TREATMENT OF ODONTOGENIC AMELOBLASTOMAS & THEIR LONG TERM FOLLOW UP AT TERTIARY CENTRE

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ABSTRACT

Fifty-two patients of ameloblastoma were operated with special emphasis on radiographic and histological appearance. The unicystic radiographico-histological (38) cases were managed conservatively with marsupialization followed by enucleation (Group A' 15 Patients) and enucleation with peripheral ostectomy (Group B' 23 Patients). The radiographico-histological multicystic (solid) variety (Group C' 14 Patients) was treated aggressively by resection. In conservative treatment regimens Carnoy's solution was applied after enucleation of the tumour whereas, the patients of aggressive surgery were operated with minimum 5mm safety marginal clearance of the tumour. The recurrence rate with average four years follow up was 0.0% for resection, 13.33% for marsupialization followed by enucleation and 8.69% for enucleation with peripheral ostectomy. The results were encouraging for unicystic ameloblastoma treated patients (Group A' & B'), in best interest of jaw bone contour preservation.

INTRODUCTION

Ameloblastoma is a benign, slow growing, locally aggressive tumor, originating from reduced enamel epithelial of developing tooth. It is the second most common odontogenic tumor after Odontomes and accounts for 11% of prevalence with no specific gender predilection.[1] The tumor is more observed in mandible comparative to maxilla with molar ramus commonly involved area.[2] The ameloblastoma clinico-radiographically is classified as unicystic, solid intraosseous (multicystic) and less common peripheral ameloblastoma. Histopathologically it has six types namely acanthomatous, granular cell, desmoplastic, basal cell, follicular and plexiform whereas, the last two types are more common, though the treatment options and recurrence rate are irrespective to its histological variants.[3] The initial diagnosis of the tumour is made on the history, clinical examination and specific radiographic images but the diagnosis is confirmed with histopathological evaluation of the tumour.[4]

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Management of ameloblastoma is primarily complete removal of tumor. The treatment modalities are controversial worldwide and reasons are distinct aggressive biological behavior with high rate of reoccurrence of this tumour.[5] The aggressive management included segmental or en bloc resection for ameloblastoma with 1 cm to 1.5 cm marginal clearance, clinically and radiographically.[6] The conservative management includes marsupialization, enucleation with curettage and excision with peripheral ostectomy. Recurrence rate for both treatment modalities varies and ranges from 75-90% in conservative and 15-25% in radical treatment.[7]

Unicystic ameloblastoma was first described in 1977 by Robinson & Martinez. It is a distinct clinicopathological entity because of general unicystic radiographic appearance, associated with an unerupted tooth, typical histologic findings, common in the mandible of younger patients and low recurrence rate after conservative surgical treatment than that of its other counterparts.[8] Vickers & Gorlin in 1970 described three distinct histopathological features for unicystic ameloblastoma and these were slightly modified by Leider et al in 1985 into three mural, intramural and luminar types on histopathological, prognostic and therapeutic implications.[9] Various treatment modalities for unicystic ameloblastoma have been used, such as segmental or marginal resection, conservative treatment including enucleation and curettage, and marsupialization

followed by second stage surgery. The reported recurrence rate after treatment of unicystic ameloblastoma ranges from 10 to 25% and there is no adequate evidence to prove which treatment modality is the most effective and its reason for the practical variability.[10] The solid or multicystic type ameloblastoma is also clinically common odontogenic tumor. It is locally aggressive and has a significant impact of morbidity and mortality. The tumor is reported to be treated with a segmental or en bloc resection of the mandible. The recurrence rate after segmental resection is 13-15% of solid ameloblastoma.[11] Peripheral ameloblastoma is a rare epithelial odontogenic tumor, limited to the soft tissues of the gingiva or oral mucosa. This extraosseous odontogenic soft tissue tumor was first reported in the literature by Kuru in 1911 but Stanley and Krogh reported in 1959 the first well-established and true case of peripheral ameloblastoma and it accounts for approximately 2% to 10% of all ameloblastomas. Later on the peripheral ameloblastoma has been reported frequently in literatures.[12,13] The ameloblastoma is always considered to be benign, but occasionally it may be locally aggressive or with malignant potential.[14]

In this study, the patients were grouped as A' B' C' and were treated conservatively by marsupialization followed by enucleation, enucleation with peripheral ostectomy and aggressive management with segmental or en bloc resection, respectively. The objective of the study is to evaluate the rate of recurrence of the tumour with and without aggressive treatment in best interest of human jaw bone contour preservation in long term follow up.

