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Case Report

Dentistry Section

Recurrence of Ameloblastoma on the Right Side of the Mandible: A Case Report

KR SHAKILA[1], LP RAGHUPATHY[2], RM ARAVINDH SWAMY[3], J SOWMIYA[4]

ABSTRACT

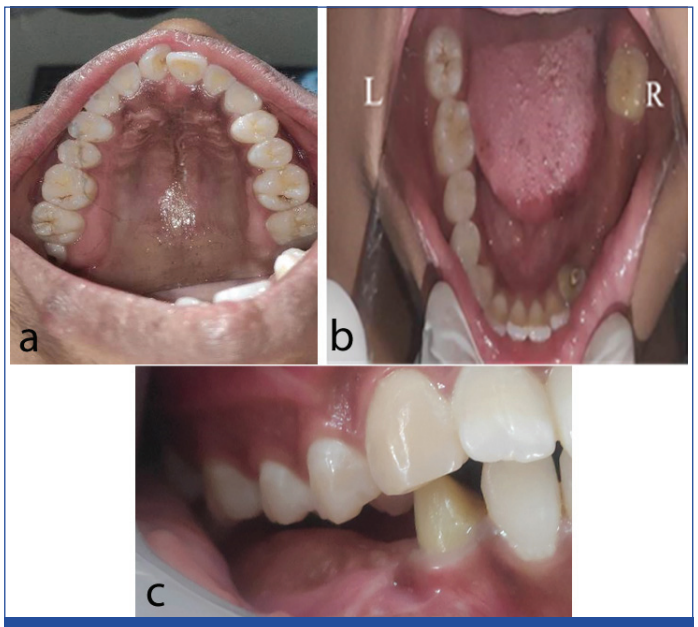
Ameloblastoma is a benign, locally invasive epithelial odontogenic tumour occuring in the jaw. Among all types, multicystic ameloblastoma is a locally aggressive lesion and prone to recurrence. Hereby, the authors present a case report of a 23-year-old female with a chief complaint of painful swelling on the right lower third of the face for the past one month. History revealed a gradual increase in size of the swelling, along with intermittent and non radiating pain that worsened during mastication and improved with rest. Patient past history revealed similar swelling in the same region twice, with the first occurring five-years-ago and a recurrence after two years of treatment. Both the episodes were treated with laser ablative surgery and extraction of the affected tooth. The patient presented with recurrent swelling one month ago and underwent an incisional biopsy, which revealed a histopathological diagnosis of unicystic ameloblastoma. Additional investigations, including an occlusal radiograph, orthopantomogram, and Computed Tomography (CT), were performed. Based on the recurrence, clinical examination, and investigations, the present case was diagnosed as unicystic ameloblastoma. It was treated with segmental resection followed by reconstruction of the mandible using a free fibula graft was planned. The prognosis was good, and regular follow-up has been maintained for the last four months, with the patient still under review. The uniqueness of present case lies in the patient's second decade of life, three episodes of recurrence within five years, and the treatment approach of segmental mandibulectomy with immediate free fibula graft reconstruction. This procedure reduces the risk of recurrence, allows for full rehabilitation within a short period, and minimises the number of surgical procedures.

Keywords: Acanthomatous ameloblastoma, Mixed/solid type, Squamous metaplasia

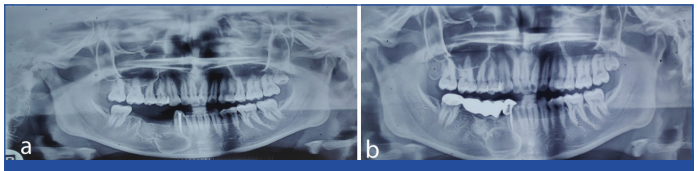
CASE REPORT

A 23-year-old female patient presented to the Department of Oral Medicine and Radiology, Karpaga Vinayaga Institute of Dental Sciences, with a chief complaint of painful swelling in the lower back jaw region of the face for the past one month. The history of the presenting illness revealed that the swelling gradually increased in size and was accompanied by intermittent, non radiating pain that worsened during mastication and was relieved by rest. The intra oral images have been shown in [Table/Fig-1a-c]. The patient had no relevant medical history. Past dental history revealed two previous episodes of similar swelling in the same region. The first episode occurred seven years ago, during which the patient visited a private clinic where radiographic investigation and panoramic imaging were performed [Table/Fig-2a]. The imaging showed a large radiolucent area extending from the apical region of tooth (according to the Federation Dentaire Internationale tooth numbering system). Based on the radiographic interpretation, it was diagnosed as unicystic ameloblastoma. The patient underwent endodontic procedures (Root Canal Treatment of 44), extraction of tooth 47, and laser ablative surgery in September 2017. Follow-up radiographic investigations were performed after the procedure [Table/Fig-2b].

