Conservative Management of Pediatric Unicystic Ameloblastoma in a Secondary Healthcare Facility in an Ameloblastoma's High Burden Sub-Saharan Africa: A Report of Two Cases

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ABSTRACT

Background: Surgical resection is the primary treatment for ameloblastoma but often causes facial disfigurement and complications. Conservative management of unicystic ameloblastoma (UA) may prevent these issues. Combining marsupialization, enucleation, and Carnoy's solution adjuvant therapy restores jawbone health and prevents complications.

Presentation: Two pediatric cases—a 13-year-old boy with a 3-month right mandibular ramus swelling and an 11-year-old boy with a 5-month anterior mandibular swelling—presented at General Hospital Ifako-Ijaiye. Clinical and investigative findings confirmed unicystic mural ameloblastoma.

Intervention: Both underwent marsupialization followed by enucleation with Carnoy's solution. Preoperative orthopantomograms showed unilocular radiolucent lesions; postoperative imaging revealed restored bony trabeculae.

Outcome: Successful treatment minimized negative psychosocial impact on the patients with no recurrence after one year of ongoing monitoring. There was no clinical or radiographic evidence of recurrence at 1-year follow-up. Planned surveillance per IAOMS guidelines (≥5 years for mural UA) and prosthesis to restore esthetics and function.

Conclusion: Conservative UA management with regular follow-up prevents facial disfigurement and complications associated with resection, improving quality of life. These are Nigeria's first documented pediatric UA cases.

Keywords: unicystic ameloblastoma, marsupialization, enucleation, Carnoy's solution.

INTRODUCTION

Ameloblastoma is a slow-growing, benign but locally aggressive odontogenic tumor that leads to progressive jaw bone destruction.[1] It is sub-Saharan Africa's most common odontogenic tumor, accounting for 1% of oral and 18% of odontogenic tumors; it has male predilection.[1,2] Ameloblastomas are classified clinically into conventional, extra-osseous/peripheral, metastasizing, or unicystic ameloblastoma (UA).[2] UA represents between 5–15% of ameloblastomas, mimicking odontogenic cysts radiographically but shows ameloblastic proliferation histopathologically.[2] UA is less aggressive than conventional ameloblastoma and greater than 90% occurs in the posterior mandibular area.[2] Its recurrence rate is 10–25% and because of its biologic behavior conservative approaches (marsupialization ± enucleation ± Carnoy's solution) become a viable option.[3,4] We report two conservatively managed mural UA cases from a secondary healthcare facility in Lagos, Nigeria. Informed consent was obtained for treatment and publication; anonymity was ensured.

CASE 1

A 13-year-old boy presented (12th January, 2024) with a 3-month right mandibular swelling of 12×8 cm in diameter causing disfigurement and social distress. Tenderness and swelling extended from the right mandibular body to the ramus. Intraoral examination revealed buccal expansion from the first molar to the ramus with "ping-pong" consistency. Aspiration yielded grayish-straw fluid. Orthopantomogram (OPG) showed a unilocular radiolucency with root resorption in the lower right mandible (Fig. 1). Differential diagnoses included odontogenic keratocyst, dentigerous cyst, and UA. Incisional biopsy confirmed mural UA (cystic lining with ameloblastomatous transformation, cuboidal peripheral cells, and stellate reticulum-like cells; epithelial projections into connective tissue indicated mural infiltration). Marsupialization was performed, followed by enucleation 4 months later

with meticulous Carnoy's solution application (Fig. 2). The cavity was dressed with Framycin Sulphate gauze until closure. Medications: Dalacin C (300 mg 12-hourly), ibuprofen (400 mg PRN), Astyfer syrup (15 ml 12-hourly). One-year postoperative OPG showed restored bony trabeculae (Fig. 3) and healed mucosa (Fig. 4).

Figure 1: Panoramic view of the jaws revealing a multilocular radiolucent bone lesion with resorption of the root of the lower right mandibular first molar (case 1).