METHODOLOGY

Fifty-two patients of biopsy confirmed ameloblastoma were treated in Oral & Maxillofacial Surgery Unit of de,Montmorency College of Dentistry/Punjab Dental Hospital, Lahore during 2004 to 2012. After written informed consent, the demographic data was collected from each patient, and clinical and radiographic features were carefully documented. All the patients were grouped as A' B' C' and were treated conservatively by marsupialization followed by enucleation, enucleation with peripheral ostectomy and aggressive management with segmental or en bloc resection respectively, on basis of clinical and radiographic findings.

Orthopantomograph (OPG), Postoanterior (PA) mandible, paranasal sinuses (PNS) views and in few cases CT scan was obtained to assess the site, size and extent of the tumor. Group A (15 patients) of unicystic ameloblastomas were treated with marsupialization followed by enucleation. The marsupialization was selected mainly for young patients with clinical and radiographic unicystic ameloblastoma. The average time of marsupialization was one year before enucleation. The patients included had large tumour involving ramous usually, irrespective of the perforation of the cortical plates. Group B' (23 patients) of also unicystic ameloblastomas were treated with enucleation with peripheral ostectomy. The patients selected were clinical and radiographical free of basal bone perforation. The remnants of the tumour were carefully removed and peripheral ostectomy was performed by bone shaving with large round bur to 5 mm depth or till the clinical healthy bone appeared. The Inferior Dental neurovascular bundle was identified where in close proximity of tumour and was retracted carefully during enucleation and ostectomy. Additional bone trimming was done where tumor nests were encountered in the area. All the unicystic ameloblastoma patients were applied Carnoy's solution (60% absolute alcohol, 30% chloroform, 10% G.I.A.C. acids & FeCl2 /1 gm) after enucleation of the tumour with ribbon gauze for three minutes and then toileted the area.

Group C' (14 patients) of multicystic (solid) ameloblastomas were treated aggressively with segmental or en bloc resection. Due to reported high recurrence rate after resection (13 to 15%), the solid/ multicystic tumours were removed with radiographic marginal clearance of 0.5 cm to 1.5 cm to ensure all micro cysts removal. The reconstruction titanium plate of 2.7 mm was adapted in required shape and stabilized with self threading screws on both cut ends. In patients with hemimandibulectomy, the stabilization was done only from one side.

RESULTS

Fifty two patients were treated in total in three categories for surgery with definitive male (44) and mandibular jaw (49) predilection. The presenting age group was mainly 11 to 40 years (47 patients). The conservatively treated (Groups A' and B') were managed with marsupialization followed by enucleation and enucleation followed by peripheral ostectomy, respectively. Carnoy's Solution was applied as a standard after removal of the tumour in conservative varieties. Marsupialization followed by enucleation (Group A') had relatively young patients with 13.33% (02 patients) recurrence rate comparative to Group B' (08.69 %). All these patients were followed up more than three years postoperatively. Fourteen patients were aggressively managed (Groups C') with segmental or en bloc resection. The jaw contour was initially restored with reconstruction plate for at least two years postoperatively before comprehensive bone grafting. The recurrence rate was 0.0% in this group. Special care was given to cortical perforation (06) patients, mainly of Group C' (04) with follow-up not less than 5 years whereas, 96% (50) patients had minimum follow-up period not less than two years. In this study only one patient each was found under age 10 years or above 50 years. This adult case was treated aggressively as it had large tumour with perforation. The unicystic histological classification of ameloblastoma was not implicated in treating Group A' & B' and the overall results were encouraging. One patient treated for enucleation followed by peripheral ostectomy (Group B') had recurrence after three years and was managed aggressively by segmental resection with 1cm marginal clearance. In

Group A', the marsupialization was followed by initial whitehead varnish packing for three weeks followed by acrylic stunt formation and continuous cleaning using catheter with normal saline in 50cc syringe. The cleaning period before enucleation varied from 08 months to 02 years, depending upon the size of the tumour. One patient had recurrence with the history of unicystic ameloblastoma (bone perforation) at the time of diagnosis.

