

## Case Report

### Conservative management and obturator placement for the treatment of unicystic ameloblastoma in a young 4 year old child: A case report

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#### ABSTRACT:

Unicystic ameloblastoma is a rare, benign, locally invasive odontogenic neoplasm of young age that show clinical, radiographic, or gross features of an odontogenic cyst, but histologically shows typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth. This case report describes the case of a 4-year-old boy with unicystic ameloblastoma of the mandible, which involved unerupted left first premolar. Marsupialization with obturator placement was performed to shrink the lesion. Later enucleation was performed to remove all cystic lesion. Five months post-operative, the lesion had started healing. The combination of conservative surgery and obturator placement effectively lead to shrinkage of the lesion and preserved mandibular growth.

**Keywords:** Unicystic ameloblastoma, pediatrics, marsupialization, enucleation, obturator.

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#### INTRODUCTION

Robinson and Martinez<sup>1</sup> first identified unicystic ameloblastoma (UA) as a distinct subtype of ameloblastoma in 1977. Ameloblastoma is defined by WHO in 1992 as a benign locally invasive polymorphic neoplasm made up of proliferating odontogenic epithelium that typically has a follicular or plexiform pattern and is embedded in a fibrous stroma. Histological confirmation is required because it typically develops in the mandibular molar-ramus region, resembles a nonneoplastic odontogenic cyst greatly, and is frequently misdiagnosed as a dentigerous cyst and an odontogenic keratocyst.<sup>2</sup> Three histologic subtypes of unicystic ameloblastoma were identified by Ackermann et al.<sup>3</sup>: (1) luminal (type 1), (2) intraluminal (type 2), in which the tumour is contained inside the cyst's epithelium and may be treated conservatively by enucleation. (3) mural pattern (type 3) in which tumor is present in the connective tissue wall of the cyst which should be

treated aggressively in exactly the same manner as multicystic ameloblastoma.

Cystic degeneration and unicystic ameloblastoma have unclear pathophysiology. It has been proposed that the cystic degeneration of neoplasm may be caused by epithelial dysadhesion (for example, defective desmosomes) or intrinsic proteinase production (for example, metalloproteinases, serine proteinases), enzymes that normally degrade the central zone of the enamel organ after tooth development.<sup>4</sup> Kahn<sup>5</sup> recently raised the hypothesis that the human papilloma virus may be involved in the growth of unicystic ameloblastoma. This case report highlights a case of unicystic ameloblastoma in a child presenting with a swelling related to the lower left quadrant of the jaw and its surgical treatment, as well as the placement and evaluation of the role of an acrylic obturator.

**CASE REPORT**

A 4 years-old boy was referred to the Department of Pedodontics and Preventive dentistry, Punjab Government Dental college and Hospital, Amritsar complaining of pain and swelling related to teeth 73,74 and 75. (Fig.1) Clinically intraoral examination revealed a compressible swelling of the buccal cortical plates of lower left quadrant. (Fig.2) The radiographic findings, CT scan and panoramic radiograph revealed a well-defined radiolucent lesion, with sclerotic margins, completely associated with the crown of the unerupted mandibular left first permanent premolar. The root of the 71,72,73,74 and 75 were involved in the lesion, with the presence of

root resorption.(Fig.3,4) There was no sensorineural or motor deficit at the facial structures. Following the clinical and radiographic examination, a provisional diagnosis of the unicysticameloblastomawas made. Considering the age of the patient and vicinity to the lower border of mandible, marsupialization of the cystic cavity was planned. Surgical intervention was carried out under general anesthesia in the Oral and Maxillofacial Department, PGDC&H, Amritsar. The exposure of the cyst cavity was done by opening a flap. (Fig.5) After the flap opening process, the cyst cavity was identified and the contents of the cyst were aspirated. (Fig.6)On aspiration of cyst cavity, golden yellow coloredfluid was noticed.



