Unicystic Ameloblastoma: Resection/Enucleation? 
A Case Report

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Unicystic ameloblastoma (UA) is a special or a less common variant of ameloblastoma. Often mistaken as dentigerous cyst or odontogenic keratocyst, it requires histological evaluation for its confirmation. We report a case of a 13-year-old female patient who presented with persistence of swelling and altered sensation in the right lower jaw even after undergoing surgery for the same at a private hospital. Clinical evaluation showed expansion in both buccal and lingual cortical plates in the region of complaint. Radiographic and histologic findings corresponded to that of UA. Considering the age of the patient and the morbidity associated with resection, a more conservative approach was preferred. Surgery was performed under general anesthesia that included extraction of offending teeth, cystic enucleation, peripheral ostectomy, and chemical cauterization with Carnoy’s solution. Platelet-rich plasma was placed in the defect and closure was achieved. The cystic lining along with extracted teeth was sent for histopathological evaluation which confirmed it to be a UA of the mural variety. The patient has been on regular follow-ups with no signs of recurrence and satisfactory healing.

Keywords: Conservative, Enucleation, Mural, Platelet-rich plasma, Unicystic ameloblastoma

INTRODUCTION

In 1977, unicystic ameloblastoma (UA) was described as a special or a less common variant of ameloblastoma by Robinson and Martinez.¹⁻⁴ In 1992, WHO defined ameloblastoma as “a benign locally invasive polymorphic neoplasm consisting of proliferating odontogenic epithelium which usually has a follicular or a plexiform pattern lying in a fibrous stroma.”

Often mistaken as a dentigerous cyst or odontogenic keratocyst, this neoplasm needs histological evaluation for its confirmatory diagnosis.⁵ The most common site of its origin is mandibular posterior region. UA was classified by Ackermann et al. into three histological subtypes: (1) Type 1: Luminal: Unilocular cyst lined by epithelium showing ameloblastic transformation in some areas without infiltrating the epithelium, (2) Type 2: Intraluminal: Unilocular cyst lined by epithelium showing nodular plexiform ameloblastic transformation without infiltrating connective tissue, and (3) Type 3: Mural pattern: Tumor extending into connective tissue.³⁻⁸

The pathogenesis of UA is unclear; it might be attributed to defective desmosomes or intrinsic production of proteinases.⁹ In recent literature, Kahn has pointed at the possibility of human papillomavirus as a contributing factor.¹⁰

CASE REPORT

A 13-year-old female presented to the Department of Oral and Maxillofacial Surgery, Yenepoya Dental College, Mangalore, Karnataka, with the chief complaint of the persistence of swelling and numbness in the right posterior jaw region after 6 months of undergoing cystic enucleation on the same site in a private hospital. There was no contributory medical history.

The patient had been complaining of swelling and paresthesia in the right anterior jaw for a year for which she underwent surgical enucleation 6 months back in a private hospital. The excisional biopsy from the previous surgery reported it as UA. Persistence of symptoms made her visit our department. Incisional biopsy was done which was reported as UA. Intraoral periapical radiograph (IOPAR), orthopantomogram (OPG), and cone-beam computed tomography (CBCT) were advised.

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On examination, a diffuse swelling was seen on the right side of the face extending across the lower border of mandible (from the chin to the angle of mandible) as seen in Figure 1. It was non-tender with underlying bony hard swelling. Slight altered sensation was noted on the chin region.

Intra-oral examination revealed a diffuse swelling extending from distal of right canine (43) to distal of right second molar (47) with the expansion of buccal and lingual cortical plates. Teeth extending from the right first premolar to the right second molar were filled with dental cement (intentional root canal treatment) (Figure 2).

On palpation, the swelling was bony hard, and tenderness was present at the right posterior molar regions.

**Radiographic Findings**

**IOPAR**

Multiple well-defined radiolucencies were seen in the right molar region, and an OPG was advised to get a better picture.

**OPG**

Multiple well-defined radiolucent lesions were seen extending from distal of the right first premolar to the right second molar regions (Figure 3). The margins of the lesions were scalloped. The inferior alveolar nerve was pushed inferiorly. It also confirmed that root canal treatment was done for teeth extending from the right first premolar to right second molar. There was also an impacted right lower third molar.

