The pattern and occurrence of ameloblastoma in adolescents treated at a university teaching hospital, in Kenya: A 13-year study

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ABSTRACT

Ameloblastoma presenting in the adolescent age group is rare with few studies documenting their occurrence.

Aim: The aim of this study was to carry out an analysis of the pattern and occurrence of ameloblastoma in those less than 20 years of age.

Materials and method: Patients from the University of Nairobi Dental teaching Hospital treated for ameloblastoma were included in the study over a 13-year period. The study highlights the demographic, clinic-radiographic and histologic features of benign locally aggressive lesions.

Results: A total of 127 patients were recorded of which, 27 (21.3%) were below the age of 20 years; no case was reported below the age of 10 years. 18.5% were below the age of 14 years and 81.5% were 15–19 years old. The gender predilection was ~1:1. All of the tumours occurred in the mandible, with radiographic features of a multilocular radiolucencies (85.2%); and a fewer unilocular lesions (14.8%). The management is in a staged-wise approach: resection and/or disarticulation with temporary reconstruction using mandibular stainless steel or titanium plates and delayed bone grafting.

Conclusion: The occurrence of ameloblastoma can mimic an odontogenic cyst, clinicians therefore need to be vigilant when examining adolescents so that conservative treatment is started early in order to reduce the subsequent morbidity.

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1. Introduction

Ameloblastoma is an aggressive benign slow growing epithelial odontogenic tumour and comprises of only 1% of all tumours and cysts of the jaws (Posnick, 2000). It is the most clinically significant odontogenic tumour. A prospective study done in Tanzania of odontogenic tumours reported, ameloblastoma occurring with the highest frequency followed by odontogenic myxoma (Simon et al., 2005).

Ameloblastoma occurs in three different clinicoradiographic patterns, the conventional intraosseous solid/multicystic (86%), unicystic (13%) and peripheral (1%) (Neville et al., 2002). Three histopathological variants of unicystic are luminal, intraluminal and mural (Ackermann et al., 1988). The common histological patterns include the follicular and plexiform patterns as the most common. However, less common histopathologic patterns, acanthomatous, granular cell, desmoplastic and basal cell types have also been reported (Neville et al., 2002). The conventional solid ameloblastoma is encountered over a wider age range being commonly reported in the 3rd–5th decade of life.

The occurrence of ameloblastoma in younger age group is considered a rarity and it accounts for approximately 10–15% of all reported cases of ameloblastoma (Keszler and Dominduez, 1986; Ueno et al., 1986). The tumour is even less common in children younger than 10 years of age and relatively uncommon in the 10–19-year-old age group with no significant gender predilection. A higher frequency in Blacks has been reported whereas other studies show no racial predilection. About 85% of conventional ameloblastomas occur in the mandible, most often in the body and angle-ascending ramus area (Neville et al., 2002). About 15% occur in the maxilla, usually in the posterior region. The most typical radiographic feature is that of a multilocular radiolucency ("soap bubble") appearance accompanied by resorption of the roots of teeth. The unicystic ameloblastoma is common in younger patients with 50% occurring in the second decade of life.

There is a paucity of information especially from East Africa and Kenya, regarding the incidence of ameloblastomas in the young
population of less than 20 years. These tumours cause disfigurement of the face, leading to poor self-esteem and interruption in schooling. In addition, it also causes difficulty in maintaining oral hygiene, as well as feeding leading to malnutrition. This paper highlights the age, gender, site predilection, histopathological and radiographic features of ameloblastoma in patients aged 10–19 years seen at the University of Nairobi (UON) Dental Hospital. The resulting information will assist surgeons in swift and efficient management of such tumours in the young age group before they reach a large size.

2. Materials and method

The data for this audit was obtained from the records of all the cases of ameloblastomas treated at the UON Dental teaching Hospital over a period of 13 years (1996–2009). This is a centre of excellence for Oral and Maxillofacial Surgical training in East Africa. The majority of such cases present to the department of Oral and Maxillofacial surgery for management from all over the country. The inclusion criterions were patients in the adolescent age group (between the ages of 10 and 19 years of age), histopathological confirmation of ameloblastoma and those with complete records. Treatment consisted of marginal resection (and disarticulation where necessary) with subsequent reconstruction using stainless steel or titanium plates. The records were examined by one investigator (Butt F.M.A) and the following information sought; demographic characteristics such as age, gender, site of lesion, radiographic and histopathological information. Radiographic findings were recorded from Orthopantomograms (OPGs), Computed Tomographic (CT)-scans and 3-D reformatted images. Histopathology slides retrieved from the laboratory were examined by two independent Oral pathologists. Patients with incomplete records and those with a disputed histopathologic findings were excluded from the study. Post-operatively the patients are under close up for the next 15–20 years to observe for any recurrences.

