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Case Report

Clinical Outcome of Treating Young Patient of Hybrid Ameloblastoma of Mandible, by Conservative Method: A Case Report

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Abstract

Ameloblastoma is a common benign odontogenic tumor, representing about 10% of jaw neoplasms, characterized by slow growth yet local invasiveness and a high recurrence rate. Its global incidence is approximately 0.92 cases/million person-years, with variations in prevalence across different regions. Studies conducted in China and India have highlighted its prominence. Diagnosis typically involves imaging techniques such as computed tomography scans and biopsies, with histopathological examination being crucial for definitive diagnosis. Ameloblastoma primarily consists of follicular and plexiform types, with acanthomata's and granular cell varieties following. Rare forms include desmoplastic, basal cell, clear-cell, keratoameloblastoma, and papilliferous types. A hybrid variant has been identified, displaying features of both plexiform and conventional follicular ameloblastoma. The current case exemplifies a hybrid classification combining plexiform and follicular. Ameloblastoma is a prevalent odontogenic tumor in India, with a prevalence of about 4.83 cases/100,000. Surgical resection is the primary treatment, with solid/multicystic types typically requiring marginal or segmental resection to reduce recurrence risk. Unicystic ameloblastomas may be treated conservatively, often with enucleation and adjuvants such as Carnoy's solution, especially in younger patients. Radical resection is preferred for larger defects, offering better disease-free survival, while conservative methods have higher recurrence rates, necessitating long-term follow-up and individualized treatment planning based on tumor characteristics.

Key words: Ameloblastoma, Conservative treatment, Enucleation, Tumors

INTRODUCTION

Ameloblastoma is a prevalent yet perplexing neoplasm of the jaw, accounting for approximately 10% of all tumors that develop in the mandible and maxilla. It is among the most prevalent benign odontogenic neoplasms. Its enigmatic nature is evidenced by a sluggish growth rate akin to benign tumors, yet it exhibits locally invasive properties, a high recurrence rate, and metastatic potential comparable to malignant tumors.[1]

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Ameloblastoma exhibits a heterogeneous geographic occurrence, with a global incidence of 0.92 cases/million person-years.[2] Numerous epidemiological researches have indicated that ameloblastoma is either the most prevalent or the second most prevalent benign odontogenic tumor. A Brazilian investigation of 6,231 oral lesions identified 185 (3%) as odontogenic tumors, all of which were benign. The most prevalent lesions were ameloblastomas (29%), followed by keratocystic odontogenic tumors (28%) and odontomas (19%).[3] The literature published by Avelar *et al.*, in 2011, Mehngi *et al.*, in 2018 and Intapa, in 2017 reports that ameloblastoma is the second most prevalent odontogenic tumor, predominantly manifesting in the posterior jaw. The majority of instances transpired throughout the second to fourth decades of life. The maximum incidence in Africa and South America occurred in the third decade, whereas in Europe and North America, it manifested in the fifth and sixth

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decades. Ameloblastoma appears to be more common in developing nations such as India, where it manifests at a younger age. This gap in the demographic distribution of ameloblastoma may be ascribed to an accelerated aging process resulting from inadequate nutrition and restricted access to healthcare services.[4-6] In the United States and Canada, ameloblastoma ranks as the second most prevalent odontogenic tumor, following odontoma.[7,8]

The most extensive study among Indian research was conducted in the Marathwada district of Maharashtra, involving a review of histopathological records from 1992 to 2012. Among the 125 benign odontogenic tumors, the keratocystic odontogenic tumor was the most prevalent at 45%, followed by ameloblastoma at 35%, odontoma at 7%, and adenomatoid odontogenic tumor at 5%.[9]

The diagnosis of ameloblastoma involves imaging techniques, primarily computed tomography (CT) scans and biopsies. CT scans reveal well-defined, uni- or multilocular radiolucent lesions, while dental X-rays may show lytic lesions with scalloped margins. Multilocular ameloblastomas exhibit a "soap bubble" appearance. Magnetic resonance imaging is preferred for maxillary cases and desmoplastic ameloblastoma due to its superior soft-tissue characterization. Definitive diagnosis relies on histopathological examination, distinguishing ameloblastoma from other conditions. Pre-operative staging for malignant cases is also conducted through biopsy, with techniques such as trucut needle biopsy or incisional biopsy employed based on the lesion's location.[10,11]