Three patients developed postoperative dehiscence of the sutures, out of that two patients were of Group B' (unicystic ameloblastoma) and only other was from Group C' (solid ameloblastoma), whereas primary closure was performed in Group B' & C' all patients. One patient in Group C' developed partial plate exposure and infection after dehiscence. The debridement was done at the site and drain was inserted with favorable prognosis. Group A' had relapse of the marsupialized site in two patients and second surgery was performed to reassure the patency at marsupialized site.

TABLE 1: AGE GROUPS OF PATIENTS WITH MALE TO FEMALE RATIO

S. No.	Age group (years)	No. of patients	Male	Female
1.	1-10	01	01	00
2.	11-20	08	07	01
3.	21-30	26	22	04
4.	31-40	13	10	03
5.	41-50	03	03	00
6.	51-60	01	01	00

TABLE 2: TYPES OF AMELOBLASTOMA PATIENTS WITH THEIR TREATMENT GROUPS

Patients No.	Tumour Type	Treatment groups	Recurrence average 4 yrs
15	Unicystic	A'	02 (13.33 %)
23	Unicystic	B'	02 (08.69 %)
14	Multicystic	C'	00 (0.00 %)

TABLE 3: TREATMENT GROUPS OF PATIENTS AND THEIR FOLLOW UP PERIOD

No. of patients	Follow-up (years)	Group A'	Group B'	Group C'
06	07 above	02	03	01
09	06	03	04	02
19	05	04	10	05
08	04	03	02	03
06	03	02	03	01
03	02	01	01	01
01	Less than 02	00	00	01

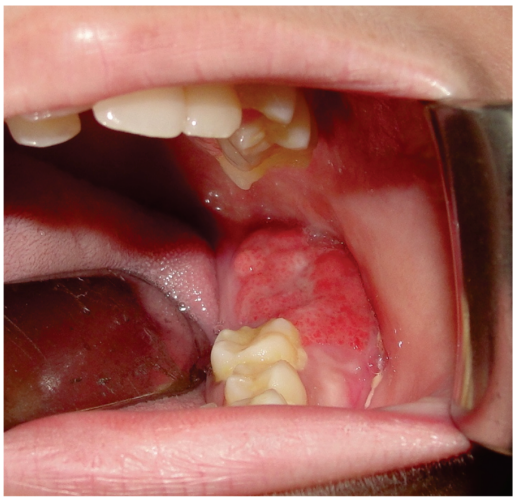

Fig 1: Left Mandibular Unicystic Ameloblastoma (Intra Oral)

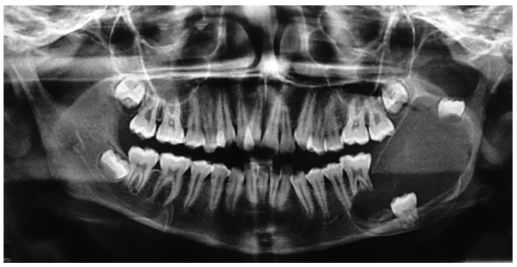

Fig. 2: Left Mandibular Unicystic Ameloblastoma (Orthopantomograph)

