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Case Report
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Marsupialization: An Understated and Underestimated Procedure to Prevent Major Morbidity to the Mandible: A Case Report

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Abstract

Unicystic ameloblastoma is a rare variation of ameloblastoma that presents as a cystic lesion in the jaw. Diagnosis can be challenging due to its radiographic similarities with other odontogenic cysts and tumors. This case report presents a 19-year-old female patient with a chief complaint of swelling in the left mandibular molar region for 7 months. Radiographic examination revealed a well-defined multilocular radiolucency with cortical bone thinning, lobulations, and resorption of tooth surfaces.

Incisional biopsy confirmed the lesion to be a plexiform unicystic ameloblastoma. Due to the patient's young age, a conservative approach was followed with a two- stage treatment plan. Stage 1 involved marsupialization of the cystic cavity using a customized acrylic plug to maintain patency and promote bone formation. After three months, the lesion size reduced and enucleation was performed in Stage 2. Radiographs at 6 months showed reduced radiolucency, and complete bone formation was observed at 12 months. No recurrence was observed during the 18-month follow-up. This case report highlights the importance of accurate diagnosis and appropriate management of plexiform unicystic ameloblastoma to achieve successful treatment outcomes.

Introduction

Unicystic ameloblastoma is a varaiation of ameloblastoma that presents as a cystic lesion in the jaw. It is often difficult to diagnose because it can mimic other odontogenic cysts and tumors on radiographic images, making it challenging to arrive at a definitive diagnosis without a biopsy or histopathological examination.

Some common radiographic features of unicystic ameloblastoma include a unilocular appearance, well-defined borders, and an association with impacted teeth. However, these features are not always present, and some cases may exhibit multilocular or ill-defined borders. Therefore, it is important to consider other clinical and histological features when making a diagnosis.

Overall, the diagnosis of unicystic ameloblastoma requires a combination of radiographic, clinical, and histopathological findings. A thorough evaluation by an experienced oral and maxillofacial pathologist is essential to arrive at an accurate diagnosis and develop an appropriate treatment plan.

Plexiform unicystic ameloblastoma is a rare variant of unicystic ameloblastoma, characterized by the proliferation of epithelium in a cystic cavity that may be mistaken for an odontogenic cyst due to its unilocular presentation. However, it should be recognized as an aggressive tumor, despite its slow growth and persistent nature. This type of ameloblastoma primarily affects the posterior area of the lower jaw in 80% of cases, making early diagnosis and management essential for successful treatment outcomes. Here we present a case to provide further insight into the diagnosis and management of plexiform unicystic ameloblastoma.

Case Report

In august 2021, an 19-year-old female patient with a chief report of swelling in the left mandibular molar region since 7 months was referred to our medical center. The patient had visited three centeres previolsy where an aggressive resection was advised. The patient had no systemic health conditions. Extraoral examination revealed swelling in the left mandibular body area 2cm from commissure of lip till 4 cm ahead of the posterior border of the mandible, extending till the lower border of the mandible

and intraoral examination revealed mucosal swelling that extended from the lower first pre-molar to the third molar region. And mobility of mandibular posterior 45 46 on the left side.

Radiographic examination of the lesion (orthopantomogram) showed a well-defined, multilocular radiolucency, involving the left side of the mandible which extended anteroposteriorly till the second molar region. Thinning of cortical bone present in the lower border of the mandible, lobulations seen on the inferior cortex of mandible, loss of cortication seen on the anterior border of the body of the mandible. Resorption of the surface of the root of the mandibular first, second pre-molar and first and second molar was also noted.

Considering the clinical and radiographic findings, differential diagnosis of unicystic ameloblastoma, dentigerous cyst, and odontogenic keratocyst was considered., an incisional biopsy was performed under local aneathsisa. Histopathology sections revealed loosely arranged fibro-cellular connective tissue stroma lined by areas of discontinuous 2-4 cell layer flattened epithelial cell with evidence of intramural and intraluminal plexiform strands. Pathologic report confirmed the lesion to be unicystic ameloblastoma of plexiform variety.

As the young age of the patient a more conserative approach was proposed. Patsch 2 treatement was followed the treatment was done in two stages. Stage 1 where the cavity was marsupilized and stage 2 where after adequate decompression of the lesion it would be enucleated. Extraction of 35 and 36 was done. The existing socket was used to marsupilize the cystic cavity and the patency of the cavity was maintained by a customized acrylic plug. Mainting the continuity between the marsupialized lesion and oral environment it would ultimately reduce the lesion size. The acrylic plug was trimmed and made suitable in monthly time periods.

