



## Unicystic Ameloblastoma - Case Report

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### Abstract

Ameloblastoma is a slow-growing locally aggressive, benign odontogenic epithelium with a greater tendency for recurrence and involves aggressive bone loss and teeth. It is believed to be derived from cell rests of the enamel organ, either remnants of dental lamina or Hertwig's sheath, the epithelial rest of Malassez.

The most common site of preference is the mandibular molar and ramus area than the anterior and premolar region. The article presents two case series of unicystic ameloblastoma of a 25 year female who had a localized swelling in the left side of the mandible which resembled a keratocystic odontogenic tumor clinically and radiographically. Based on histopathological examination the final diagnosis was unicystic ameloblastoma with the mural pattern. The second case report presented a 32-year female with a diffuse swelling in the left lower jaw which histopathologically revealed ameloblastoma with the plexiform variant. The case series presented certain unique features of female predilection and a multilocular variant with mural and plexiform pattern unicystic ameloblastoma. Both the patient was surgically managed more conservatively with enucleation and cauterization as cystic types are found to be less aggressive compared to solid variants of ameloblastoma. They were on a regular follow period postoperatively.

**Keywords:** Odontogenic tumor; Ameloblastoma; Mandible; Recurrence

### Introduction

Ameloblastoma are categorized broadly into three biologic variants: Cystic (unicystic), solid, and peripheral. The cystic variant is biologically less aggressive and features a better response to enucleation or curettage than does the solid ameloblastoma [1]. Ameloblastoma is considered a tumor of an odontogenic variant that has developed from epithelial cellular elements and dental tissues in various stages of development.

An Unicystic Ameloblastoma (UA) represents an ameloblastoma variant, that's a cyst [2]. In 1977, Robinson and Martinez first used the term "Unicystic Ameloblastoma" (UA) for such lesions [3], but the other name recognized by WHO is "cystogenic ameloblastoma" [2]. They are often related to an unerupted third molar [4], with a peak incidence within the third and fourth decade of life. Six radiographic patterns are identified for unicystic ameloblastoma, well-defined unilocular to multilocular ones. When the radiographic appearance is split into two main patterns, unilocular and multilocular, a unilocular pattern is usually associated in cases with impacted teeth [5].

### Case Series

#### Case 1

A 25-year-old young female patient presented with the chief complaints of gradual increasing painless swelling over the left side of the lower jaw for the past six months (Figure 1). The patient had an altered sensation over the left cheek region.

On clinical examination extra orally revealed a diffuse, swelling on the left side of the mandible approximately 4.5 cm × 4.5 cm. The swelling extends anteriorly from the parasymphysis region of the mandible to the body of the mandible posteriorly. On intraoral examination, diffuse swelling extends from 33 to 37 with buccal and lingual expansion. Bicortical expansion of swelling was evident from 33 to 38 regions. On palpation, the swelling was hard in consistency with smooth surface and non-tender. Pain-related to the swelling was dull, non-radiating, and intermittent. Mobility of teeth present in 34, 35, 36, 38 with missing 37 (Figures 2-4). A provisional diagnosis of odontogenic keratocyst was given.

**Panoramic radiograph:** Revealed well-defined multilocular radiolucency with large locules resembling soap bubble appearance with thin corticated borders in the left side of the mandible extending from 33 to 38 with resorption of roots in 33, 34, 35 and cervical resorption of roots of 36

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Received Date: 18 Oct 2022

Accepted Date: 03 Nov 2022

Published Date: 07 Nov 2022

#### Citation:

Gayathri Devi M, Bakyalakshmi K,  
Cap Regu P. Unicystic Ameloblastoma  
- Case Report. *Ann Surg Case Rep.*  
2022; 5(2): 1063.