Waldron and El-Mofty identified a unique type of ameloblastoma that histologically has regions of conventional follicular or plexiform ameloblastoma alongside regions of desmoplastic ameloblastoma. This was designated as a hybrid ameloblastoma (HA).[12]

HA is an uncommon variant that integrates the histological characteristics of desmoplastic and ordinary ameloblastoma. Desmoplastic ameloblastoma, a rare condition, histopathologically comprises odontogenic epithelium of different sizes embedded within a highly collagenized connective tissue stroma, including ill-defined proliferating islands and cords.[13]

Some authors remain uncertain if desmoplastic alterations in the stroma are a consequence of underlying unicystic/multicystic types or if desmoplastic changes pertain to other types.[14]

To the best of our knowledge, a few cases of the hybrid type of desmoplastic ameloblastoma have been published as per our literature search.

CASE REPORT

A 19-year-old female patient presented to the Department of Oral and Maxillofacial Surgery with a chief complaint of painless swelling on the left side of the lower jaw for 4 months. The swelling began small and progressively grew in size, causing mild facial asymmetry. There was no trauma, paresthesia, or systemic disease. Extraoral inspection presented a diffuse, hard swelling in the premolar-molar area of the left mandible, resulting in expansion of the buccal cortical plate. Intraorally, there was a well-delineated, bony hard swelling from the area of 35–38 with obliteration of the buccal vestibular area and intact overlying mucosa [Figure 1].

Orthopantomogram showed a multilocular lesion with well-defined margins separated by multiple septa, giving it a multilocular appearance, and seen extending from the left canine to the third molar area, with thinning and minimal perforation of the cortical plates with displaced and impacted mandibular second and third molars. Resorption of the root apices of involved teeth was also noted. Cone-beam CT also established buccal and lingual cortical expansion with minimal perforation areas and displacement of adjacent teeth. Contrast-enhanced CT of the site does not suggest any soft-tissue invasion of the lesion. Incisional biopsy was done, and histopathological study revealed desmoplastic ameloblastoma features in parts with dense collagenous stroma, squeezing odontogenic epithelial islands, and typical plexiform patterns of conventional ameloblastoma in other sections, establishing the diagnosis as HA [Figures 2 and 3].

The lesion was treated conservatively under general anesthesia by enucleation, curettage of deeply situated ameloblastomatous content in the region of the condylar and coronoid processes, followed by extraction of involved premolars and first molar teeth, along with removal of