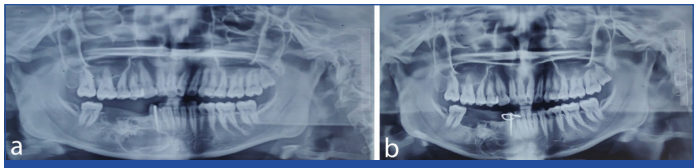
For a period of one and a half years, the patient remained normal. However, patient noticed recurrent swelling in the same region, for which patient revisited the previously visited private clinic. The patient underwent laser ablative surgery for the second time in September 2019. Subsequent radiographic investigations, including an orthopantomogram, showed mixed radiopacities and radiolucencies extending from the apical region of tooth 43 to 45 as a follow-up [Table/Fig-3a,b]. Approximately one year ago, the patient developed another painful swelling similar to previous episodes and sought treatment at a private Dental College in Chennai, India. Radiographic investigations, including an orthopantomogram, revealed multilocular radiopacities and radiolucencies extending from the apical region of tooth 43 to 47 [Table/Fig-4], with mesial root resorption on 47. An occlusal radiograph showed buccal cortical plate expansion with radiopacities [Table/Fig-5].



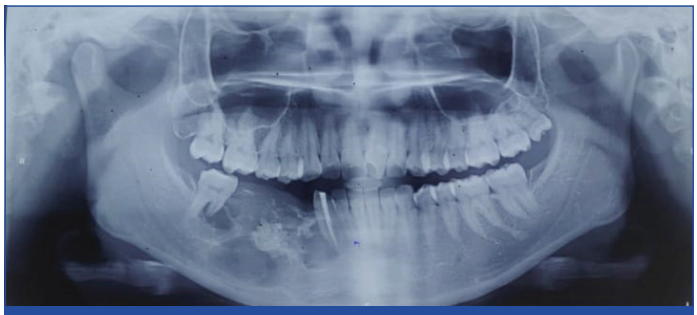
[Table/Fig-1]: Intra-oral images a) Maxillary aspect b) Mandibular aspect c) Right lateral view of mandible.



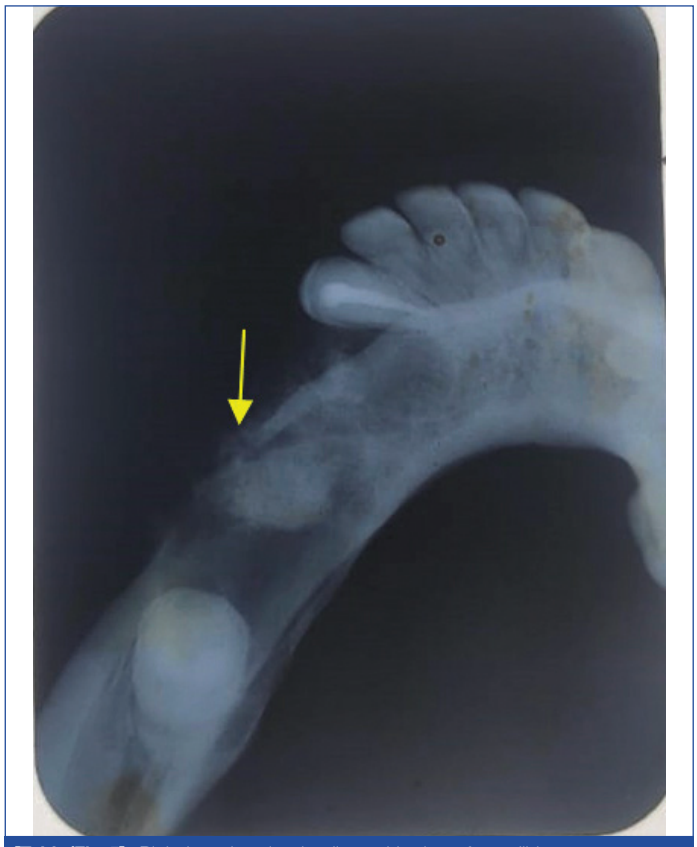
[Table/Fig-2]: a,b) Orthopantamogram reveals unilocular radiolucencies of varying sizes involving right-side of the mandible in relation to 44, 45 and 46 regions 2017-2018.