The patient was recalled at timely intervals. The parents were instructed to maintain overall proper hygiene of the oral cavity through self-irrigation with iodine solution (betatine) after marsupialization. Three months after marsupialization, the lesion diminished in size, new bone formation was observed in the periphery. At this stage due to enough bone formation, under local aneathsia enucleation was performed.

Radiographs obtained 6 months after enucleation showed that the lesion margin had lost clarity and that the regenerated bone was replaced by normal trabeculae. The radiolucent area was significantly reduced. At 12 months after marsupialization, complete bone formation was observed. To date, no evidence of tumor recurrence has been observed on examinations during the 18-month follow-up.

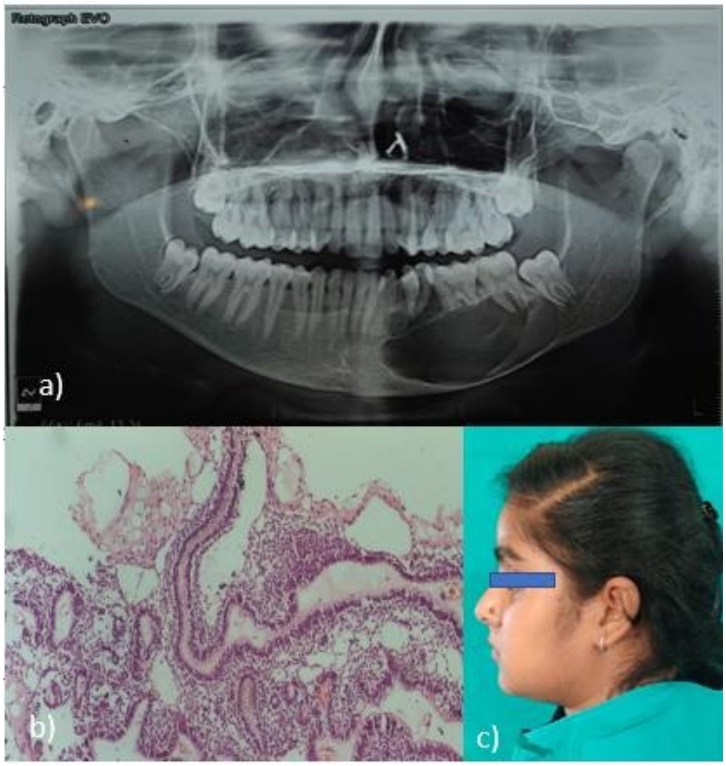

Figure 1: a) Orthopantomograph shows unilocular radiolucency with sclerotic borders from 33 to mesial root of 38 region.
b) Photomicrograph at 10 x showing plexiform pattern of ameloblastoma.
c) Photo of patient on arrival showing swelling in the body region of the mandible.

