ORIGINAL ARTICLE

CLINICAL OUTCOMES OF ENUCLEATION VERSUS MARSUPIALIZATION FOLLOWED BY ENUCLEATION IN THE MANAGEMENT OF UNICYSTIC AMELOBLASTOMA

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ABSTRACT

Objectives: To compare the clinical outcome of enucleation versus marsupialization followed by enucleation in managing unicystic ameloblastoma.

Material and Methods: A retrospective chart review of 40 patients equally divided into marsupialization and enucleation groups was carried out. Pre and Post-operative clinical features and radiographic bone resorption volume were evaluated using SPSS version 22. The independent sample T-test was used to compare the mean ages of the two groups. Categorical variables were subjected to the Chi-square test or Fisher's exact test. A logistic regression model included the variables with $p < 0.05$ to determine the odd ratios.

Results: Data analysis showed male dominancy in the enucleation group (12, 60%) and marsupialization groups (11, 55%). The mean age was 31.35±SD7.0507 and 29.80 ±SD9.1450, respectively, for group 1 and group 2. Post-operative pain, paresthesia, and swelling predominated in the enucleation group. A good percentage of reduction in bone resorption was noted in the marsupialization group. A significant difference was observed between the groups in terms of Postoperative paresthesia, Postoperative Swelling, and % reduction in Bone resorption (0.050, 0.004, and 0.001, respectively).

Conclusion: Conservative approach in the form of marsupialization showed less post-operative pain, paresthesia, and swelling as compared to the enucleation group. A good percentage of bone remodeling and less recurrence or progression of the disease was documented in the marsupialization group, resulting in a positive patient response to this strategy.

Keywords: Ameloblastoma, Enucleation, Marsupialization

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INTRODUCTION

Ameloblastoma is the second-most common odontogenic tumor, representing about 1% of all jaw tumors. The site predilection is such that they are much more common in the lower jaw than in the upper jaw and more common in the posterior mandible as compared to the anterior. Ameloblastomas are benign tumors with aggressive behavior and morbidity. Multicystic ameloblastoma is the most common type of these tumors, accounting for 86% of cases, causing considerably large swellings, disfigurement, teeth displacement, and even pathological fractures. [1, 2] UC ameloblastoma presents clinically and radiologically as a dentigerous cyst. Although their histological characteristic represents a typical epithelial lining composed of ameloblastoma epithelium. [3, 4]

Treatment of Unicystic ameloblastoma is debatable and surgeon-specific. The treatment modalities range from simple enucleation, enucleation, curettage, and Marsupialization to block resection with or without sacrificing the continuity of jaw bone. [5] Decompression or marsupialization was first described by Lean Scultet in 1671 as a means of changing the internal pressure of cystic lesions. With this type of treatment, the constant stimulus that causes peripheral bone reabsorption is reduced and nullified, favoring a decrease in the size of the lesion with subsequent bone healing. [6, 7]

In the management of unicystic ameloblastomas, sometimes, unnecessary aggressive treatment is offered to the patient, resulting in morbidity and loss of function in the jaw. Thus emphasizing the importance of providing a conservative treatment to the patient with this type of tumor to prevent the functional and esthetic sequelae entailed by the excision of the tumor through a conventional treatment. The present study is carried out to compare the clinical outcome of enucleation versus marsupialization followed by enucleation in the management of unicystic

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ameloblastoma.

MATERIAL AND METHODS

A retrospective chart review of patients treated for unicystic Ameloblastoma with Enucealtion and/ Or marsupialization was carried out from the departmental and individual records from January 2019 to December 2022 in the Department of Oral & Maxillofacial Surgery, Khyber College of Dentistry Peshawar after the ethical approval was obtained from the Research Review Board at Khyber College of Dentistry (06/ADR/KCD, dated: 17/01/2023). All patients fulfilling the inclusion criteria were divided into two groups (The enucleation group and the Marsupialization group). Demographic data such as age, gender, primary site, Pre and Post-operative clinical features such as Pain, swelling, paresthesia, and radiographic bone resorption volume (maximum anteroposterior length in cm multiplied by the maximum vertical distance in cm). [8] A reduction in the target resorbed area by 50% was considered a good result. When there was a reduction of less than 50% but more than 10%, it was denoted as a moderate result. Ineffective regression of the tumor was considered only when the reduction in the volume of the resorbed area was less than 10%. An increase in the size of the tumor at any stage was considered as Tumor progression.

