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Effect of radiographic & histopathologic variants in management of ameloblastoma: An original research

Dr. Bharathiram Guduri
Senior lecturer, Dept. of Oral & MaxilloFacial Surgery, GSL Dental College, Rajahmundry
Email: drgbharathiramomfs@gmail.com

Dr. Shraddha C. Jugade
Assistant Professor, Oral Medicine and Radiology, Dr. D Y Patil Dental College & Hospital, Dr. D Y Patil Vidyapeeth, Pimpri, Pune
Email: jugadeshraddha@gmail.com

Dr. Saurabh Shekhar
Lecturer, Oral pathology, AIMST University, Bedong, Kedah, Malaysia
*Corresponding author email: saurabhshekhar2001@gmail.com

Dr. A. Rashmi Pillai
MDS, Oral and Maxillofacial Surgeon, Rashmis Dentofacial Multispeciality Centre, Kavanad, Kollam, Kerala
Email: drarashmipillai@gmail.com

Dr. Ankita Sharma
Senior Lecturer, Dept of Pathology, NIMS Jaipur
Email: ankitasha313@gmail.com

Dr. Amrita Das
Senior lecturer, Department of Periodontology and Implantology, Dr. HSRSM dental college , Hingoli. Maharashtra, India
Email: das.amrita94@gmail.com

***Abstract*---**Introduction: Ameloblastoma is a rare, benign odontogenic tumor that accounts for 1% of all tumors of the jaws. We aim to compare the ameloblastoma recurrence rate according to the type of treatment: radical or conservative and as per the radiographic, histologic features. Material and methods: we conducted a retrospective study and collected the data based on the radiological, and histological and the type of treatment: conservative or radical and to compare the recurrence rate according to the type of treatment.

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Results: Thirty Patients were included, managed by conservative treatment (CT) in 26 cases and radical treatment (RT) in 14 cases. The recurrence rate was 90.9% in the CT group and 9.1% in the RT group (P = 0.025) with a mean follow-up of 56.2 months. Conclusion: The recurrence rate after conservative treatment was higher than that after radical treatment. The choice of treatment must be adapted to the macroscopic and histological characteristics of each tumor and to the patient.

***Keywords*---**ameloblastoma, management, radiographic, histologic features.

Introduction

Ameloblastoma is a rare, benign, slowly-growing odontogenic tumor. It accounts for 1% of all tumors of the jaws and 11% of all odontogenic tumors [1]. Ameloblastomas are described by an aggressive potential for local invasion and a high recurrence rate, needing a precise histological diagnosis and surgical treatment, the modalities of which have not yet been well defined. Although conservative treatment (marsupialization, enucleation, curettage) preserves integrity of the bones and allows continued growth of the mandible [2,3], it appears to be associated with a high recurrence rate, ranging between 55% and 90% [4], while radical treatment can leave major cosmetic and functional sequelae and may require free flap reconstruction [5,6]. The selection between these two treatment modalities therefore appears to be an essential issue in the management of these tumors. Hence the purpose of this study was to compare the ameloblastoma recurrence rate according to the type of treatment: radical or conservative and as per the radiographic, histologic features.

Material and Methods

We conducted a retrospective study from 200-2019. We collected demographic data, clinical features as radiographic presentation: unilobed defect, multilobed defect, cortical bone invasion, root resorption, impacted tooth, soft tissue infiltration, fracture;

- histological type: follicular, plexiform, mixed (follicular and plexiform), desmoplastic, unicystic, peripheral, acanthomatous, granular;
- first-line, second-line and third-line treatments: conservative or radical
- number of recurrences;
- Duration of follow-up.

Comparison was done between the types of the treatment and the management for the type of the lesion was done keeping the $P < 0.05$ was considered significant.

A total of 30 patients were included in the study and 27 were included in statistical analysis.

Results

The men were 60%. The mean age of the patients was 46.3 years±17.4 years. The common site was the mandible. The most common histological types were follicular (29.6%) (Table 1). The mean duration of follow-up was 56.2 months with a SD of 44.2 months. 22 events in the conservative treatment (CT) group and 14 events in the radical treatment (RT) group, as 8 patients required a second treatment for recurrence and 2 patients required a third treatment. The recurrence rate was significantly higher (90.9%) in the conservative treatment group than in the radical treatment group (9.1%) (P = 0.025) (Table 2).

Table 1
Radiographic characteristics presentation

Unicystic	15	55.5
Multicystic	10	37
Other		
Root resorption	9	33.3
Bone invasion	22	81.5
Soft tissue thickening	6	22.2
Histological type		
Follicular	8	29.6
Cystic	3	11.1
Folliculocystic	2	7.4
Fibroblastic	0	
Acanthomatous	2	7.4
Basaloid	1	3.7
Granular	1	3.7
Plexiform	6	22.2
Follicular and plexiform	0	
Folliculocystic and plexiform	1	3.7
Desmoplastic	0	
Unicystic	0	
Undetermined	3	11.1

Table 2
Recurrence rate

	Conservative treatment	Radical treatment	*P*
Recurrence	10 (90.9%)	1 (9.1%)	
No recurrence	12 (48%)	13 (52%)	
Total	22 (61.2%)	14 (38.8%)	0.025a

Discussion

In our study, the recurrence rate after conservative treatment was greater than that after radical treatment. The therapeutic management of ameloblastoma is a complex issue, as it must be as minimally destructive as possible due to the

benign nature of this lesion, but must be sufficiently extensive to prevent subsequent recurrence. Two substitute approaches are therefore proposed at the present time. The conservative approach can consist of enucleation or curettage, sometimes preceded by marsupialization. Enucleation has been exactly defined as dissection of an intraosseous cavity, while preserving its integrity [7]. When this definition cannot be completely seen, curettage must be associated, consisting of revision of the residual cavity using a drill (sphere or resin) or bone curette. The radical approach consists of segmental bone resection.