Figure 2: Intra-operative picture during procedure of the enucleation of the cystic lesion (case 1)

Figure 3: Panoramic view of the jaws revealing a healthy right mandibular bone with an erupting molar tooth a year post-operative (case 1)

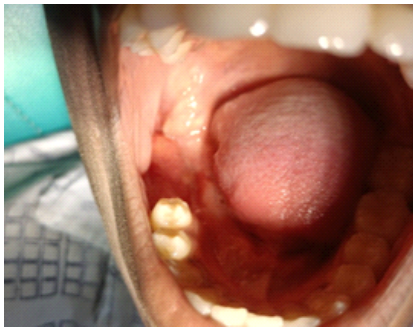

Figure 4: Post-operative picture showing the healed oral mucosa in the lower right mandible (case 1)

CASE 2

An 11-year-old boy presented (3rd February, 2024) with a 5-month anterior mandibular swelling causing disfigurement measuring 8×5 cm in its widest diameter. Tenderness spanned the parasymphyseal region. Intraoral examination revealed labio-lingual swelling; serosanguinous fluid was aspirated. OPG revealed a radiolucent lesion with an erupting tooth and cortical plate expansion (Fig. 5). Differential diagnoses included odontogenic keratocyst, dentigerous cyst, and UA. The initial and final biopsies revealed islands of ameloblastomatous proliferation of cuboidal to low columnar peripheral cells and central stellate reticulum-like cells infiltrating within cystic walls and into connective tissue stroma, thus confirming mural UA. Marsupialization was performed, followed by enucleation 3 months later with Carnoy's solution application; affected teeth including the one at the inferior border of the mandible were extracted (Fig. 6). The cavity was irrigated and dressed with Framycin Sulphate gauze until closure (Fig. 7). Medications included Dalacin C (150 mg 12-hourly), ibuprofen syrup (200 mg PRN), and Astyfer syrup (10 ml 12-hourly). One-year postoperative OPG showed restored bony trabeculae (Fig. 8). There are plans to restore function and aesthetics with prosthetic replacement of the missing teeth.

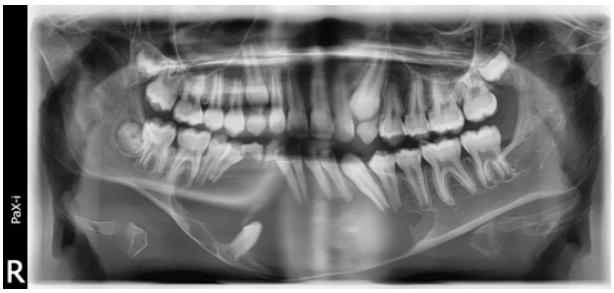

Figure 5: Panoramic radiograph revealing unilocular osteolytic radiolucency of the anterior mandibular area (case 2)

Figure 6: Intra-operative picture immediately after enucleation of the cystic lesion in the anterior mandibular area (case 2)

Figure 7: Post-operative picture of the healed oral mucosa in the anterior mandibular area (case 2)

Figure 8: A panoramic view of a healthy anterior mandibular bony trabeculae a year post-operative (case 2)

Timeline:

Event	Time
Follow-up:	Ongoing (1+ year)
Histopathologic diagnosis	September 2024
Enucleation	June -July 2024
Marsupialization	March 2024
Incisional biopsy	February 2024
Presentation:	January-February 2024

DISCUSSION

Findings: Robinson and Martinez first described unicystic ameloblastoma (UA) in 1977 as a distinct variant[5] with clinical/radiographic features resembling odontogenic cysts but histopathologic ameloblastic proliferation. Ackermann et al.[6] later classified three subtypes based on ameloblastic cell infiltration: Mural (60%) with cells invading the fibrous cystic lining; Luminal (30%) with cell proliferation confined to the cyst lining; and Intra-luminal (10%) with tumorous-cell nodules projecting into the cyst cavity.[2,5] In these present cases, the aspirates from the UA were straw-colored in case I and serosanguinous in case II, aligning with reports from a Nigerian study.[7] Both affected male pediatric patients, corroborating previous reports[3,6,8] that UA mostly affects younger patients with higher male preponderance, though Ahmed[2] reported equal sex predilection with a mean age of 32.8 years. The two cases showed different radiographic appearances; case I presented with a multilocular radiolucency while case II had a unilocular radiolucency. These observations align with reports by Meshram et al.[6] and Sinduja & Ramani,[8] confirming radiographic unpredictability in UA's presentation.