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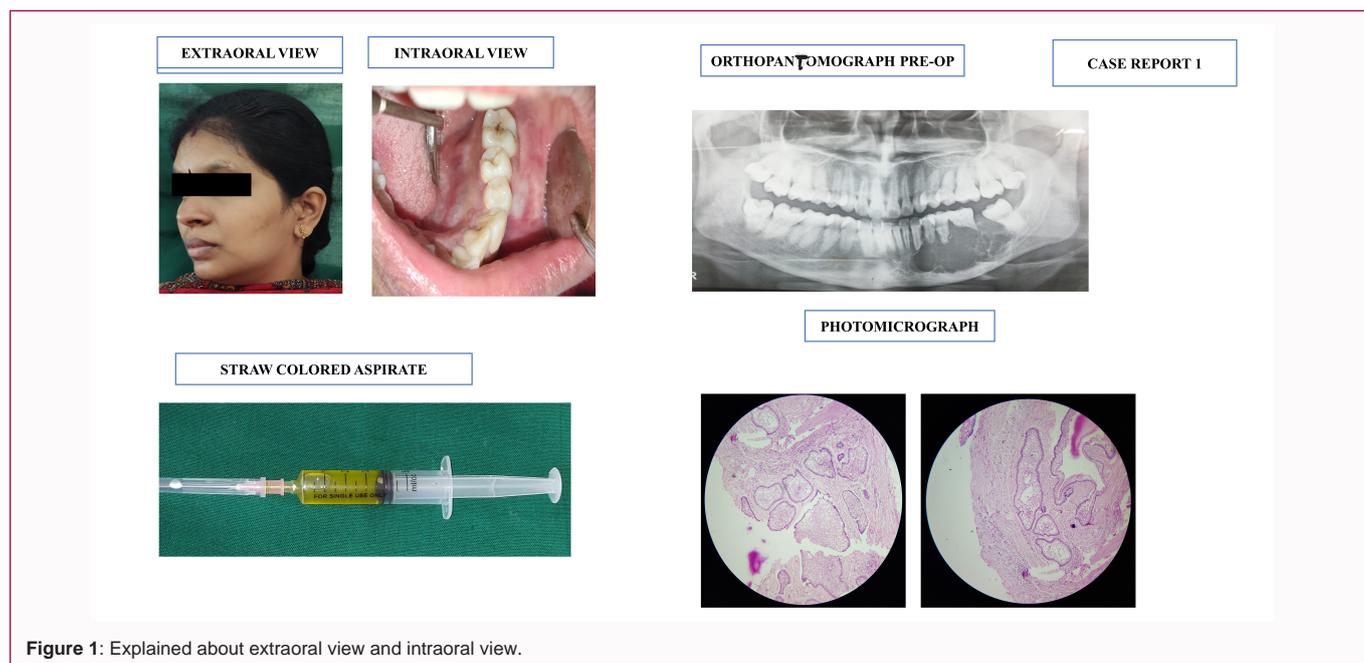


Figure 1: Explained about extraoral view and intraoral view.

