-mm axial images were obtained of the maxilla without contrast. There was a 1-cm diameter defect in the left anterior portion of the maxilla, presumably the surgical site. There was mild prominence of the adjacent soft tissue which is probably scarring. There was no evidence of a pathologic fracture. There was no lymphadenopathy. There was a small fluid level in the maxillary sinus.

Although there was no indication of recurrence in this case, re-entry surgery with curettage and cryotherapy with liquid nitrogen, as prophylactic surgical intervention before recurrence, was suggested.

Clinical Case Report: re-entry Cryosurgery
A 51-year old female Jamaican of African descent presented to the Cornwall Dental Centre in Montego Bay with slight
pain in the upper anterior region on the right side. On examination, no clinical swelling was present. An ortho-
panthomogram revealed a well-defined radiolucent lesion (Fig. 1) surrounding a tooth or a denticle apical to the upper
left lateral incisor. The cyst outline was moderately corti-
cated with evidence of root resorption of the upper left lateral incisor. The cyst measured about 7 mm x 6.5 mm on radi-
graph. The differential diagnoses included dentigerous cyst, ameloblastoma and odontogenic keratocyst. Because of the small size of the lesion, an excisional biopsy (definitive treat-
ment) was done without an initial incisional biopsy, bearing in mind that the lesion maybe an ameloblastoma or odonto-
genic keratocyst rather than a dentigerous cyst. Histopatho-
logical confirmation of ameloblastoma was made (Fig. 5).

Based on the histological diagnosis, it was decided to do an early radiological follow-up with CT scan at six months. Although the CT scan at six months showed no evidence of recurrence (Fig. 2) it was suggested to the patient that a re-entry cryosurgery procedure and biopsy of any residual pathological tissue be done. 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 left lateral incisor which was mobile, was extracted and curetted, and all the soft tissue (Fig. 3) in the healing cavity preserved for histopathology. There was no evidence of recurrence on histological evaluation.

**DISCUSSION**
The maxillary ameloblastoma can be solid or multicystic, unicystic, peripheral or malignant. The solid or multicystic type is the classical ameloblastoma and the other three are variants (16). Most cases of maxillary ameloblastoma occur in the posterior part of the maxilla (11, 16) and are potentially more troublesome than the mandibular ameloblastomas. The mandibular ameloblastoma, unlike the maxillary counterpart, can be readily diagnosed before it advances. However maxil-
ary ameloblastoma often reaches a considerable size before diagnosis. This is because of anatomical factors. The maxil-
lar sinus plays an important role in the nature of spread and size of the tumour before detection. The pain related to Case 5 was most likely due to the further intraosseous expansion and pressure effect on the apicies of tooth # 21. Her nasal cavity on the right side was also always stuffy. Hence as previously documented, localized painful facial enlargement with nasal obstruction may be the characteristic clinical presentation of maxillary ameloblastoma when compared to mandibular ameloblastoma, which typically presents as a painless mass (5, 8, 9).

As ninety per cent of the maxillary ameloblastomas are found in the posterior maxilla with extension into the pterygomaxillary area, the infra-temporal fossa and the floor of the middle cranial fossa, there may be a problem in the management of this tumour. The suggested resection margin of 10 mm may not be achievable in the maxilla without damage to vital anatomic structures. The radiological pre-
sentation of Cases 4 and 5 further emphasizes the signifi-
cance of a dentigerous lesion. The treatment of Case 4 was based on accurate detection of the extent of the tumour by CT scan. This confirmed that the tumour involved both the
anterior and posterior maxilla with extension to the nasal cavity.

The type of ameloblastoma was not indicated, however, because of the prevalence of the plexiform type of ameloblastoma in children, which is thought to be less aggressive than the follicular type (17). The treatment of this child was that of initial conservative treatment involving curettage of the lesion with reasonable margins, together with the removal of the inferior turbinate which was affected by the tumour. A long term follow-up with advanced imaging was suggested.

Clinical follow-up of this patient up to four years post-surgery revealed no evidence of recurrence. The cosmetic appearance was very satisfactory despite the extent of surgery (Fig. 3). Unfortunately, the CT scan or MRI at two years interval has not been done, because of lack of funds. This patient was advised that clinical follow-up alone is inadequate to survey for recurrence. Radiological follow-up advanced imaging rather than conventional imaging – is so important as it will detect recurrence prior to it being clinically evident. The choice of radiological follow-up for this patient was CT scan, because it is less expensive than MRI which has the advantage of no exposure to ionizing radiation and also better detection of soft tissue recurrence.

The treatment of Case 5 was initially that of an excisional biopsy of a radiologically confirmed dentigerous lesion of the anterior maxilla, which was later confirmed histologically as maxillary ameloblastoma. A postoperative CT scan at six months revealed no evidence of recurrence (Fig. 3). It was suggested that the patient opt for a re-entry surgery with cryotherapy. This treatment modality is “the prophylactic re-entry cryosurgical intervention for the prevention of recurrence” we advocate in the management of all maxillary ameloblastomas, as this will prevent or limit the extent of recurrence.

CONCLUSION

Maxillary ameloblastoma is a rare histopathological entity which is treatable with early detection, appropriate surgical removal and utilisation of cryotherapy. Long term clinical and radiological follow-up should be emphasized to both clinicians and patients. Re-entry surgery with liquid nitrogen application may play a great role in prevention of recurrence of this tumour.

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REFERENCES