3. Results

During the 13-year period, a total of 127 patients were treated for ameloblastoma of the oral and maxillofacial region. The male to female ratio was ~1:1. The majority of patients (90.6%) were within the age bracket 10–49 years. The modal age group was 20–29 years (Table 1). 27 (21.3%) patients were under 20 years of age and of these 14 were males and 13 females giving a male: female ratio ~1:1. The average age of presentation was 16.1 years. All the lesions were noted to have occurred in the mandible with none in the maxilla. Only five (18.5%) of the 27 patients were under 14 years of age, while 22 (81.5%) of them were between the ages of 15–19 years. None of the patients were under 10 years old. The ratio of males to females in the 10–14 and 15–19-year-old age brackets was ~1:1 similar to that in the total population (Table 2). The youngest patient was a 10-year-old and the oldest 19 years of age. The most frequently affected age group of patients with ameloblastoma was 18 and 19 years (44.4%), with the modal age group being 18 years (25.9%).

Extra-oral features noted were facial swelling with asymmetry. Intra-orally, there was cortical expansion, mobility of teeth and occlusal derangement (Fig. 1). 16 patients had involvement of the body/ramus with extension into coronoid and condyle, seven had only the body and ramus affected and the remaining four had the symphysal and body areas affected (Fig. 2). Common radiological features exhibited by the majority (85.2%) were the typical “soap bubble” (multilocular) appearance that is characteristic of solid ameloblastomas and the minority (14.8%) were unilocular radiolucencies. In addition there was root resorption and tooth displacement. In 11 (40.7%) patients, there was an associated impacted tooth (Fig. 3). A computed tomogram (CT) formatted as an axial image showed bucco-lingual expansion and perforation of the lingual cortex (Fig. 4). The treatment modality consisted of

![Fig. 1. (A) Clinical features: extra-oral – facial swelling (B) intra-orally – bucco-lingual expansion.](https://example.com/fig1.jpg)

**Table 1**

<table>
<thead>
<tr>
<th>Age range of ameloblastoma in the study population.</th>
<th>Males</th>
<th>Females</th>
<th>Males + females (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age bracket (years)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>10–19</td>
<td>14</td>
<td>13</td>
<td>27 (21.3)</td>
</tr>
<tr>
<td>20–29</td>
<td>23</td>
<td>21</td>
<td>44 (34.6)</td>
</tr>
<tr>
<td>30–39</td>
<td>12</td>
<td>15</td>
<td>27 (21.3)</td>
</tr>
<tr>
<td>40–49</td>
<td>10</td>
<td>5</td>
<td>15 (11.8)</td>
</tr>
<tr>
<td>50–59</td>
<td>0</td>
<td>5</td>
<td>5 (3.9)</td>
</tr>
<tr>
<td>60–69</td>
<td>2</td>
<td>3</td>
<td>5 (3.9)</td>
</tr>
<tr>
<td>70–79</td>
<td>1</td>
<td>1</td>
<td>2 (1.6)</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td>64</td>
<td>63</td>
<td>127 (100)</td>
</tr>
</tbody>
</table>

**Table 2**

<table>
<thead>
<tr>
<th>Distribution of adolescents with ameloblastoma by age and gender.</th>
<th>Male</th>
<th>Female</th>
<th>Total (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>10–14</td>
<td>3</td>
<td>2</td>
<td>5 (18.5)</td>
</tr>
<tr>
<td>15–19</td>
<td>11</td>
<td>11</td>
<td>22 (81.5)</td>
</tr>
<tr>
<td><strong>Total</strong></td>
<td>14</td>
<td>13</td>
<td>27</td>
</tr>
</tbody>
</table>

resection and reconstruction using non-vascularized iliac bone graft secured with stainless steel or titanium plates. 16 (59.3%) patients had resection with disarticulation due to the extension of the tumour into the coronoid and condyle (Fig. 5).

Three histopathological variants of ameloblastoma were noted in our study: follicular (77.8%), plexiform (18.5%) and desmoplastic (3.7%) (Fig. 6). The male to female ratio of the follicular and plexiform was ∼1:1; there was only one patient (female) with the desmoplastic type (Fig. 7). Each of the subtypes exhibited some degree of microcystic degeneration within the ameloblastic islands; this did not dominate the overall histological characteristics.