[Table/Fig-3]: Postoperative follow-up images. a,b) After laser ablative surgery performed in year 2019, mixed radiopacities and radiolucencies extending from apical region of 43 to 45. 2020-2021.



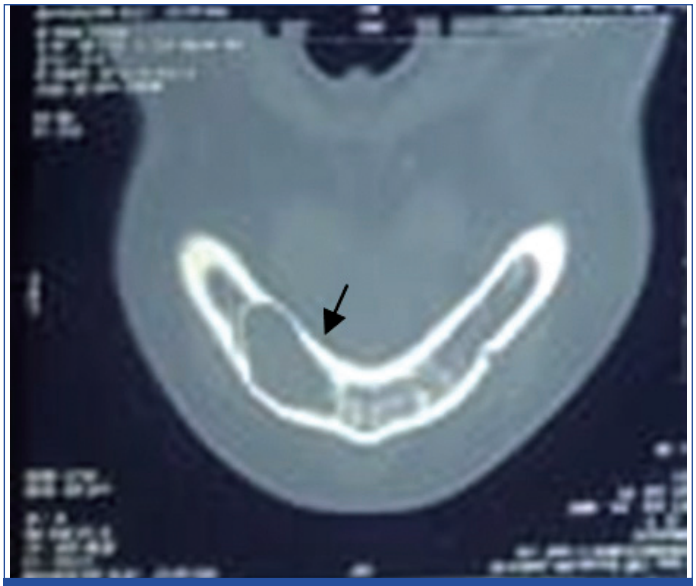
[Table/Fig-4]: Multilocular mixed radiopacities and radiolucencies extending from apical region of 43 to 47, with mesial root resorption on 47.



[Table/Fig-5]: Right lateral occlusal radiographic view of mandible.

Computed tomography revealed buccal and lingual cortical bone expansion of the right body of the mandible [Table/Fig-6]. An incisional biopsy was performed, and the results indicated a diagnosis of unicystic ameloblastoma.

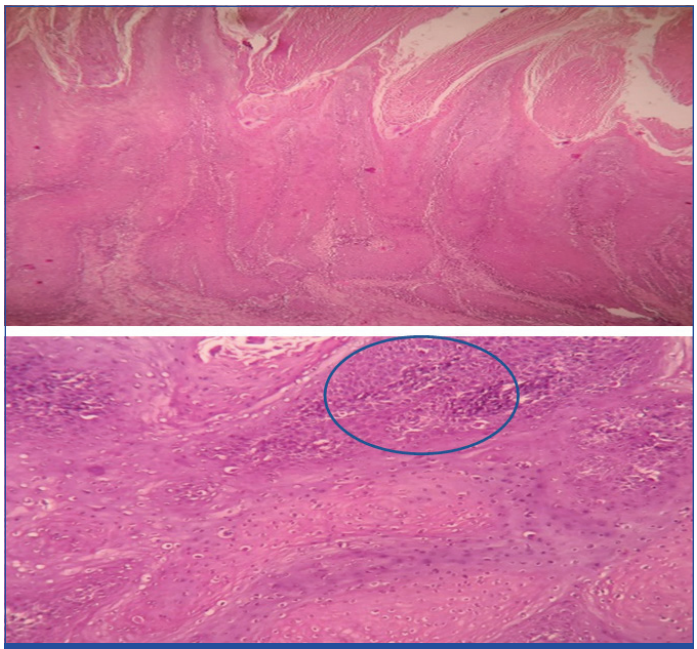
On extraoral examination and inspection, evidence of facial asymmetry was observed on the right side of the face, attributed to a single diffuse swelling measuring approximately 5×4 cm. The swelling was roughly oval in shape and extended anteriorly from 2 cm below the right commissure of the lip at the parasymphysis region, posteriorly up to the right angle of the mandible, superiorly 1 cm below the infraorbital region, and inferiorly along the right lower border of the mandible. The skin over the swelling appeared normal with no secondary changes. Palpation confirmed the size, shape, and extension of the swelling, which was found to be hard in consistency and tender, without any evident bleeding or pus discharge. Intraoral examination revealed an exophytic bony swelling over the right lower alveolar ridge, extending anteriorly from the distal aspect of 43 to the posterior aspect of tooth