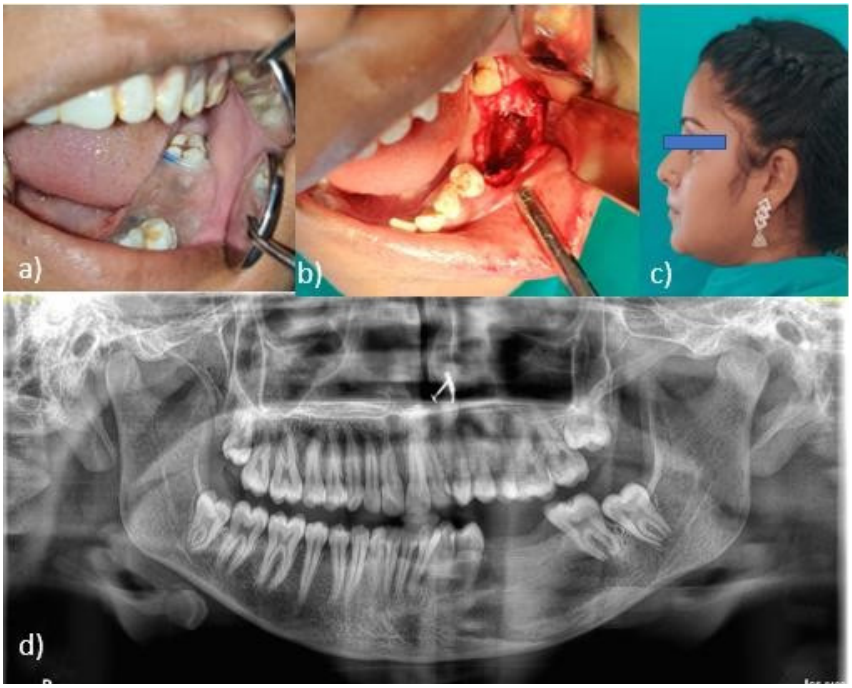

Figure 2: a) Acrylic plug was placed after marsupialization of cavity, which was maintained for 6 months.
b) Enucleation of cavity was done after 6 months.
c) Patient seen during marsupialization of the cavity, showing less amount of swelling in the mandibular body region.
d) Orthopantomograph shows mixed radiopaque radiolucent zone with boner formation around the lower border of the mandible from 33 to mesial root of 38 region.

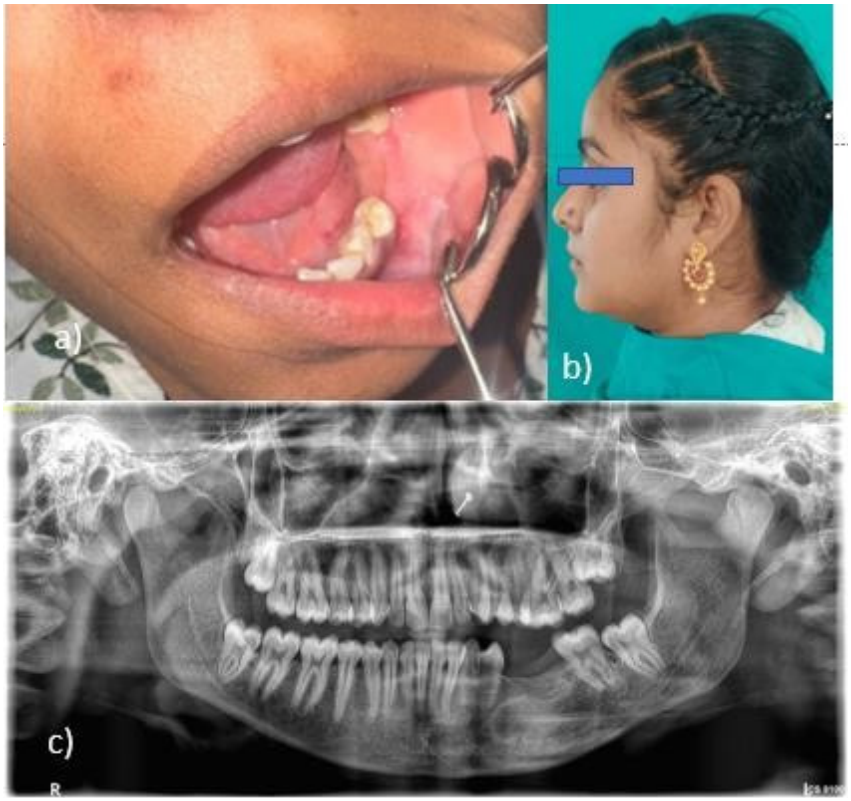

Figure 3: a) Intra oral shows complete closure of the marsupialized walls of the cystic cavity taken at 3 months after enucleation.
b) Patient as seen after 3 months post enucleation showing complete resolve of lesion.
c) Orthopantomograph radiopaque area between 33 to 38 region showing complete resolution of lesion.

Declaration of Interest Statement

Discussion

The treatment of plexiform unicystic ameloblastoma has been controversial. It can be radical or conservative. Segmental or marginal resectioning of the lesion followed by reconstruction is an aggressive approach that can be used to treat certain conditions. On the other hand, more conservative treatments involve enucleation, enucleation followed by the application of carnoy's solution, or marsupialization followed by enucleation. However, there is currently no conclusive evidence as to which treatment method is the most effective, and controversy exists regarding the best course of action. This is due in part to the rarity of the tumor, which makes it difficult to reach a definitive conclusion on the matter.

The recurrence rate is a crucial factor in determining the effectiveness of a treatment, and it can vary depending on the type of ameloblastoma. Unicystic ameloblastoma is less aggressive than the solid type, but it still has a potential for recurrence, with reported rates ranging from 10% to 25% after treatment [1].