For patients who presented with large, extensive tumours involving the condyle, coronoid and overlying soft tissues, resection with disarticulation was done regardless of it being unicocular or multicocular. It is in our protocol to try and eliminate the tumour in our first attempt and adhering to the practice of leaving a 2 cm safety margin of cancellous bone after resection as has been advocated (Muller and Slootweg 1985). Reconstruction was done immediately with a condylar prosthesis on a stainless steel or titanium plate (Fig. 8). In cases where there was spread of the tumour from the mandible into the soft tissues, they were sacrificed to prevent the development of peripheral ameloblastoma. Excess facial skin was contoured for cosmesis before closure. There were two patients who developed post-operative infection which was resolved with course of broad spectrum intravenous antibiotics. The patients who have been followed so far have not presented with recurrence of the ameloblastoma (Fig. 9).

4. Discussion

In 1955, Waldron and Small documented a review of 1,036 ameloblastomas in which the average patient age was 38.9 years, with only 2.2% under 10 years old and 8.7% between 10 and 19 years old (Small and Waldron, 1955). Reichart in his biological profile of 3677 cases reported that in the developing countries, ameloblastomas occur in younger patients (average age = 27.7 years) compared to those from industrialized countries (average age = 39.1 years). As per the racial distribution the average age of Blacks was 28.7 years compared to 39.9 years in Caucasians and 41.2 years in Asians (Reichart et al., 1995) Kesler and Dominquez reported that 8.7% less than 16 years of age, whereas Kahn found 12.2% of ameloblastomas were in patients younger than 19 years (Kahn, 1989; Kesler and Dominquez, 1986). Mehlisch et al. found 12.6% in those aged less than 20 years but, only one patient was under 10 years of age, while Sandler et al. found 15% of their patients under 20 years old and 3% to be under 10 years old (Mehlisch et al., 1972; Sandler et al., 1983). Patients from Asian and African countries show a higher percentage of children with ameloblastoma to be younger than 20 years with 18.2% for Japanese patients and 19.7% for Thai patients (Sirichitra and Dhiravangkura, 1984; Ueno et al., 1986). A recent study done in China documented a series of 27 patients of which only 8.1% were under the age of 11 years and 91.9% were between the ages of 11 and 20 years (Zhang et al., 2010). In Nigeria, 25% of children under the age of 20 years show an increased frequency of ameloblastoma (Arotiba et al., 1997). In Zimbabwe, Chidzonga studied 117 patients over a period of 10 years who were treated for ameloblastoma and reported 17.1% were under 18 years of age, the youngest being 11 years: the average age at presentation being 15.5 years (Chidzonga, 1996).

In our study the incidence of ameloblastoma in the 10–19 years old relative to all cases of ameloblastoma in all age groups was 21.3%. In comparison to other studies done in Africa, it shows a similarity to the one reported by Arotiba et al. (21.9%), and a bit higher than the 17.1 % reported by Chidzonga (Chidzonga, 1996; Arotiba et al., 2005). The gender predilection shows a male to female ratio of 1:1, which is similar to that reported by Arotiba as 1.3:1. Our finding of equal gender distribution has also been reported by other authors (Arotiba et al. 2005). Available statistical evidence suggests that this tumour is more common in the Black population; in our study all the patients were Blacks of African decent (Shear and Singh, 1978; Sawyer et al., 1985).

The rarity of this tumour in the younger age group is demonstrated by our study findings in which only 18.5% were younger than 14 years of age with the majority being in the older group, and the youngest patient being 10 years. This is in accordance with...
previous reports of the low occurrence of this tumour in the first decade of life. In our study we had two 10-year-old patients (7.4%) which is higher than the 4.7% reported by Arotiba et al., and Adekeye whereas, Chidzonga reported none in his group (Adekeye, 1980; Chidzonga, 1996; Arotiba et al., 2005). The most frequently affected age group of patients with ameloblastoma was between 18 and 19 years (44.4%), the majority being 18 years (25.9%) This is close to the one reported in Nigeria in which the modal age was 17 years (17.5%) (Arotiba et al., 2005).

The most prevalent site of occurrence of ameloblastoma is the mandible in all races (Potdar, 1969; Huffman and Thatcher, 1974; Adekeye, 1980; Olaitan and Adekeye, 1997). Reichart observed that in Blacks, ameloblastomas affected the incisor region of the mandible significantly more often than in the white population (Reichert et al., 1995). The anatomic location of ameloblastoma in our study was mainly in the posterior ramus (85.2%). The unilocular type was less common (14.8%) mainly located in the ramus and body of the mandible and associated with an impacted tooth. However in Whites the unicystic subtype has been reported as the most common in those less than 20 years of age (Robinson and Martinez, 1977; Leider et al., 1985; Zhang et al., 2010).