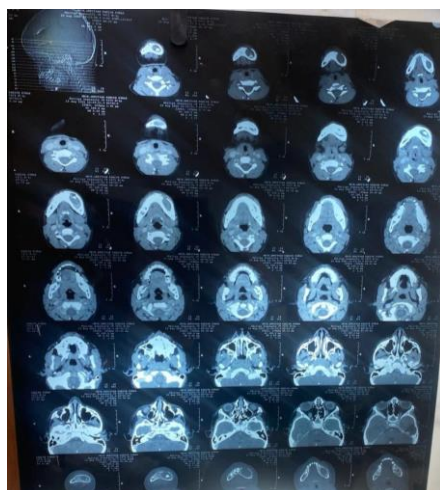
**Fig.1 Preoperative picture**



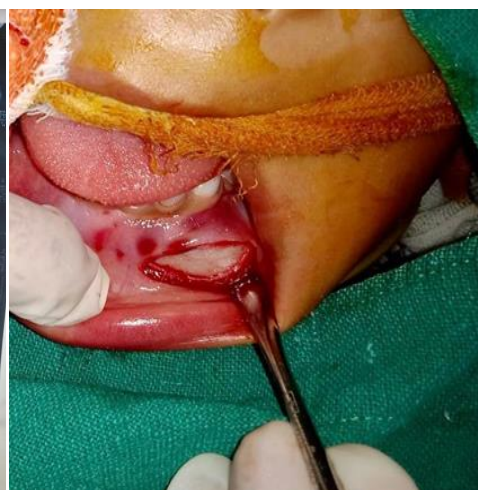
**Fig.2 Preoperative intraorally**



**Fig.3 OPG**



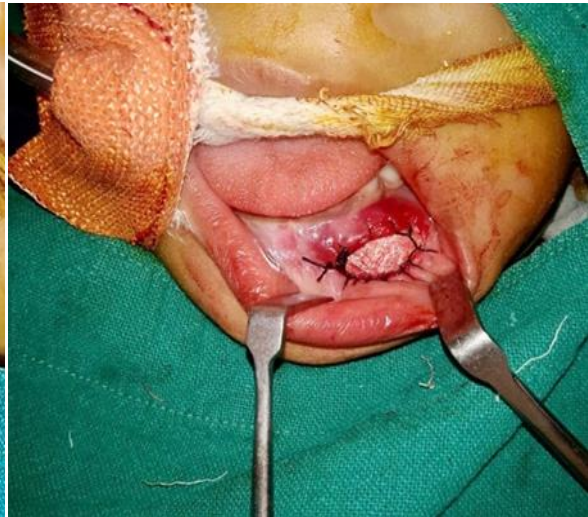
**Fig.4 CT scan**



**Fig.5 Creation of a surgical window**



**Fig.6**Aspiration of fluid out of cystic



**Fig.7** Cystic lining sutured to the mucosacavity for histological examination.



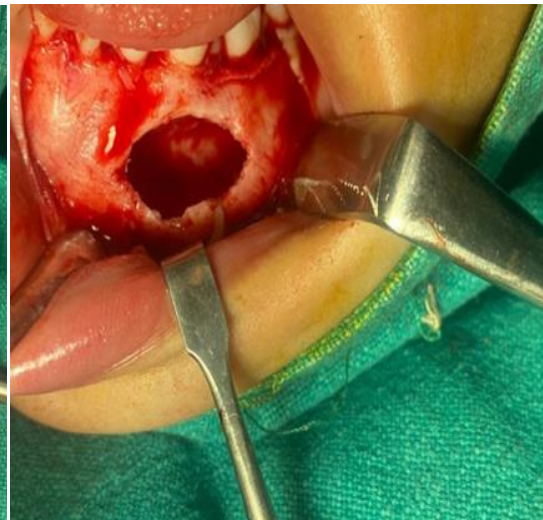
**Fig.8** Window created after to cover the defect



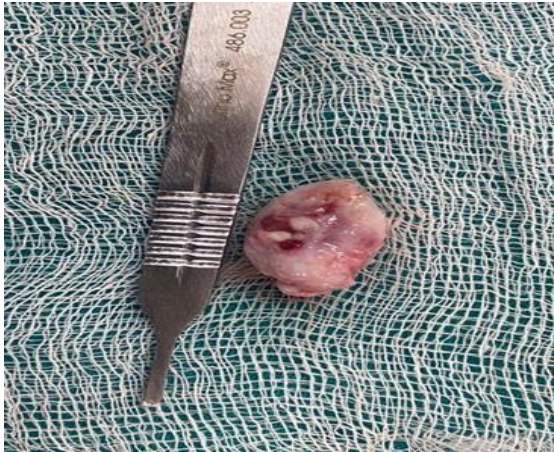
**Fig.9** Placement of the obturator to marsupialisation



**Fig.10** Creation of second surgical window



**Fig.11** Enucleation of entire lesion



**Fig.12 Excised cystic mass**



**Fig.13 Placement of iodoform gauze**



**Fig.14 flap sutured post surgery**



**Fig.15 Post operative intraoral photographs after 6 months**



**Fig.16 Post operative extraoral photographs after 6 months**

On the basis of age, location, clinical, and radiographical features, preoperative diagnosis of aninflammatory dentigerous cyst and unicysticameloblastoma was made. Contents were sent for histopathologic evaluation where the diagnosis of unicysticameloblastoma was confirmed.

The surgical procedure of marsupialization was completed with no complications and further analgesics and antibiotics were prescribed. The cystic cavity was filled with iodoform gauze that was changed after seven days following washing and disinfecting the wound. (Fig.7) Thin alginate

impression was prepared, so that its placement was soft. On the seventh day a removable acrylic obturator, that has been previously adjusted to the cystic window (Fig.8), was delivered to the patient. (Fig.9)The patient and his parents were informed about how to clean the lumen of the cyst using Betadine mouthwash twice per day.