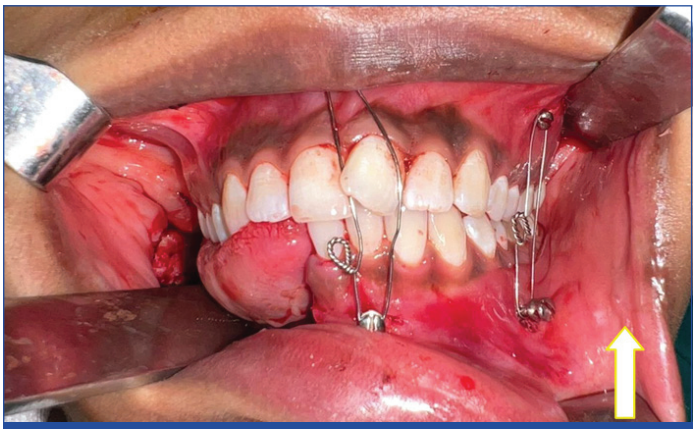
[Table/Fig-6]: CT axial section of mandible. The arrow mark shows well-defined unilocular radiolucency on the right-side of the mandible with buccolingual expansion of the cortical plate surrounded by sclerotic border.
CT: Computed tomography

48. The affected area exhibited hardness and tenderness, with missing teeth 44, 45, 46, and 47.

Based on the history of recurrence, clinical findings, radiographic findings, and histopathological report [Table/Fig-7a,b], a provisional diagnosis of recurrent unicystic ameloblastoma of the right body of the mandible was made, considering the differential diagnosis of odontogenic keratocyst, calcifying odontogenic cyst, peripheral giant cell granuloma, and odontogenic myxoma. The treatment involved intermaxillary fixation [Table/Fig-8], followed by segmental mandibulectomy and free fibula graft reconstruction. The patient was positioned supine, and general anesthesia was administered via right nasoendotracheal intubation. A 2% lignocaine with adrenaline was injected along the incision line. Incisions were made using a no. 15 Bard Parker (BP) blade. A subplatysmal flap was raised, and the right common facial vein and facial artery were identified and ligated. The right submandibular gland, along with its duct, was excised using an oscillating saw. A vertical cut was made from the 42 region [Table/Fig-8], with extraction of 42, while another cut was made beyond the third molar at the angle region, completing the segmental mandibulectomy.



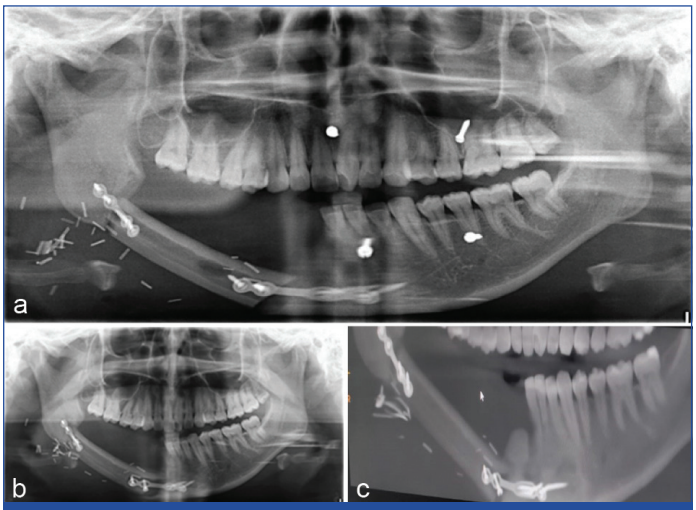
[Table/Fig-7]: a,b) Histopathological image of unicystic ameloblastoma. Focal areas showing islands and follicles of ameloblastic epithelium with palisading nuclei and central stellate reticulum (H&E).



