CASE REPORT

Unicystic ameloblastoma with mural proliferation: conservative or surgical approach?

Soumi Samuel,¹ Freddy K Mistry,¹ Shilpa Chopra,¹ Ajay Pillai²

¹Department of Oral and Maxillofacial Surgery, AB Shetty Memorial Institute of Dental Sciences, Mangalore, Karnataka, India

²Department of Oral and Maxillofacial Surgery, People's Dental Academy, Bhopal, Madhya Pradesh, India

Correspondence to
Dr Ajay Pillai,
drajaypillai@yahoo.co.in

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SUMMARY

Ameloblastoma occurs in a wide variety of forms. Various forms of ameloblastomas have various treatment modalities ranging from a conservative approach to surgical resection with reconstruction. We report a case of unicystic ameloblastoma with mural proliferation in a 17-year-old girl, who presented with a swelling in the lower left jaw associated with dull aching pain and was managed initially by a conservative approach followed by surgical enucleation on recurrence.

BACKGROUND

Ameloblastomas are perplexing odontogenic tumours. Their name suggests a resemblance to cells of enamel forming organs. True ameloblastomas are classified into three subtypes: solid and multicystic ameloblastoma, unicystic ameloblastoma and peripheral ameloblastoma.¹

Unicystic ameloblastoma, variously termed as mural, monocystic, intracystic, cystogenic or cystic ameloblastoma, occurs in a relatively younger age group.^{2,3}

The diverse literature shows that unicystic ameloblastoma is thought to have recurrence potential. The purpose of publishing this case report is to show that conservative treatment (eg, enucleation)

Figure 2 Intraoral examination.

is not enough for the treatment of unicystic ameloblastoma with mural proliferation and that resection of the entire tumour along with uninvolved bony margins is required.

CASE PRESENTATION

A 17-year-old female patient presented with a 4-month history of swelling in the lower left back region of the jaw. The swelling was insidious in onset and gradually progressed to the present size. The patient had dull aching pain.

Clinical examination revealed no gross asymmetry of the face and no palpable nodes were found (figure 1).

Figure 1 Extraoral examination.

Figure 3 Orthopantomogram showing a well-defined multilocular radiolucent region.

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Figure 4 Histological examination.

Intraoral examination revealed a tender diffuse bony swelling approximately 3×3 cm in size and extending mesiodistally from the mesial aspect of 36 to the distal aspect of 38, superiorly up to the gingival crevice and inferiorly obliterating the buccal vestibule ([figure 2](#)). On palpation, the swelling was bony hard in consistency with buccal cortical expansion. The overlying mucosa appears normal and smooth. Mobility was present in relation to 36 and 37.

A provisional clinical diagnosis of odontogenic cyst was arrived at based on the clinical presentation.

INVESTIGATIONS

Panoramic radiograph: It revealed a well-defined multilocular radiolucent lesion extending from the medial aspect of 36 to the distal aspect of 37. Calcification was not evident on the radiograph ([figure 3](#)).

Aspiration: Straw-coloured fluid.

Histopathological examination: H&E stained slides show a lumen lined by an epithelium and surrounded by a connective tissue capsule. The lining epithelium shows tall columnar ameloblast-like cells with hyperchromatic, reverse polarised nuclei. Numerous large and small isolated islands were noticed in the connective tissue capsule, composed of peripheral layer of columnar ameloblast like cells with some islands shows central stellate reticulum like cells undergoing squamous metaplasia. Few well hyalinised areas around these island were also noticed. The connective tissue appeared to be dense collagenous with fibroblast, bony trabeculae, nerve fibres, muscle fibres and blood vessels with extravasated RBC's ([figure 4](#)).

A histological diagnosis of unicystic ameloblastoma with mural proliferation was arrived at.

Figure 5 Orthopantomogram showing postoperative view after enucleation of the lesion with extraction of 36 and 37.

Figure 6 Orthopantomogram showing recurrence after 6 months.

DIFFERENTIAL DIAGNOSIS

- ▶ Calcifying epithelial odontogenic tumour.
- ▶ Adenomatoid odontogenic tumour.
- ▶ Unicystic ameloblastoma.
- ▶ Ameloblastic fibro-odontoma.
- ▶ Odontoma.
- ▶ Dentigerous cyst or other odontogenic cysts.
- ▶ Multicystic ameloblastoma.
- ▶ Keratocystic odontogenic tumour.

TREATMENT

Enucleation of the lesion along with extraction of 36 and 37 was initially performed with primary closure ([figure 5](#)). Following recurrence after 3 months ([figure 6](#)), marginal resection was performed with extraction of 35 and 38 ([figure 7](#)). The patient then underwent rehabilitation with dental implants after a period of 1.5 years from marginal resection ([figures 8 and 9](#)).

OUTCOME AND FOLLOW-UP

After marginal resection of the recurring lesion, a close follow-up was performed. The lesion healed without any complication within a period of 1.5 years.

DISCUSSION

According to WHO 1992 definition, ameloblastoma 'Is a true neoplasm of enamel organ like tissue which does not undergo differentiation to the point of enamel formation'.

Ameloblastomas in young people (ie, those 19 years old and younger) are thought to be rare. They account for approximately 10–15% of all reported cases of ameloblastoma.^{4 5}

Unicystic ameloblastoma is the second important clinical type of ameloblastoma. They are commonly noticed in the second and third decades of life which is considerably younger than the average of discovery for all types of ameloblastoma.⁶

Conservative treatment is suggested especially in the younger population, in the light of the divesting impacts on the

Figure 7 Orthopantomogram showing marginal resection of the ameloblastoma with extraction of 35 and 38.

Figure 8 Prosthetic rehabilitation of 35 and 36 with dental implants.

developing jaw, masticatory function, facial growth and psychosocial aspects.^{7–12} The literature suggests that initially a conservative treatment can be applied for unicystic ameloblastomas with more aggressive approaches being reserved for later recurrence. Based on the evidence from the literature, enucleation was planned for the patient as a conservative modality followed by a close observation for recurrence.

Regarding unicystic ameloblastoma, recurrence following conservative treatment is reported to be approximately 7–25%.^{13–15}

Ackermann *et al*¹⁶ classified unicystic ameloblastomas into three histological subgroups: luminal, intraluminal and mural. Among the three subtypes, mural type has the highest recurrence rate since the epithelium penetrates and breaches the fibrous wall with a high potential to invade the adjacent cancellous bone.¹¹ According to Li *et al*,^{17–18} a recurrence rate of 35.7% is reported for mural unicystic ameloblastoma.

In our case, we observed early recurrence after a period of 6 months, following which marginal resection was performed. Follow-up for 1.5 years shows healing with regeneration of the bone and no signs of recurrence.

The patient then underwent prosthodontic rehabilitation with two dental implants which are well osseointegrated after a period of 4 months from their time of placement followed by a prosthetic placement of a three unit splinted ceramic fused to metal bridge.

Figure 9 Prosthetic rehabilitation of 35 and 36 with dental implants.

An early recurrence for mural type of unicystic ameloblastoma has been reported, hence we suggest marginal resection with follow up rather than starting with conservative treatment and making the patient undergo various surgeries.

Learning points

- ▶ Unicystic ameloblastomas with mural proliferation should be treated more aggressively as compared to its other variants keeping in mind their recurrence rate.
- ▶ A close follow-up for this variant should be performed as the recurrence rate is high.
- ▶ Postsurgical radiographic examinations should be performed at frequent intervals for early detection of recurrence.

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Competing interests None.

Patient consent Obtained.

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