Data were analyzed using the latest Statistical Package of Social Sciences (V-21). Continuous variables such as age were expressed as mean and SD. Frequency and percentages were determined for categorical variables such as gender, Pre and Post-operative pain, swelling, paresthesia, and percent reduction in bone resorption. To compare the mean ages of the enucleation and marsupialization group, an independent sample T-test was applied. Categorical variables were subjected to the Chi-square test or Fisher's exact test. The variables with $p < 0.05$ were included in a logistic regression model to determine the odd ratios.

RESULTS

Data analysis showed male dominancy in both enucleation group (12, 60%) and marsupialization groups (11, 55%). The mean age of all the patients of both groups was 31.35±SD7.0507 and 29.80 ±SD9.1450 respectively, for group 1 and group 2 with a mean age difference of 1.55, 95% CI -4.05 to 6.77, p-value= 0.552 (Table-2).

Analysis of other variables showed that pre-operative pain, paresthesia, and swelling most commonly occurred in the Marsupialization group and compared to the Enucleation group. While post-operative pain, paresthesia, and swelling predominated in the enucleation group. A good percentage of reduction in bone resorption was noted in the marsupialization group. Progression of the disease process was noted frequently in the enucleation group (10%) as compared to the Marsupialization group. A detail of these findings is given in Table 1.

Stratification of different variables with Enucleation

Table No 1: Descriptive statistics (n=20 each)

Variables		Enucleation Group		Marsupialization Group	
		(n)	(%)	(n)	(%)
Gender	Male	12	60%	11	55%
	Female	8	40%	9	45%
Mean Age		31.35±SD7.0507		29.80 ±SD9.1450	
Pre op Pain	No	12	60%	11	55%
	Yes	8	40%	9	45%
Post op Pain	No	6	30%	13	65%
	Yes	14	70%	7	35%
Pre Op Paresthesia	No	17	85%	12	60%
	Yes	3	15%	8	40%
Post Op Paresthesia	No	9	45%	15	75%
	Yes	11	55%	5	25%
Pre Op Swelling	No	9	45%	8	40%
	Yes	11	55%	12	60%
Post Op Swelling	No	5	25%	14	70%
	Yes	15	75%	6	30%
% Reduction in Bone resorption	Good	5	25%	12	60%
	Moderate	7	35%	5	25%
	Ineffective	6	30%	2	10%
	Progression	2	10%	1	5%

Table No 2: Mean age difference between the Enucleation Group and Marsupialization Group

Groups		Number of patients (n)	Mean age + S.D	Mean Difference	%95 Confidence Interval	P value
Age	Enucleation	20	31.35±SD7.0507	1.55	4.05- to 6.77	0.552*
	Marsupialization	20	29.80 ±SD9.1450			

*Significant at 0.05

Table No 3: Stratification of different variables with Enucleation and Marsupialization

Variables		Enucleation Group		Marsupialization Group		P-value
		(n)	(%)	(n)	(%)	
Gender	Male	12	(60%)	11	(55%)	0.749*
	Female	8	(40%)	9	(45%)	
Pre op Pain	No	12	(60%)	11	(55%)	0.749*
	Yes	8	(40%)	9	(45%)	
Post op Pain	No	6	(30%)	13	(65%)	0.27*
	Yes	14	(70%)	7	(35%)	
Pre Op Paresthesia	No	17	(85%)	12	(60%)	0.077*
	Yes	3	(15%)	8	(40%)	
Post Op Paresthesia	No	9	(45%)	15	(75%)	0.050*
	Yes	11	(55%)	5	(25%)	
Pre Op Swelling	No	9	(45%)	8	(40%)	0.749*
	Yes	11	(55%)	12	(60%)	
Post Op Swelling	No	5	(25%)	14	(70%)	0.004*
	Yes	15	(75%)	6	(30%)	
% Reduction in Bone resorption	Good	5	(25%)	12	(60%)	0.001**
	Moderate	7	(35%)	5	(25%)	
	Ineffective	6	(30%)	2	(10%)	
	Progression	2	(10%)	1	(5%)	

*Chi-square **Fisher Exact

Table No 4: Regression Analysis

Variables in the Equation		Odds Ratio	P- value	95% Confidence interval
Post op Swelling	No	Reference		
	Yes	.793	.805	.126 - 5.003
Postop Paresthesia	No	Reference		
	Yes	.186	.078	.029 - 1.209
% Reduction in Bone resorption	Good	Reference		
	Moderate	.050	.049	.003 - .985
	Ineffective	.148	.204	.008 - 2.811
	Progression	2.230	.590	.121 - 41.217