In the second stage of surgery, enucleation was planned as the cystic lesion had shrunk. The lesion was enucleated along with unerupted first premolar. (Fig.10,11,12) Copious irrigation of bony cavity was done with betadine solution and normal saline. Carnoy's solution was applied in the bone cavity for 3 min with cotton applicators. The bony cavity was rinsed with normal saline and packed with iodoform gauze. (Fig.13) The flap was sutured with mersilk. (Fig.14) The patient was recalled for follow-up every two weeks till 6 months till complete healing of the lesion. All the follow-up visits were uneventful with no complications.

## DISCUSSION

Unicystic ameloblastoma is a tumor of young age group, typically unilocular radiographic appearance, macroscopically cystic nature and most important, it relatively better response to conservative treatment makes it a distinguishable entity. It accounts for 10% to 15% of all intraosseous ameloblastoma.<sup>3</sup> Although most commonly found in association with the crowns of impacted teeth, it may be found in interradicular, periapical, or edentulous region.<sup>2</sup> Commonly associated manifestations include painless swelling, facial asymmetry, unilocular lesion with defined sclerotic borders, tooth impaction, displacement, mobility, root resorption, root divergence, occlusal interference, and extrusion of tooth.<sup>6</sup> This distinct prognostic entity is predominantly observed in the mandibular molar-ramus region. The posterior region of maxilla is considered to be rare and atypical. The ratio of mandibular to maxillary unicystic ameloblastoma has been reported to be 13:1.<sup>7</sup> The present case report describes the conservative management unicystic ameloblastoma of mandibular body region.

The treatment of unicystic ameloblastoma has been controversial and can be conventional, radical, or conservative. Conventional approaches consist of segmental or marginal resectioning with subsequent reconstructive procedures often being required. Conversely marsupialization is considered a good way to exteriorize or decompress a cyst. Marsupialization followed by enucleation is one therapeutic approach for ameloblastoma.<sup>8</sup> No substantial evidence proves one treatment modality as the most effective, and many reasons exist for this uncertainty.

There is a consensus among various authors that ameloblastoma has to be treated aggressively to avoid recurrences. However, an aggressive surgical approach in pediatric patients could result in numerous complications, such as functional and masticatory

changes, mutilations, and loss of permanent teeth involved in the tumor and facial deformities. Therefore, there is a dilemma regarding the applicability of an initial radical extensive surgery in children.

In children, the treatment of unicystic ameloblastoma is complicated by the following key factors: 1) continued facial growth, different bone physiology (greater percentage of cancellous bone, higher bone turnover, and reactive periosteum), and the presence of unerupted teeth; 2) difficulty in making the initial diagnosis; and 3) predominance of the unicystic type of ameloblastoma. Given that mandible resection in pediatric patients may lead to complications, such as dysfunction and deformity, our patient was submitted to conservative treatment to preserve the permanent teeth and as much bone as possible, to avoid esthetic deformities.<sup>9</sup>

In this case report, the surgical procedure was completed with no complications and a week later a removable acrylic obturator was prepared and delivered to the patient. The obturator prevents contamination of the lesion or food accumulation in the cystic pouch, its smooth surface prevents the removal of the formed blood clot, moreover, it hinders the formation of fibrous scar.<sup>10</sup>

In the second stage of treatment enucleation was performed. Enucleation alone yielded the highest recurrence rate among all treatment (30.5%). Two possible explanations: firstly, cystic lining of the tumor is inadequately removed; secondly, ameloblastic tumor cells can invade the cancellous bone to a certain extent.<sup>11</sup> Enucleation followed by application of Carnoy's solution has resulted in a recurrence rate of 16.0% which is the best except for resection. The recurrence rate could even be lower than reported, if the closely related teeth with tumor are extracted. Because in an attempt to preserve the tooth without damage, tumor remnants may be left around the tooth apex or root and these may lead to recurrence.<sup>11,12</sup> In the present case reports, teeth in close relation of tumor were extracted. Carnoy's solution, a powerful fixative penetrates the cancellous spaces and thus fixes the remaining tumor cells. Usually, Carnoy's solution is applied for 3-5 min. However, Frerich *et al*,<sup>13</sup> suggested that the application of Carnoy's solution should not exceed 3 min and should not be directly applied over the nerve as it could lead to nerve impairment.

After 5 months, we observed that the lesion had partially healed. There are several reports of cystic ameloblastoma cases in which the tumor completely disappeared after marsupialization alone, suggesting that marsupialization helps avoid wide resection of the mandible in patients with unicystic ameloblastoma.<sup>14</sup>

## CONCLUSION

All clinical and histological factors should be taken into account when determining the optimal course of treatment for children with unicystic ameloblastoma.

The unilocular radiolucencies of the jaws should be reported to oral healthcare professionals because this lesion may be unicystic ameloblastoma. Early intervention, conservative surgical procedures, Carnoy's solution application, and the excision of teeth that are closely connected to the lesion may help to reduce the risk of problems from greater resections and enhance treatment outcomes. The use of a precisely fitted removable acrylic obturator that was finely adjusted according to the size of the lesion at each follow-up appointment was a strong adjunct in the success of the treatment.

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