**CBCT**

Confirmed the OPG findings and also revealed perforations of the buccal cortical plate (Figure 4).

On the basis of location, nature of lesion, age of the patient, and radiographic and clinical findings, differential diagnosis of odontogenic keratocyst, and ameloblastoma were made.

**Biopsy Report**

The section stained with H and E showed cystic lining consisting of tall columnar epithelial cells and superficial stellate reticulum-like cells. The lining showed intraluminal and mural proliferations. The connective tissue capsule presented with ameloblastomatous follicles with peripheral tall columnar cells and central stellate reticulum-like cells.

The final diagnosis was given as UA.

**Surgical Procedure**

The patient was posted for surgery under general anesthesia after obtaining consent for the same. Patient draped under aseptic conditions. 2% lignocaine with adrenaline (1:100,000) was infiltrated in the right mandibular region. A crevicular incision was placed extending from distal of lower right canine to lower right second molar regions, this was further extended till the anterior border of the ramus. Releasing incision was placed distal to lower right canine. Envelope flap was reflected. Extraction of teeth 45, 46, 47, and impacted 48 was done. The cystic linings were identified and enucleated (Figure 5a). Apicectomy was done for 44 with retrograde filling. Chemical cautery with Carnoy’s solution was done for 4 min. This was followed by copious irrigation of cystic cavity (Figure 5b) with saline.

Platelet-rich plasma was prepared with autologous blood and placed in the cavity (Figure 5c).

Simple interrupted sutures using 3-0 vicryl were placed to close the flap (Figure 5d).

The patient has been on regular follow-ups since then.
DISCUSSION

UA occurs at a very young age (second decade). Macroscopically, it is cystic in nature, and radiographically, it has a typical unilocular appearance and it has a positive response toward conservative management, and hence considered a special variant. Its predominance is 10-15% of all intraosseous ameloblastomas. Associated clinical features include swelling, facial asymmetry, unicocular radiolucency with well-defined sclerotic borders, displacement or mobility of tooth, root divergence, and tooth extrusion. Most common region of occurrence is mandibular posteriors, its ratio to maxilla in terms of occurrence being 13:1.

This case of UA had features very close to odontogenic keratocyst such as radiolucency with scalloped margins and clinical features not correlating with the extent of the lesion. Yet there were features of UA such as cortical bone expansion, which is seldom in keratocysts, which expand in an anteroposterior direction.

This being a case of recurrence or incomplete removal of the lesion, an aggressive treatment protocol is advised like segmental or marginal resection. Although resection has the lowest recurrence rate (3.6%), life more conservative approaches are preferred for a better quality of life. Marsupialization is another conservative method to reduce the size of the lesion, especially useful in large lesions. Its recurrence rate when combined with other modalities is about 18%.

Enucleation performed alone had the maximum recurrence rate (30.5%). This is probably due to two possibilities: The lining of the cyst not being removed completely and the ability of ameloblastic tumor cells to invade the cancellous bone.

Enucleation with chemical cauterization further lowers the recurrence rate to 16%. If the tooth associated with the tumor is extracted, the recurrence can be further minimized.

It is suggested by various authors that for UA, conservative therapy can be employed initially, and in case of recurrence, a more aggressive modality can be used. The treatment modality also depends on the histological variant luminal, intraluminal, or mural. The luminal and intraluminal variant cannot be treated with enucleation, but with mural patterns, resection is recommended for the fear of leaving the lining behind. Insipre of resection being the primary choice, considering the age of the patient resection was not advocated. Hence, a less aggressive surgical intervention was carried out, which included extraction of associated teeth (45, 46, 47, 48), enucleation followed by peripheral ostectomy, and chemical cauterization with Carnoy’s solution. Application of Carnoy’s solution was done for 4 min, as prolonged application can cause nerve damage. We further placed platelet rich plasma into the defect to promote better healing. The patient is on regular follow-up.
CONCLUSION

We conclude that UA poses a major dilemma to the surgeon regarding its management. Resection, which is the treatment of choice to prevent recurrence, is also associated with esthetic and functional morbidity. Considering the histopathological variant and the age of the patient, a less aggressive modality was considered. This gave the patient better functional and esthetic outcomes and also prevented any post-operative complications. We would also keep the option of resection for further treatment in case of recurrence.

REFERENCES


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