Studies suggest that radical treatment may be more effective than conservative treatment in reducing the recurrence rate [1-6]. According to lau and samman, the highest recurrence rate was observed with enucleation alone (30.5%), while the lowest (3.6%) was seen with resection [1]. Seintou et al. Reported a recurrence rate of 29.4% after enucleation or excision, and all recurrent cases were related to the conservative approach with enucleation or excision. No recurrence was observed after resection [2]. Similarly, sampson and pogrel found that all patients treated with curettage alone developed recurrence [3].

It is essential to strike a balance between success and overtreatment when choosing the resection option, as a wide bone margin removal can reduce the chances of recurrence. However, in young patients, radical resectioning of an ameloblastoma is often avoided due to several factors. Radical surgery may lead to deformity, dysfunction, and complications, such as masticatory dysfunction, abnormal jaw movement, and tooth removal, even after successful reconstruction.

In young patients, the lack of mandibular growth can cause severe facial deformity that directly impacts their quality of life. Therefore, despite the high success rate of resection, more conservative treatments have been recently preferred. Tanaka et al. Suggested that minimal surgical treatment should be the first-choice procedure for any case of oral and maxillofacial benign tumors in young patients [4].

The biologic behavior of unicystic ameloblastoma is typically less invasive, and it responds well to conservative treatment compared to a multicystic ameloblastoma. In our case, we opted for conservative therapy and used marsupialization as the initial treatment to reduce the tumor's size. Once the tumors had regressed in size, enucleation was done.

Marsupialization is aimed at decreasing the size of the tumor, which reduces the need for more extensive surgery. By decompressing the internal contents, bone remodeling and osteogenesis are promoted. There are several benefits of decompression, including preservation of pulp vitality, preservation of the inferior alveolar nerve or maxillary sinus, maintenance of the mandibular contour and growth, prevention of jaw fracture, and a low risk of recurrence. According to lau and samman's systematic review, the recurrence rate (18%) after marsupialization with or without further treatment was lower than that of enucleation alone (30.5%).

The effectiveness of marsupialization can be influenced by several factors, such as the patient's age, the technique used, the removal of solid growths during incisional biopsy, close radiographic follow- up, and the effectiveness of enucleation after marsupialization [7]. Nakamura et al. Have found that the effectiveness of marsupialization depends on the potential for new bone formation, the technique used, and the growth characteristics of the tumor [8, 9]. They have also reported that younger patients, particularly those in the second decade of life, have a higher potential for bone formation and are more likely to benefit from marsupialization. On the other hand, older patients may require a longer time for marsupialization to be effective, and it may be more difficult to decompress lesions in these patients [10].

It is important to note that the choice of treatment for unicystic ameloblastoma should be individualized for each patient. Factors such as age, location, size, and growth pattern of the tumor, as well as the patient's overall health and preferences should be considered. Conservative treatment may be a viable option for younger patients with larger, well-circumscribed lesions, while more aggressive surgical approaches may be necessary for cases of recurrence. In any case, close follow-up and monitoring are essential to detect any signs of recurrence or progression, and to ensure the best possible outcomes for the patient.

Conclusion

In most instances, unicystic ameloblastoma is considered as a tumor and treated more aggressively. However, conservative treatment for plexiform unicystic ameloblastoma in young patients is more advantageous. Here, to save continuity of mandible as well as asthetics of a young patient to maintain the form and aesthetic mansupilization followed by enucleation was done. If there is recurrance then resection can be performed at a later stage [11-18].

Hence, we conclude that marusiplzation is a better choice Our patient who was treated by conservative anagement did not have any complications, and the condition was well maintained, with no signs of recurrence after a follow up 1 ½ year.

Declaration of Interest

"In consideration of the of taking action in Marsupilization ; an understated and undestimated procedure , to prevent a major mobirdity to mandible : A case report' reviewing and editing my (our) submission, the author(s) undersigned hereby transfer(s), assign(s), or otherwise convey(s) all copyright ownership to the Journal. In the event that such work is published in the Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology The undersigned author(s) understands that if the manuscript is accepted, the Editors reserve the right to determine whether it will be published in the print edition or solely in the Internet edition of the Journal. Articles accepted for publication are subject to editorial revision.

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