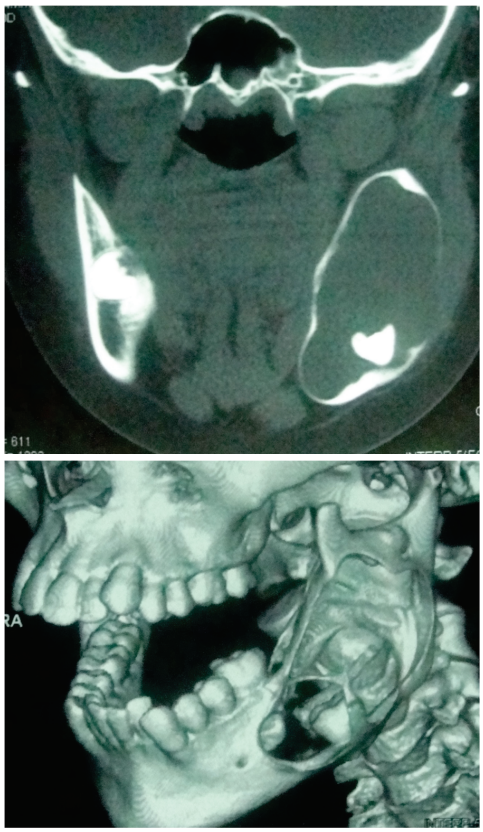

Fig 3: Left Mandibular Unicystic Ameloblastoma (Coronal CT Scan & 3D)

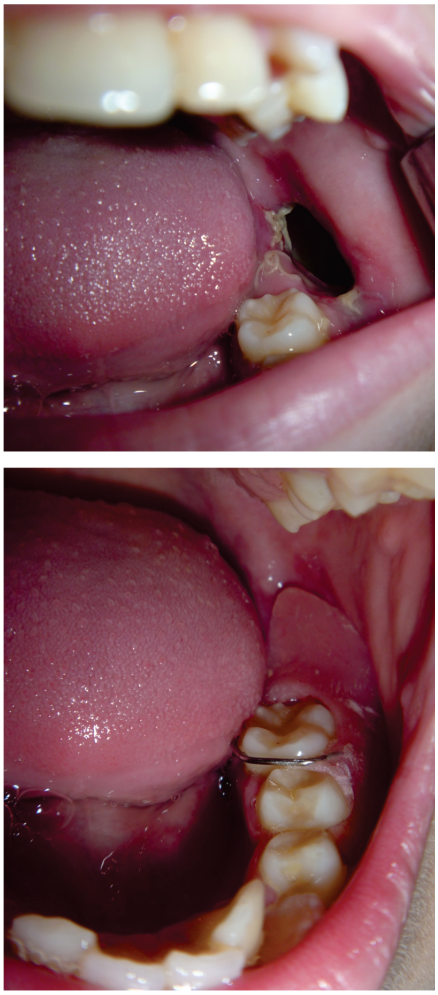

Fig. 4: Left Mandibular Unicystic Ameloblastoma (Marsupialization & Acrylic Stunt)

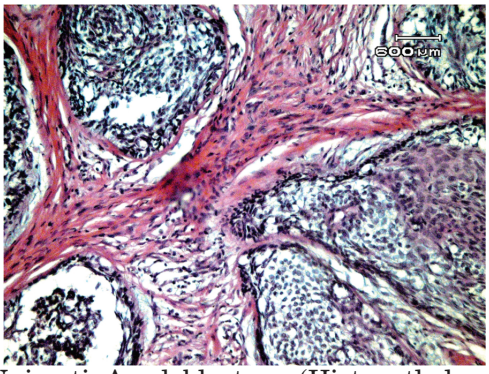

Fig 5: Unicystic Ameloblastoma (Histopathology 5 x 5x)

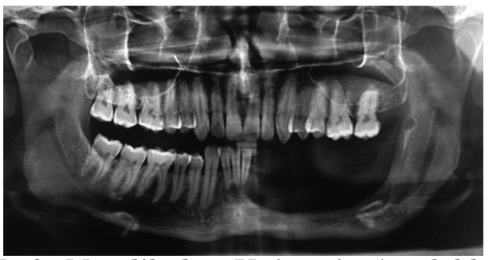

Fig 6: Left Mandibular Unicystic Ameloblastoma (Postoperative Orthopantomograph)