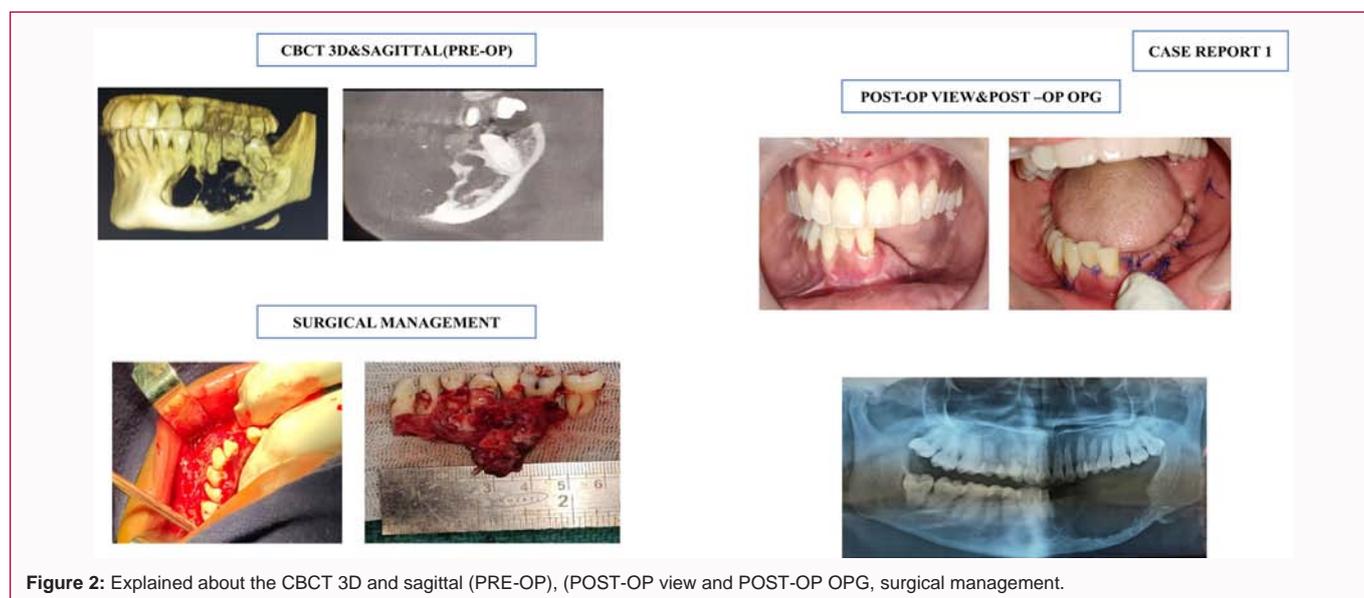


Figure 2: Explained about the CBCT 3D and sagittal (PRE-OP), (POST-OP view and POST-OP OPG, surgical management.

(Figure 5a).

**CBCT:** Of mandible presented with multilocular hypodense, ill-defined corticated border, bony erosions, obliteration of inferior alveolar nerve canal, root resorption of 33, 34, 35, 36 and bicortical expansion of buccal and lingual plates. The radiological diagnosis based on clinical correlation as odontogenic keratocyst was given.

On fine needle aspiration, straw-colored fluid was obtained. Routine blood investigation was done before the biopsy procedure. An incisional biopsy was done to get a definitive diagnosis.

The histopathological section shows cystic lesions characterized by odontogenic epithelial lining with mural follicular growth pattern showing peripheral tall columnar ameloblast like cells and inner stellate reticulum-like cells in the connective tissue wall. Reactive bone is seen in the periphery of the lesion. The biopsy was confirmed unicystic ameloblastoma.

The patient was surgically managed with enucleation and chemical cauterization. Peripheral osteotomy is done with vulcanite bur. A postoperative panoramic radiograph was taken and was found to be satisfactory. The patient was on regular follow-up.

**Case 2**

A 32-year female reported a diffuse swelling of the lower jaw for the past five months. Extraoral examination revealed a facial asymmetry with diffuse ill-defined swelling on the left body, angle, and extending to the ramus of the mandible. Superiorly involving inferior border of the mandible, inferiorly submandibular region, medially submental region and posteriorly ramus of mandible intraorally mild swelling with bicortical expansion, non-tender, firm in consistency with a smooth surface was evident from 34 to 37 regions. Mobility present in relation to 33, 34, 35, 36, 37. On aspiration, clear straw-colored fluid was obtained.