*Chi-square **Fisher Exact

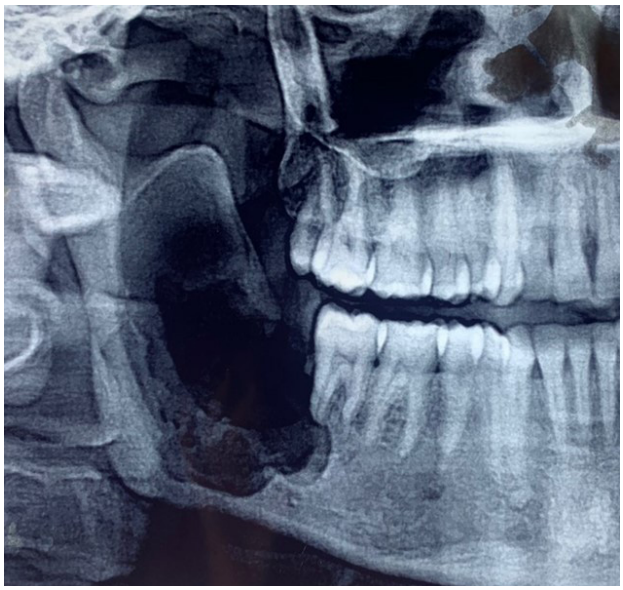

Fig-1 Pre-operative

Fig-1 Post-operative

Fig-2 Pre-operative

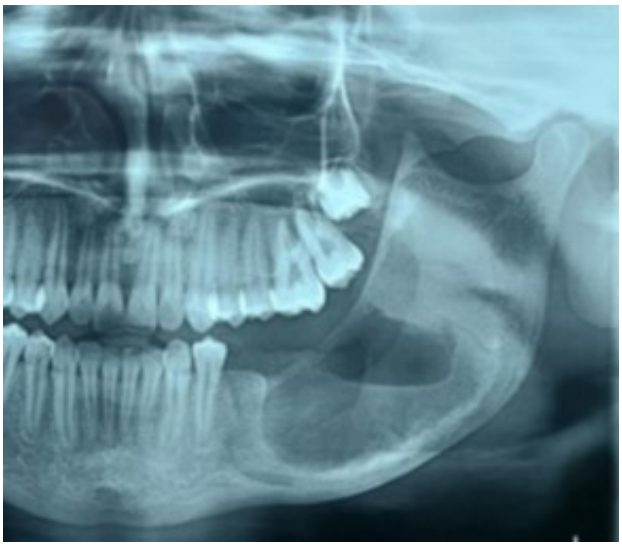

Fig-2 Post-operative

and Marsupialization was performed and showed that a statistically significant difference was observed between enucleation and marsupialization groups in terms of Post-operative paresthesia, Postoperative Swelling, and % reduction in Bone resorption (0.050, 0.004 and 0.001 respectively). See Table 3 for details.

Logistic regression analysis showed that the odds ratio (OR) for Post-operative swelling was 0.793 (95 % CI .126 - 5.003) and post-operative Paresthesia was 0.186, 95% CI .029 -1.209. Similarly, the OR for Moderate reduction in % bone resorption was 0.050 (95 % CI .003 - .985), ineffective reduction in % bone resorption 0.148 (95 % CI .008 - 2.811) and Progressive resorption 2.230 (95 % CI. 0.121 – 41.217). In other words, greater odds of Post-operative swelling, Post-operative paresthesia, moderate % reduction in bone resorption, ineffective bone formation, and progression of resorption were associated with patients who received enucleation as the treatment modality as compared to the marsupialization Group. Detail is given in Table 4

DISCUSSION

In the present study, the majority of the cases were reported in males (57.5%) as opposed to their female counterparts, with a female-to-male ratio of 1: 1.29. Figueiredo et al. [9], Reichart et al. [11,] and Cosola et al.[12,] during analysis of their results, have given the same results. Age

analysis revealed that ameloblastomas typically occur in the late second and early third decades of life. Most patients with ameloblastoma present at 3rd to 5th decade with an average age at the time of diagnosis that varies from continent to continent, estimated to be approximately 42.3 and 30.4 years in Europe and Africa. [13, 14] The pediatric population accounts for a very small percentage of instances of ameloblastoma, which varies by geographic location. [15]