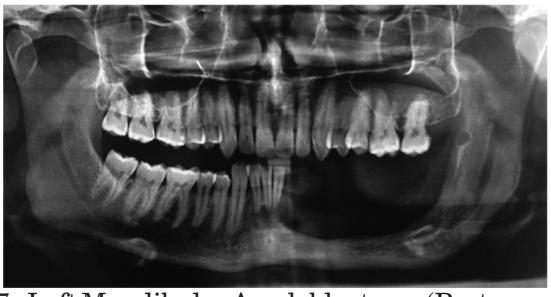

Fig 7: Left Mandibular Ameloblastoma (Postoperative Peripheral Ostectomy OPG)

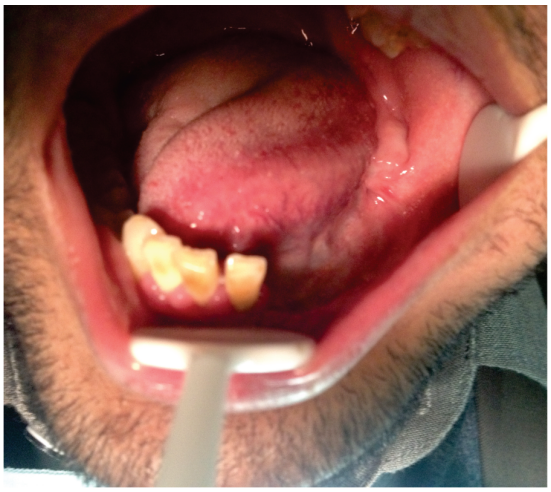

Fig 8: Left Mandibular Ameloblastoma (Postoperative Intraoral view)

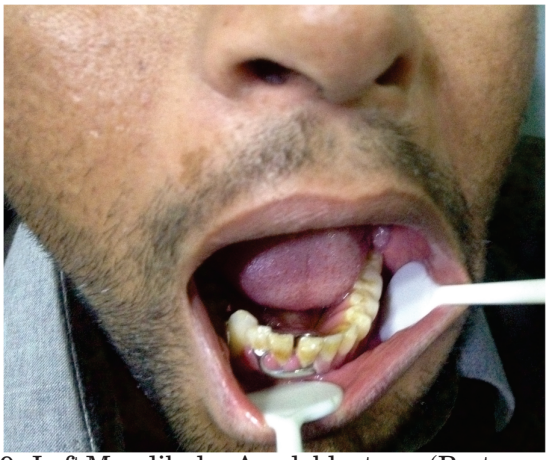

Fig 9: Left Mandibular Ameloblastoma (Postoperative Intraoral view with denture)

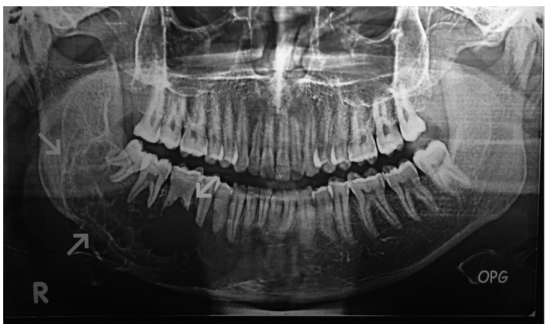

Fig. 10: Right Mandibular Solid Ameloblastoma (Orthopantomograph)

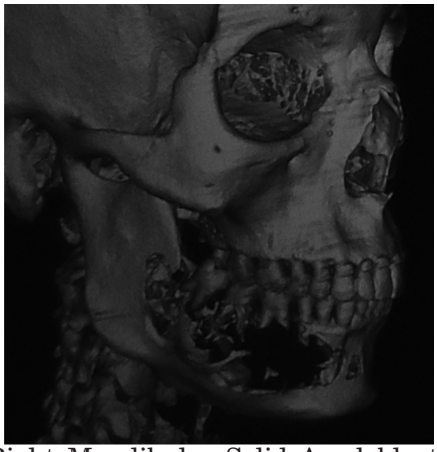

Fig 11: Right Mandibular Solid Ameloblastoma (3D Scan)