Figure 1: Pre-operative clinical photographs

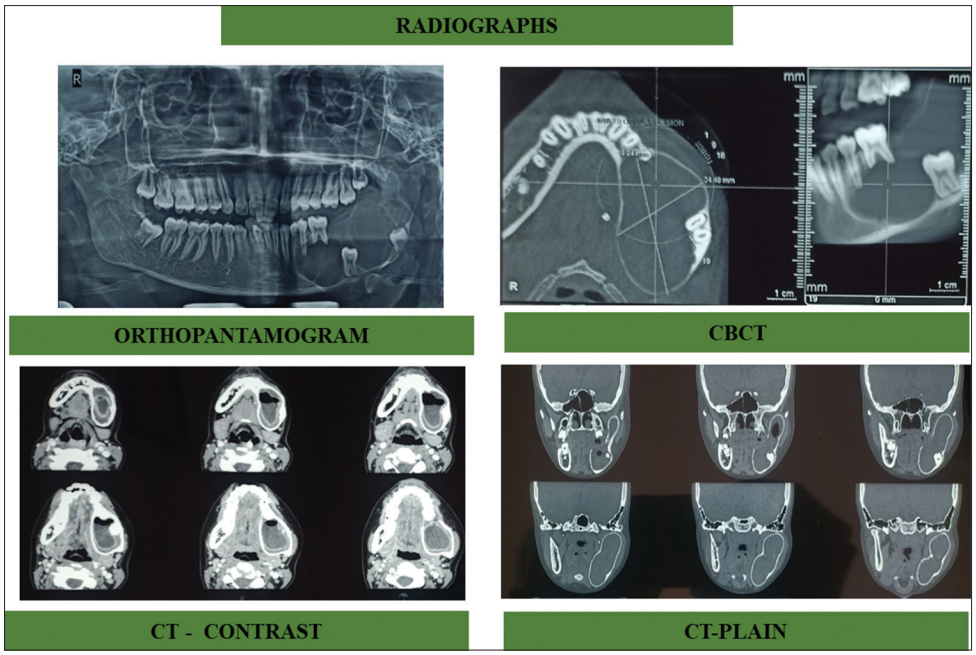

Figure 2: Pre-operative radiographs and cone-beam computed tomography (CT), CT

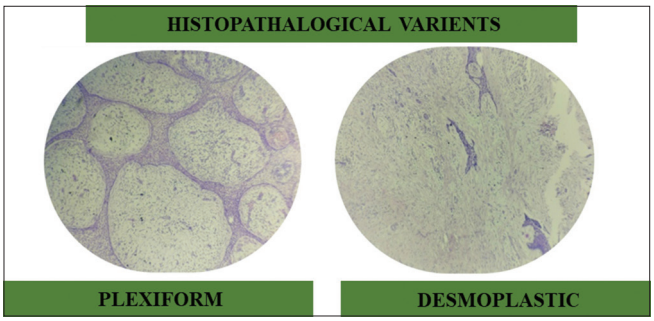

Figure 3: Histopathological slides of the hybrid ameloblastoma

deeply impacted and displaced second molar and third molar tooth buds. After complete curettage of the lesion, chemical cauterization was done using modified carnoy's solution. The wound was closed primarily at the time of operation, but gaping and inward rolling of soft-tissue margins was observed on post op day 12. Then the patient was advised an obturator plate to avoid any food debris to stuck inside the bony cavity. In the upcoming post-operative course, the site was regularly irrigated with the dilute antiseptic solution with frequency of twice to three times daily, and post-discharge, and patient's parents were educated about the thorough irrigation of the site, and the patient was regularly followed up for almost 2½ years with no evidence of recurrence clinically or radiographically [Figures 4 and 5].

Conservative treatment (enucleation ± curettage ± chemical cauterization) was selected for this HA to meet oncologic control with maintenance of form and function of the jaw as the tumor was anatomically accessible, and to

Figure 4: Intraoperative surgical procedure

liberate the patient from functional or esthetic morbidity of radical treatment. While pooled analyses indicate increased recurrence rates following conservative treatment as opposed to radical resection, some recent series and case reports illustrate acceptable control using conservative methods when strict selection is applied (intact cortical plates or unicystic component, young patient, feasibility of long-term follow-up), and when adjuncts such as chemical cauterization, peripheral ostectomy or staged dredging are added to minimize residual tumor load. Conservative therapy thus constitutes a logical, patient-oriented choice for certain hybrid lesions in which retention of jaw ultimate quality of life, as long as the patient is informed regarding

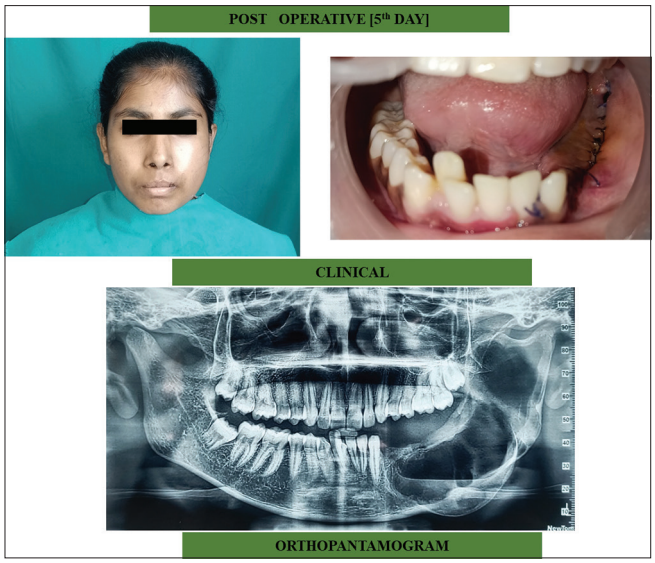

Figure 5: 5th Day clinical and radiographical investigation for healing assessment

the increased risk of recurrence and entered into close, long-term radiographic follow-up so the recurrences will be recognized early and quickly treated. Here, the major factor was the age consideration of the patient, and to avoid post-operative morbidity.[12,13]

The patient was followed up for 2½ years of time after which the periodic evaluation was done clinically and radiographically. There was a sound decrease in the extraoral swelling in the immediate post-operative period [Figure 6]. Moreover, at 6 months, a sound bone formation with a distinct trabecular pattern was observed radiographically along with satisfactory intraoral healing [Figure 7].