The issue of recurrence begins at the time of diagnosis: either a biopsy has been done prior to surgery to confirm the diagnosis and determine the histological type in order to propose radical or conservative treatment, or conservative biopsy resection is performed, with no possibility of reliable frozen section examination, and the suggestion for radical treatment is then decided on the basis of the histological results or after a surveillance period. Several studies have reported a higher recurrence rate after conservative treatment compared to radical treatment [8–14]. Our study results therefore consistent with previously published results. Apart from the study by Hong et al. in 2007 [3], which shown a significant difference ($P = 0.004$) based on direct comparison of the 2 groups, a review of the literature failed to demonstrate a statistically significant difference between conservative treatment and radical treatment. Our study confirms this tendency by reporting a higher recurrence rate in the conservative treatment group than in the radical treatment group.

However, several studies nevertheless recommend conservative treatment depending on the macroscopic appearance of the ameloblastoma. Similarly, Reichart et al. [5] distinguished unicystic forms from the other forms of ameloblastoma and recommended conservative treatment for this group of tumors. The histological type also appears to be an important factor determining the potential for recurrence of ameloblastomas. Hong et al., in a series of 239 patients published in 2007 [3], reported a higher recurrence rate for follicular tumors (25%) with no cases of recurrence for extraosseous/peripheral and desmoplastic tumors. Ruhin-Poncet et al. [6] also compared recurrence rates according to histological type in a series of 109 patients: 5 patients with a follicular form presented one recurrence and 16 patients presented two recurrences. Nakamura et al. [2], in a series of 78 cases, reported a higher recurrence rate for follicular (26.3%), plexiform (21.7%) and mixed (follicular and plexiform) (33.3%) tumors, regardless of the type of treatment.

These various studies led to the new WHO classification in 2005, which distinguishes various macroscopic types of ameloblastomas: solid/multicystic, extraosseous/peripheral, desmoplastic, unicystic, allowing the most appropriate treatment to be proposed for the most common histological types. According to the WHO histological classification, extraosseous/peripheral and unicystic tumors have a better prognosis than solid/multicystic and desmoplastic tumors and could be suitable for conservative management consisting of simple enucleation. The WHO emphasizes the need for negative resection margins for solid/multicystic and desmoplastic tumors. Due to its retrospective design, this study comprise a case-mix of all macroscopic and histological forms of

ameloblastoma. The results of this study must therefore be interpreted cautiously.
The most appropriate treatment for young, growing patients remains controversial. In one study in a mean age of 12.3 years, they recommended conservative treatment because of an often less aggressive histological type (plexiform), in order to allow continued mandibular growth and consequently limit the major cosmetic and functional sequelae observed at this age. Secondary resection in the event of recurrence proved to be effective and allowed less mutilating treatment [17]. Surgical resection is considered to be the only effective treatment for ameloblastoma. In their review of the literature, Carlson et al. [18] reported the radioresistance and chemoresistance of ameloblastomas. Radiotherapy can also be responsible for second tumors, such as sarcomas [9]. However, some studies, such as that by Sauk et al. [10], have tried to develop targeted chemotherapy for sonic hedgehog (SHH)-dependent tumors in order to block signals responsible for systemic dissemination of certain ameloblastomas.

According to Dissanayake et al. [11], the recommended treatment for metastatic ameloblastoma associated with cervical lymph nodes comprises cervical lymph node dissection, while the addition of chemotherapy and radiotherapy protocols did not provide any conclusive responses in this study. Kurppa et al. [12] reported the presence of a BRAF V600E mutation in 63% of cases of ameloblastoma in their study. This mutation is responsible for resistance of ameloblastomas to anti-epidermal growth factor receptor (EGFR) targeted therapies. These studies could open up new therapeutic perspectives based on the use of targeted therapies in selected patients. The predominant site of metastasis of ameloblastoma is the lung [13], for which curative treatment consists of surgery. Slootweg et al. [14] also highlighted the possibility of lung metastases in patients with multiple recurrences. We have even observed a case of bilateral lung metastases in a patient with malignant transformation into ameloblastic carcinoma. This adult patient presented with follicular ameloblastoma of the maxilla treated by partial maxillectomy, with recurrence 4 years after initial management.

Lastly, some ameloblastoma can be fatal due to the anatomical site. Nastri et al., in a series of 13 ameloblastoma of the jaws printed in 1995 [15], reported 3 cases of local cerebral invasion resulting in the death of 2 patients. Posterior mandibular or maxillary tumor sites require larger resection margins than tumors of the mandibular symphysis due to the higher risk of local invasion and more difficult redo surgery. While radical treatment has been clearly established as first- line treatment for follicular and non-unicystic forms, the issue of resection margins has not been clearly resolved. Hong et al. [3] did not observe any statistically significant difference between resection with wide margins and segmental resection. In contrast, based on a review of the literature, Carlson et al. [8] recommended 1 to 1.5 cm resection margins. Nevertheless, this study is limited by its small sample size and the results must therefore be interpreted cautiously. This type of problem is frequently encountered in the case of rare diseases such as ameloblastoma.

Conclusion

It can be concluded that a higher recurrence rate in the group treated conservatively in the various forms of the histological and radiographic presentations, confirming the results of most of the published studies on this subject. However, conservative treatment still plays a major role in the management of certain forms of ameloblastoma. The macroscopic type, histological type, and the patient's age and medical history are major determinants.

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