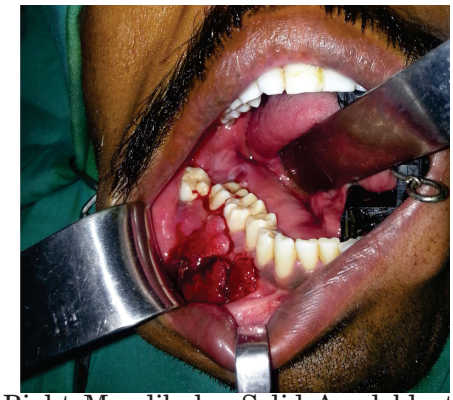

Fig 12: Right Mandibular Solid Ameloblastoma (Intraoral View)

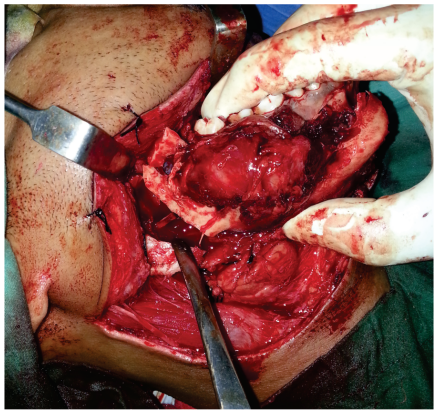

Fig 13: Right Mandibular Solid Ameloblastoma (Intraoral Operative View)

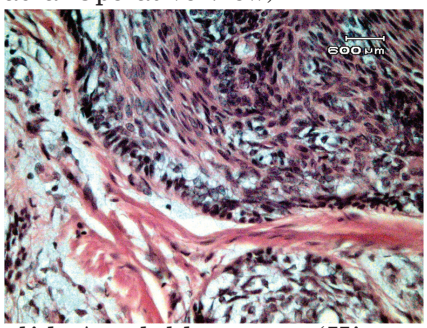

Fig 14: Solid Ameloblastoma (Histopathology 10 x 10_x)

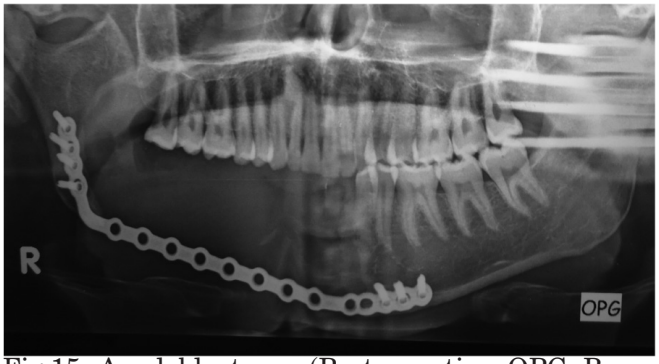

Fig 15: Ameloblastoma (Postoperative OPG Recon Plate after Resection)

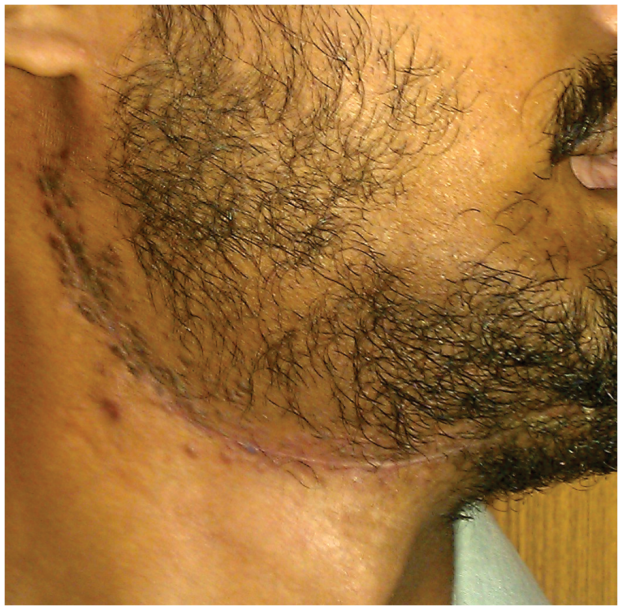

Fig 16: Right Mandibular Solid Ameloblastoma (Late Postoperative View)