DISCUSSION

Consensus on the surgical management of HA is lacking, as the biological behavior of this unusual entity remains undetermined.[14] Numerous papers have delineated treatment regimens for ameloblastoma. A multitude of surgeons favor a drastic surgical intervention, specifically excision, for the management of all variants of ameloblastoma, owing to its challenging characteristics, including local aggressiveness and a propensity for recurrence.[15,16] Escande *et al*. proposed that unilocular ameloblastomas of <5 cm in diameter, characterized by many medium-sized "soap bubbles," should be managed conservatively with enucleation and curettage.[17] The Carnoy solution serves as a chemical curettage agent for treating benign jaw lesions, especially keratocysts, owing to its cauterizing, penetrating, and fixing capabilities.[18] Numerous studies have indicated that the supplementary application of Carnoy's solution after enucleation or curettage of aggressive lesions, such as ameloblastoma, may yield certain advantages.

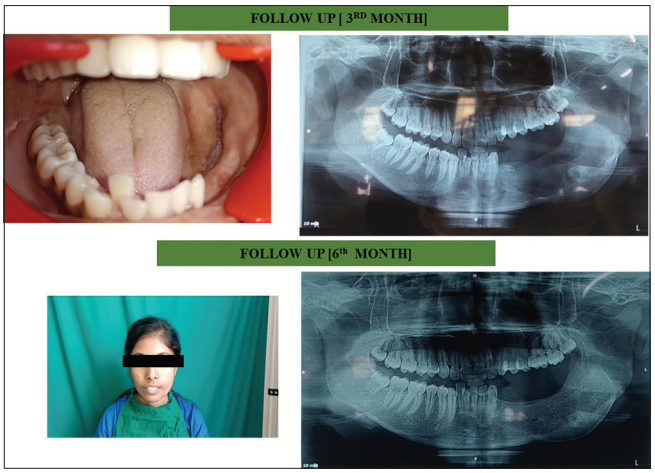

Figure 6: Follow-up at 3 months and 6 months

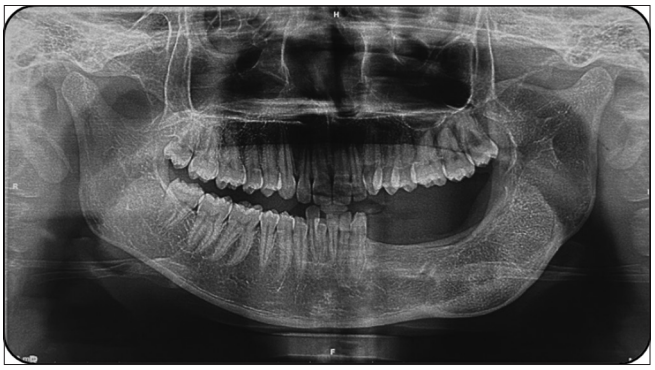

Figure 7: Follow-up radiograph at 2 years 6 months

Lee *et al*. conducted a retrospective study indicating a reduced recurrence rate following the application of Carnoy's solution for 3 min post-enucleation of unicystic ameloblastomas with 93% mural invasion. Lau and Samman reported a recurrence rate of 3.6% following resection, 30.5% with enucleation alone, and 16% with the use of Carnoy's solution after enucleation. Thirteen, the findings of the available studies suggest that enucleation of ameloblastoma may effectively eliminate leftover ameloblastoma tissue and diminish the chance of recurrence, but this conclusion is based on only two studies cited.[19,20] In this case, we opted for enucleation, curettage, and chemical fixation with Carnoy's solution to preserve the inferior alveolar nerve and enhance quality of life by prioritizing aesthetics and function, resulting in successful outcomes.

CONCLUSION

We demonstrate that HA in the younger demographic can be effectively managed with a conservative approach, specifically enucleation accompanied by bone curettage. We opted for cautious surgery in this case, partly due to the patients' youth.

In every instance, prolonged monitoring using radiographic imaging is essential. We continue to do annual surveillance of our patients by radiography and, if necessary, biopsies.

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