Panoramic radiograph revealed well-defined multilocular

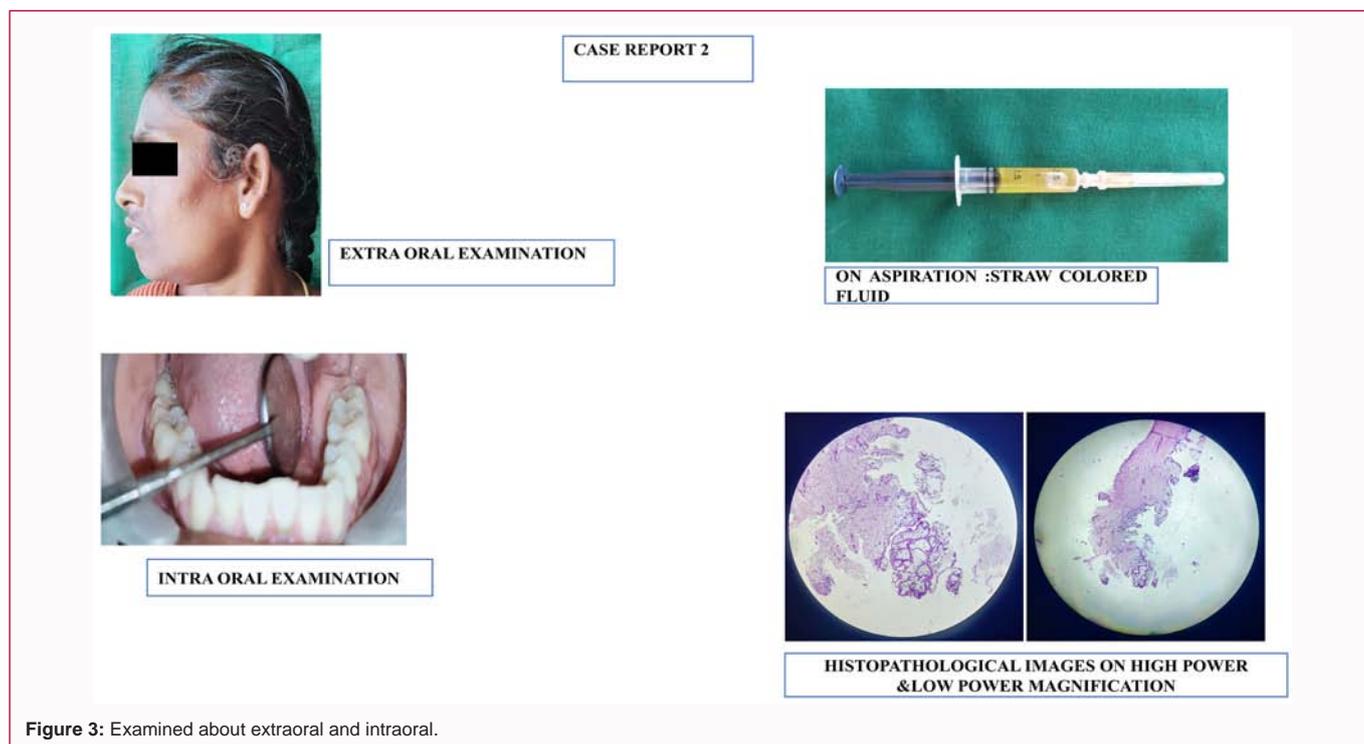


Figure 3: Examined about extraoral and intraoral.