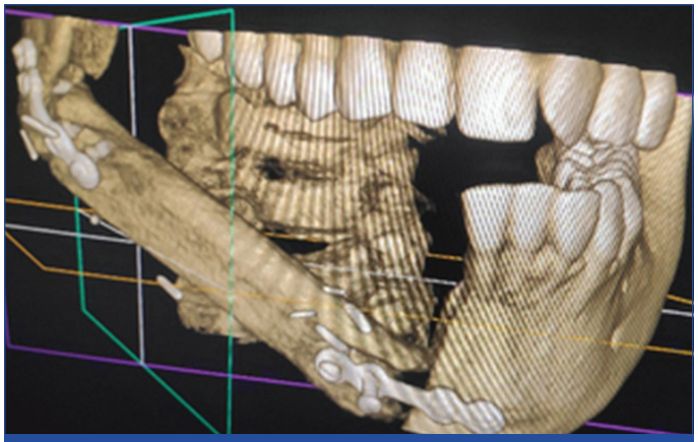
[Table/Fig-8]: Surgical image of intermaxillary fixation. The treatment done was intermaxillary fixation, followed by segmental mandibulectomy and free fibula graft reconstruction.

A tourniquet was placed on the left thigh and inflated to 300 mmHg. Painting and draping were done in the usual manner. The fibula above the left leg was marked using Doppler and skin paddles to identify and mark the myocutaneous flap perforation. Incisions were made over the markings, and layer-by-layer dissection was performed. The flexor hallucis longus muscle, along with its lateral and anterior compartments, was dissected. The posterior intermuscular septum was dissected, and the peroneal artery and vein were identified in the posterior compartment. The fibula bone was exposed along the plane of dissection, and approximately 8 cm of fibula bone was harvested along with the skin paddle. Osteotomy cuts were made with an oscillating saw at the distal and proximal ends. The donor site was closed primarily. Approximately 5.5 cm of fibula was customised to match the shape of the segmental defect in the mandible and was fixed using two titanium mini plates. Microvascular anastomosis was performed, connecting the facial artery to the peroneal artery and the facial vein to the peroneal vein. The osteomyocutaneous flap was sutured to the lower vestibule using 3-0 Vicryl. Suturing was done in two layers using 3-0 Vicryl and 2-0 Ethilon.

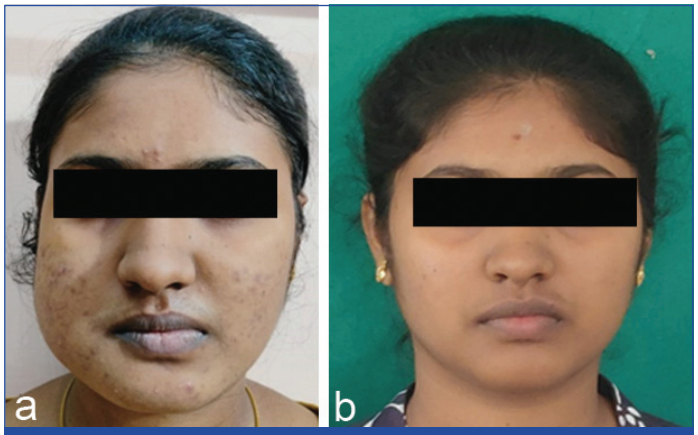
Orthopantomogram and Cone Beam Computed Tomography (CBCT) showed successful bony reconstruction of the right side of the mandible, from the symphysis to the angle of the mandible, using titanium miniplate fixation screws. There were no signs of relapse during the one-month, two-month, and four-month follow-up [Table/Fig-9a-c,10,11a,b]. The prognosis was good, and regular follow-up with the patient has been maintained for the last four months, with the patient still under review.



[Table/Fig-9]: a,b,c) Orthopantamogram shows bony reconstruction of the right-side of the mandible from symphysis to the angle of mandible with titanium miniplate fixation screws without any relapse at 1 month, 2 month and 4 month follow-up. 1st month follow-up; 2nd month follow-up; 3rd month follow-up.



[Table/Fig-10]: CBCT-3D reconstruction of mandible.
CBCT: Cone beam computed tomography



[Table/Fig-11]: a) Preoperative image b) Postoperative image.