Histopathologically, some studies have shown that the plexiform type of ameloblastoma is more common than the follicular type in children and is thought to be less aggressive hence a conservative treatment can be considered (Shear and Singh, 1978; Sawyer et al., 1985; Matsuo and Ueno, 1991). In contrast, our findings showed the follicular (77.8%) to be most common followed by the plexiform (18.5%) and then the desmoplastic (3.7%) The mean age of patients presenting with the follicular and plexiform was 16 years and there was only one patient, a 15-year-old boy, with the desmoplastic type. These results are consistent with those of another study done in Kenya by Vilembwa et al. who in a study of clinicopathologic features of ameloblastoma, reported, that patterns of the follicular and plexiform types were predominant (Sirichitara and Dhiravarangkura, 1984; Vilembwa et al., 2008).

The management of ameloblastoma in the younger age group can be very challenging despite using a reconstruction plate, as this has a profound effect on the subsequent growth dynamics of the craniofacial skeleton, dentition and soft tissues that ensues. Conservative management for unicystic ameloblastomas involves enucleation, curettage and use of physico-chemical methods (dredging and carnoy’s solution). However, high recurrence rates of up to 60% have been noted which is similar to the rates when solid and multicystic ameloblastomas are enucleated (Small and Waldron, 1955; Ueno et al., 1986; Ueno et al., 1989; Li et al., 2000; Lau and Samman, 2006; Vilembwa et al., 2008).

In our study population, most of the ameloblastomas are at an advanced state and these lesions present with mural invasion and
possible involvement of cancellous bone. This hardly leaves room for conservative methods which are indicated for small sized and unicystic tumours. Interestingly, it has been reported that ameloblastomas in patients from the developing countries are larger compared to those from the industrialized countries with Blacks having larger dimensions than Caucasians (Reichart et al., 1995). In the majority of cases extensive surgery is the management of choice in our centre. Lesions smaller in size are accessed using an intra-oral approach. For large tumours, extending to the condyle and coronoid, an extra-oral approach is used. In our teaching hospital patients are encouraged to adhere to a regular long-term follow-up in order to detect any recurrences. A staged-reconstruction method using a non-vascularized free bone graft is preferred, whereby the primary reconstruction with a bone graft is secured with a mandibular reconstruction plate as shown in our studies. A one-stage reconstruction using a vascularized bone graft is often lengthy and costly for our patients. In addition to challenging socioeconomic conditions, difficulties arise in close follow-up, given the distances patients have to travel to hospitals and the cost for multiple procedures and their poor patient compliance. Due to limited resources and general poverty, this forces the surgeon to resect and reconstruct in the shortest time possible. These are some of the main reasons against adopting conservative measures involving decompression and enucleation with peripheral ostectomy for paediatric and/or adolescents patients as reported by in recent study on Taiwanese patients (Huang et al., 2007).

Post-operative difficulties for these young individuals include, interference with craniofacial growth dynamics and function (mastication and speech), disturbance in psychological well-being and their progression in education. So far, there have been no recurrences in this study: this could probably be attributed to our treatment protocol as mentioned earlier.

It is recommended that multi-centre prospective studies should be done in our region to record the histopathological types of ameloblastoma and age of presentation with detailed emphasis on post-operative craniofacial growth dynamics of those who are less than 14 years of age.

5. Conclusion

The occurrence of ameloblastoma can mimic an odontogenic cyst, clinicians therefore need to be vigilant when managing adolescents with such lesions so that conservative approaches involving decompression and/or enucleation and peripheral ostectomy can be considered. As long as the lesions are detected early, that is before the destruction of the lower border, perforation of the buccal and lingual cortices and involvement of the condyle or coronoid regions of the mandible, much of the anatomy can be preserved during surgery and this will reduce the subsequent morbidity affecting young patients.

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Conflict of interest statement
None

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References


Fig. 8. Treatment: extensive surgery involving mandibulectomy with disarticulation and reconstruction using a stainless steel plate.

Fig. 9. Showing orthopantomogram of an 18-year-old patient at different ages. (A) Pre-operative (B) 2-months post-operative with bone graft (C) 4-years post-operative showing complete healing of the bone graft.