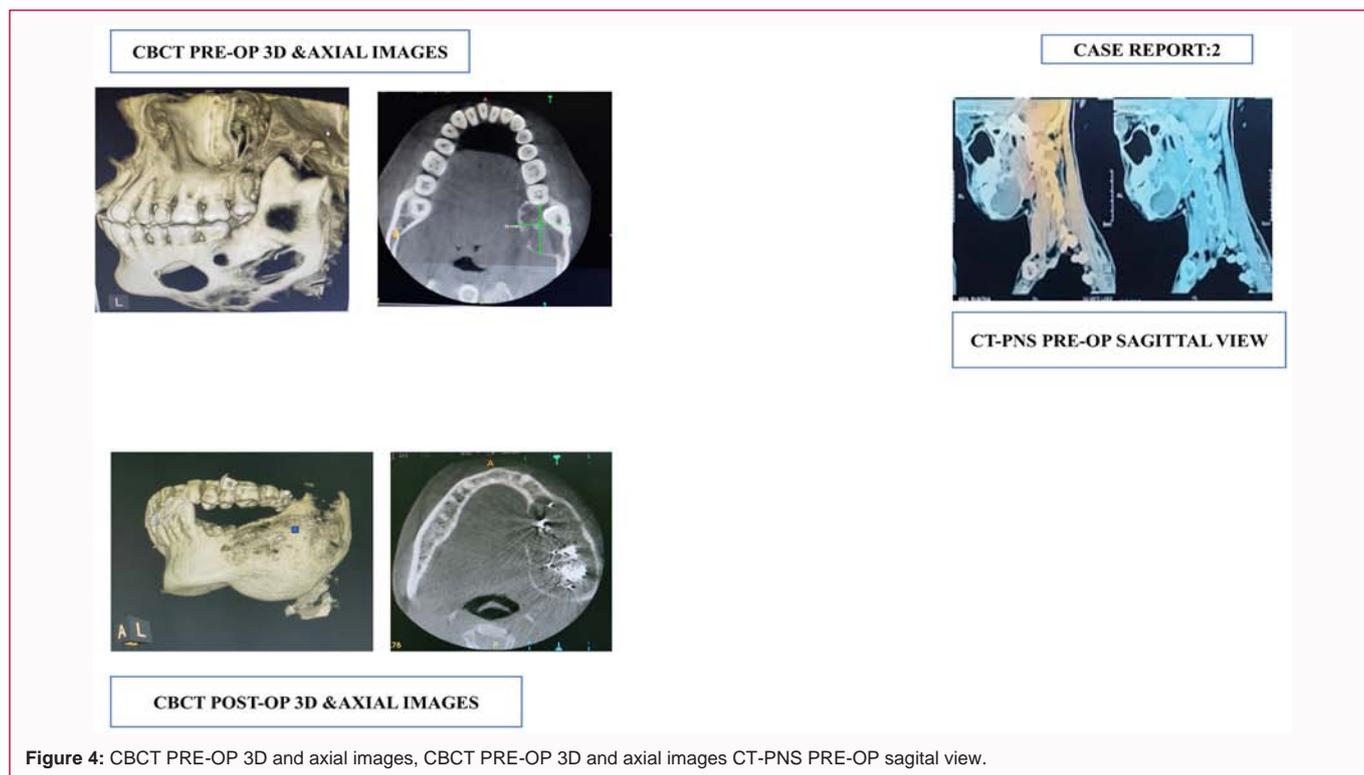


Figure 4: CBCT PRE-OP 3D and axial images, CBCT PRE-OP 3D and axial images CT-PNS PRE-OP sagittal view.

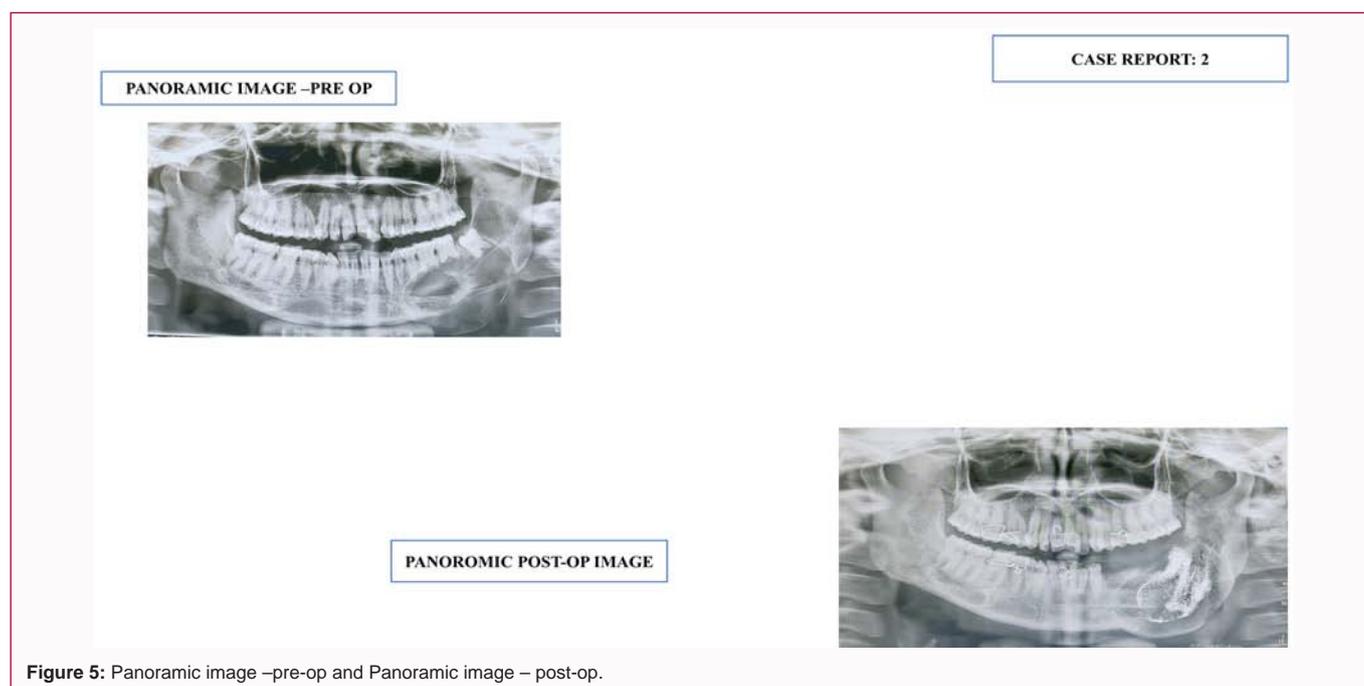
radiolucency with large locules resembling soap bubble appearance with thin corticated borders in the left body, ramus of the mandible, and impacted 38 within the multilocular radiolucency. Resorption of roots in 35, 36, 37 was evident (Figure 3).