DISCUSSION

Ameloblastoma is defined as unicentric, non functional, intermittent in growth, anatomically benign, and clinically persistent, as described by Robinson HBG [1]. It is one of the most common odontogenic tumours, known for its potential to grow extensively and cause severe bone deformity. Ameloblastoma arises from remnants of the tissues involved in tooth formation and represents approximately 1% [2] of all oral ectodermal tumours and 11% of all odontogenic tumours. It predominantly occurs in the third, fourth, and fifth decades of life, with a higher incidence in males, and most commonly affects the posterior mandible. Around 50-80% of cases are associated with an unerupted or impacted tooth. Ameloblastoma can be classified into unicystic, multicystic, peripheral, and intraosseous subtypes. Histopathological variants include follicular, plexiform, acanthomatous, and granular cell types, with uncommon variants such as desmoplastic, basal cell, clear cell, and keratoameloblastoma [3].

Clinically, ameloblastoma is characterised as a slow-growing, locally aggressive lesion, which can cause mobility and root resorption in the associated teeth. Radiographically, it appears as unilocular or multilocular radiolucencies with well-defined borders. The multilocular appearance resembles soap bubbles, indicating the presence of multiple bony spaces separated by trabeculae. Treatment options for ameloblastoma include enucleation, curettage, marsupialisation, cryosurgery, electrocautery, sclerotherapy, radiotherapy, and segmental or complete resection with reconstruction. The first description of ameloblastoma was made by Cusack JW in 1827 [4], and the term "ameloblastoma" was suggested by Churchill in 1934 to replace the term "adamantinoma" coined by Malassez L [5] in 1885. Ameloblastoma is a benign epithelial odontogenic tumour that is often aggressive and destructive. It originates from various sources, including cell rests of the dental organ (such as remnants of the dental lamina (cell rests of Serres) or remnants of Hertwig's sheath), epithelium of odontogenic cysts (such as dentigerous cysts and odontomas), disturbances in

the development of the enamel organ, and basal cells of the surface epithelial rests of Malassez L of the jaws [6].

Ameloblastoma occurs predominantly in young adults, with a mean age of 35 years, and shows a higher incidence in males. It is more commonly found in the mandible than in the maxilla, with a ratio of 13:1. It accounts for approximately 97.4% of mandibular cases, with the body, ramus, and angle being the most frequently affected sites. In the 2017 classification by the World Health Organisation (WHO) [7], ameloblastoma is categorised as conventional/solid/multicystic (86% of cases), unicystic (13% of cases), peripheral/extraosseous (about 1% of cases), and malignant ameloblastoma. Typically, ameloblastoma is asymptomatic and exhibits slow growth, although it may rarely cause pain and paresthesia. The lesion is commonly associated with the crowns of mandibular third molars but can also be found in interradicular, periapical, and edentulous regions. It can penetrate the cortical bone and cause tooth resorption. Conventional ameloblastoma often infiltrates the cancellous bone trabeculae at the periphery of the lesion before radiographic evidence of bone resorption becomes visible. Due to its local invasiveness and destructive nature, ameloblastoma can lead to deformity of the jaws, masticatory dysfunction, and abnormal jaw movement, including involvement of the Temporomandibular Joint (TMJ). Radiographically [8], ameloblastoma presents with soap bubble-like, honeycomb, or eggshell cracking appearances, which may mimic other tumours such as odontogenic keratocysts, aneurysmal bone cysts, or giant cell tumours. Differential diagnosis includes dentigerous cysts, odontogenic keratocysts, odontogenic myxomas, aneurysmal bone cysts, fibrous dysplasia, odontomas, and osteosarcomas [9]. In this particular case, the patient was a female in second decade of life who had a history of recurrent ameloblastoma at the same site. The initial surgical treatment performed was marginal resection. The last recurrence occurred three years ago, and now the patient has presented for the third time with radiological findings suggestive of a multilocular radiolucency. According to a study by Li Y et al., [10], radiographic examinations can be used for diagnosing and planning the treatment of ameloblastoma, especially in cases of unicystic ameloblastomas characterised by a fibrous cyst wall lined by ameloblastic epithelium.