Treatment remains controversial. The radical approach (resection), preferred for conventional ameloblastoma, minimizes recurrence (3%)[2] but causes facial disfigurement, tooth loss, and masticatory dysfunction. Conversely, conservative management for UA—including marsupialization, enucleation, and Carnoy's solution application—reduces morbidity.[2] Carnoy's solution alone lowers recurrence to 16% versus 30.5% for enucleation alone.[5] It was first clinically used by Cutler and Zollinger in 1933.[2] Aside from reducing recurrence rate, Carnoy's solution provides critical advantages such as bone penetration (1.54 mm depth), optimized hemostasis, and rapid fixation/elimination of residual ameloblastic cells.[2] Its adjunctive use in oral and maxillofacial surgery after enucleation is well-established for UA and keratocysts.[2]

In the current report, we adopted a combination of marsupialization, enucleation, and application of Carnoy's solution. Our method is similar to that adopted by several previous workers.[6,8,9] The choice of procedure was influenced by the strong preference for conservative management and aversion to surgery by the patients' mothers, their willingness to commit to long-term follow-up, and concerns about the psychological impact of jaw resection on their children. This perspective aligns with reports by Meshram et al.[6] and Demir & Gunhan[5] and supports our rationale that a conservative surgical approach is justified. We considered the risk reasonable given pediatric patients' superior osteogenic regenerative capacity compared to adults.[6,9] Hence, the observed favorable clinical outcome eliminates the risks of permanent growth plate disruption, loss of regenerative periosteum, and impaired quality of life associated with radical surgical approaches.[6,9,10]

Furthermore, our approach of initial marsupialization to decompress the mandible, followed by enucleation and application of Carnoy's solution, achieved excellent bony regeneration (Figs. 4 & 8), even in the thinned anterior mandible in Case II. We attribute this to meticulous application of Carnoy's solution, its penetration of osteolytic bone,[2,6] and careful avoidance of nerve injury, as evidenced by the absence of inferior alveolar/mental nerve deficits.[2] While UA mostly arises in the posterior mandibular area and less frequently in the

anterior region, our cases confirm that any area of the mandible may be involved, consistent with previous studies.[2,3,5] The erupting teeth in both cases suggest that UA might have developed from dentigerous cysts around the crown of erupting teeth, confirming that 50–80% of UA cases are of dentigerous cyst origin.[2,6,8,11]

Implications: To our knowledge, these are Nigeria's first documented pediatric UA cases. Both occurred in young males (ages 11 and 13 years) with a mean age of 12 years, corroborating reports by Meshram et al.[6] that the mean UA age is approximately 13 years. This further strengthens reports that UA has a predilection for younger age groups[6,7] despite controversial sex distribution.[2,6,8] Also, while UA prevalence is 80% in the posterior mandible and 20% in the anterior area,[2,3,5] Case II highlights diagnostic variability. Probably the most important implication of our report is the viability of conservative UA management, the benefits of preserving post-surgery quality of life, and its financial relief in a resource-limited setting like ours—provided long-term follow-up is assured.

Trade-Offs (Limitations): Our 1-year recurrence-free period is promising but short. Mural UA (the most aggressive subtype) requires ≥5-year surveillance per IAOMS guidelines. We continue monitoring these patients.

Take-Home (Conclusion): Conservative UA management minimizes disfigurement while preserving quality of life—particularly crucial in pediatric patients. Our cases demonstrate that meticulous enucleation + Carnoy's solution enables excellent bony regeneration even in high-risk scenarios (anterior location, cortical thinning).

Expectation for Future Research: Prospective studies with longer follow-up (≥5 years) in larger cohorts to establish definitive recurrence rates.

Recommendation: We recommend conducting larger multicenter trials with long-term follow-up to compare conservative versus radical outcomes in both pediatric and adult UA patients.

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