**CT neck:** 4.3 cm × 6.5 cm sized well-defined expansile lytic lesion involving angle of mandible with a small extension into the body and left ramus; focal areas of the cortical breach; the cyst is associated with an unerupted left third molar which was suggestive of dentigerous

cyst.

**CBCT:** Multilocular hypodense expansile lesion with obliteration of inferior alveolar nerve canal, expansion of bicortical plates with root resorption in 35, 36, 37 and impacted 38 which had a differential diagnosis of dentigerous cyst and ameloblastoma (Figure 4, 5).

Routine blood investigation was done before the biopsy procedure. An incisional biopsy was done to get a definitive diagnosis.



**Figure 5:** Panoramic image –pre-op and Panoramic image – post-op.

Histopathology revealed fragments of cystic lesions characterized by a luminal proliferation of odontogenic epithelium in a plexiform pattern and a fibrous wall. Peripheral reactive bone was also noted. Histopathology was suggestive of unicystic ameloblastoma. The patient was surgically managed with enucleation and chemical cauterization. Peripheral osteotomy was done. A postoperative panoramic radiograph was taken and was found to be satisfactory. The patient was on regular follow-up.

## Discussion

Eversole et al. and Paikkatt et al. identified predominant radiographical patterns for unicystic ameloblastoma as unilocular, scalloped, multilocular, pericoronal, inter radicular, or periapical expansile radiolucencies [6]. Unicystic ameloblastoma have a unilocular pattern and both cases had the multilocular variant. Ameloblastoma is considered a slow-growing aggressive tumor with a high rate of recurrence if inadequately removed [7].

WHO system for ameloblastoma in 2003, based on differences in biological behavior, treatment plan, and recurrence rate classified as:

- 1) Classic solid/Multicystic ameloblastoma,
- 2) Unicystic ameloblastoma,
- 3) Peripheral ameloblastoma
- 4) Desmoplastic ameloblastoma, including the so-called hybrid lesions [8].

In 1970, Vickers and Gorlin were the first to describe the early ameloblastic changes within the cyst wall and their histological criteria included for diagnosis was a cyst lined by ameloblastic epithelium with a tall columnar basal layer, subnuclear vacuoles, reverse polarity of the hyperchromatic nucleus, and a thin layer of edematous, degenerating stellate reticulum-like cells on the surface [9]. When the thickened lining penetrates nearby tissues, it is usually a mural unicystic form [10].

Ackermann based on histological pattern classified the unicystic

ameloblastoma [11].

Group I-luminal UA (tumor confined to the luminal cyst surface);

Group II-intraluminal/plexiform UA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall);

Group III-mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).

By Ackermann's classification, our case report belongs to Group III mural unicystic ameloblastoma. Unicystic Ameloblastoma (UA) accounts for 10% to 15% and least encountered. The gender distribution shows a slight male predilection and most of the unicystic types are associated with impacted mandibular molar [12]. The case report 1: Presented here was a young female with mural unicystic pattern, cyst not involving impacted teeth and had a multilocular variant and in case 2 middle-aged female with plexiform variant, cyst involving impacted teeth and multilocular variant. The definite diagnosis of unicystic ameloblastoma can be made by histopathological examination. Surgical management was done conservatively to preserve bone loss in both cases.

The recurrence rate for unicystic ameloblastoma after conservative surgical treatment whether curettage or enucleation was generally reported to be 10% to 20% [13]. The patient was under regular follow-up visits for any recurrence.

## Conclusion

Unicystic ameloblastoma with a mural growth pattern has a tendency for recurrence if inadequately managed surgically. Proper management avoids the propensity of recurrence rate. Unicystic ameloblastoma with mural and plexiform pattern involving impacted and not involving impacted tooth with a multilocular radiolucency is the characteristic of this case report with proper surgical management and follow-up.

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