Histopathologically, follicular ameloblastoma is characterised by islands of epithelium resembling enamel organ epithelium with a core of stellate reticulum-like cells embedded in a mature fibrous connective tissue stroma. A plexiform pattern is characterised by long, anastomosing chords or sheets of odontogenic epithelium. Unicystic ameloblastoma, on the other hand, presents as a circumscribed unicystic lesion lined by ameloblastic epithelium and containing eosinophilic to haemorrhagic material [Table/Fig-7]. The underlying dense fibrous stroma shows proliferating fibroblasts, scattered osteoclast-like giant cells, areas of newly formed woven bone trabeculae, and vascular channels. Radiographs provide valuable information about bone destruction, the presence of multiple locules, and bone reactions.

A literature review revealed that the most common histologic pattern of ameloblastoma was follicular (48.1%), followed by reticular (37%), basaloid (7.4%), and mixed (7.4%) [11]. In males, the predominant tumour patterns were reticular (50%), follicular (35%), mixed (10%), and basaloid (5%). In females, the most common type was follicular (six out of seven cases; 85.7%), followed by basaloid (14.3%). Various treatment modalities have been employed, including electrocautery, cryosurgery, injection of sclerosing agents, enucleation, curettage, marsupialisation, cryosurgery, and segmental or complete resection with reconstruction. Recurrence rates were reported by Lau SL and Samman N as 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application (which consists of 60% ethanol, 30% chloroform, 10% glacial acetic acid, and 1 gram of ferric chloride), and 18% for marsupialisation followed by enucleation [12]. Simple treatment with enucleation or curettage often presents a high potential for recurrence (60-80%). Therefore, the interpretation of clinical and imaging data is crucial for early diagnosis and effective treatment of ameloblastoma. The therapeutic decision should aim to eliminate the pathology, considering the treatment method's morbidity and the patient's quality of life during rehabilitation.

According to a retrospective study conducted by Vayvada H et al., involving 21 patients with mandibular ameloblastoma who underwent segmental mandibulectomy with free fibula graft reconstruction, was conducted [13]. Only four patients experienced complications or recurrence during an average follow-up of five years [13]. Segmental mandibulectomy with free fibula graft reconstruction is considered an ideal treatment method for mandibular ameloblastoma, providing satisfactory functional and aesthetic results. Another retrospective study by Ooi A et al., reviewed 30 consecutive patients with unicystic ameloblastoma who were treated with segmental mandibulectomy and free fibula graft reconstruction [14]. At an average follow-up of five years, no tumour recurrences were observed, and 96% of the patients reported satisfaction with their appearance.

Due to its slow growth, ameloblastoma can reappear after several years. Therefore, annual follow-ups for at least 10 years are recommended. Despite undergoing laser ablative surgery twice, the patient experienced a recurrence within five years. Complete resection of the lesion, involving wide surgical excision with clear margins, is crucial to prevent recurrences due to the aggressive nature of ameloblastoma.

CONCLUSION(S)

The treatment of marginal resection with a safety margin proved to be ineffective in this reported case of mandibular ameloblastoma. Due to multiple recurrences, segmental resection with free fibula reconstruction was performed. However, long-term follow-up remains crucial in light of the local aggressiveness and high recurrence rate associated with this condition. If the patient had undergone complete resection of the lesion during the initial episode, it would have ultimately reduced the time, cost, number of surgical procedures, and improved functional and aesthetic outcomes. Therefore, factors such as the patient's age, the aggressive nature of the lesion, its location, and the high likelihood of recurrence should all be carefully considered, before deciding on any surgical interventions.

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PARTICULARS OF CONTRIBUTORS:
1. Professor, Department of Oral Medicine and Radiology, Karpaga Vinayaga Institute of Dental Sciences, Chengalpattu, Tamil Nadu, India.
2. Senior Lecturer, Department of Oral Medicine and Radiology, Karpaga Vinayaga Institute of Dental Sciences, Chengalpattu, Tamil Nadu, India.
3. Senior Lecturer, Department of Oral Medicine and Radiology, Karpaga Vinayaga Institute of Dental Sciences, Chengalpattu, Tamil Nadu, India.
4. Postgraduate, Department of Oral Medicine and Radiology, Karpaga Vinayaga Institute of Dental Sciences, Chengalpattu, Tamil Nadu, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. KR Shakila,
Professor, Department of Oral Medicine and Radiology, Karpaga Vinayaga Institute of Dental Sciences, Chinnakolambakkam, Madhuranthagam,
Chengalpattu-603308, Tamil Nadu, India.
E-mail: shakilasuresh100@yahoo.com

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