When compared to the Enucleation group in our study, the Marsupialization group had pre-operative pain, paresthesia, and edema more frequently. In contrast, edema, paresthesia, and post-operative discomfort were frequent in the enucleation group. A good percentage of reduction in bone resorption was noted in the marsupialization group. Progression of the disease process was noted frequently in the enucleation group (10%) as compared to the Marsupialization group. Pre-operative pain was present in 40% of cases of the enucleation group and 45% in the marsupialization group. In contrast, Al-Khateeb et al.[16], White, and Pharoah [10] found a majority of their cases as painless swelling and showed that pain is rarely a presenting complaint in ameloblastoma. Juliansyah et al. [17] showed that 39.1% patient of ameloblastoma presented with Swelling at the initial presentation, while 21.7% had pain. The variations in the previous results could be attributed to variations in geography, sample size, and investigative methodology. The current study's findings are consistent with those of the Juliansyah et al. investigation. In a retrospective study by Dandriyal et al., [18] of 20 patients reasoned that 10 (half) patients went through enucleation with bone curettage. They assessed the general impact of enucleation with bone curettage as good in 10% of cases, moderately effective in 30% of cases, and insufficient in 20% of cases. In our review, 60% of the cases had good quality bone formation when treated with marsupialization, while 30-35% of cases presented ineffective bone remodeling after enucleation. The difference may be attributed to the fact that the majority of their cases were treated with enucleation and curettage, while in our study, we compared the difference between the two surgical modalities and analyzed their outcomes. Another reason for this difference in findings may be because we included only unicystic ameloblastomas while they investigated different histopathological types.

Wu et al. [8]. studied 233 subjects with ameloblastomas of the jaw treated with marsupialization or enucleation to evaluate clinical outcomes, recurrences, and post-operative sequelae. Forty-three cases of unicystic ameloblastomas received marsupialization modality, with a 50% success rate. With curettage, the cystic cavity of 24 patients was reduced by more than 50%. Their statistics revealed that the decreased pace of the cystic cavity in the decompression bunch was higher than that in the curettage group ($P<0.001$). Conversely, the results of the present investigation show that the percent reduction of bone in the cystic cavity in the marsupialization group is better than the enucleation group ($P<0.001$). Genetic variation and differences in sample size may be the reason for this gross difference between the results of these two studies. [19]

Tumor recurrence is the most common complication of any odontogenic and non-odontogenic tumors of the jaws, depending upon multiple factors, including histopathological type, surgical modality employed, and the extent of involvement of bone and soft tissues. Investigators from different countries showed that the conservative approach in the treatment of ameloblastomas has a greater chance of recurrence or progression of the tumor. [20]

The results of our study showed an overall low progression or recurrence rate in both groups. However, the marsupialization group proved itself to be superior in that the recurrence or progression rate was higher in the enucleation group (10%).

The only limitation here was that only a short follow-up of 12 months was carried out for each case, and this might have affected the overall recurrence rate as compared to other large-scale cross-sectional studies. As per the methodical survey of Lau SL et al. [21], different recurrence percentages were demonstrated by different modalities, with a lowest of 3.6% for resection and as high as 30.5 for enucleation. Nakamura N et al.[22,] while working on unicystic ameloblastomas, have the highest recurrence rates for marsupialization and conservative enucleations. In their study, almost no recurrence rate was demonstrated by radical excision and marsupialization.

The current study contains several shortcomings. This research was restricted to one public sector hospital. To examine the actual occurrence and behavior of the uncommon histological subtypes, a comparatively limited number of patients were chosen from the oral and maxillofacial surgical facility. Additionally, because there are so few of these tumors, we did not examine the impact of the

unicystic ameloblastoma histological pattern on treatment outcomes. An information bias about both clinical and morphological data may be present in the study. The minimum follow-up period was 12 months, and recurrences might emerge and show up at a later time. Nonetheless, the study's data is extremely thorough and comparable to other research reports. Additionally, it offers fresh data that may help improve our knowledge of tumor epidemiology and guide the creation of effective treatment plans in this region of the globe.

CONCLUSION

It can be concluded that the Conservative approach in the form of marsupialization followed later on by enucleation showed less post-operative pain, paresthesia, and swelling as compared to the enucleation group. A good percentage of bone remodeling and less recurrence or progression of the disease was documented in the marsupialization group, resulting in a positive patient response to this strategy.

The retrospective results of our study confirm the importance of developing less invasive approaches to ameloblastoma elimination while maintaining patients' quality of life.

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Authors Contribution:

Following authors have made substantial contributions to the manuscript as under

Authors	Conceived & designed the analysis	Collected the data	Contributed data or analysis tools	Performed the analysis	Wrote the paper	Other contribution
Rehman B	✓	×	✓	×	✓	×
Khan M	✓	✓	×	✓	✓	×
Murad B	×	✓	×	×	✓	×
Ahmad T	✓	✓	✓	×	✓	✓
Rahman AU	✓	✓	×	✓	✓	×

Authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Ethical Approval:

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