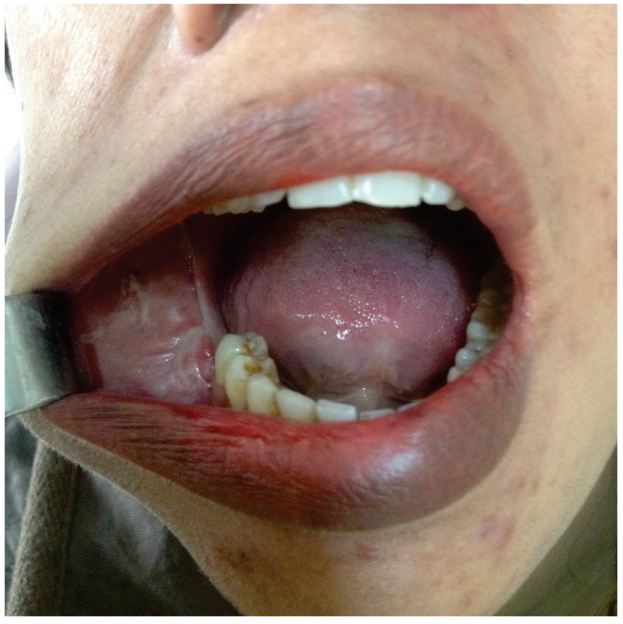

Fig 17: Right Mandibular Ameloblastoma (Late Postoperative Recurrence)

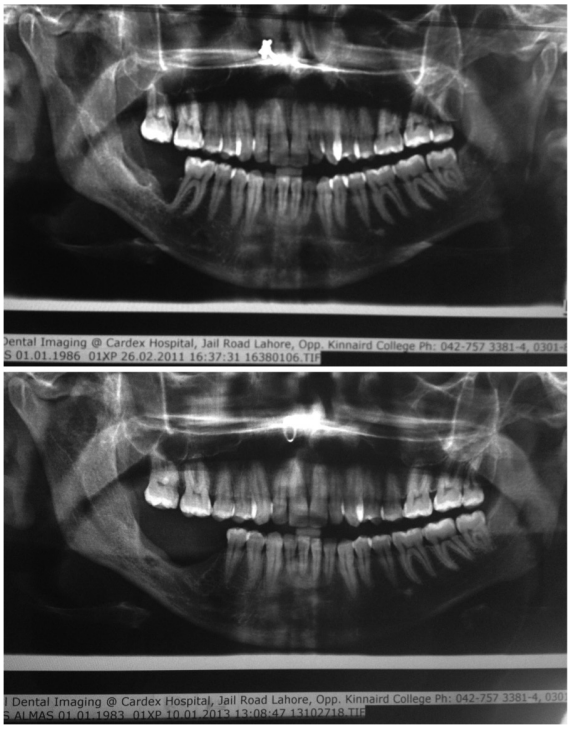

Fig 18: Right Mandibular Ameloblastoma (Preoperative and Primary Postoperative Follow up)

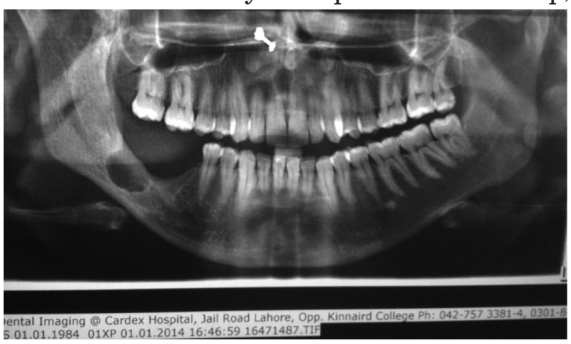

Fig 19: Recurrence Right Mandibular Ameloblastoma (Late Postoperative Follow up)

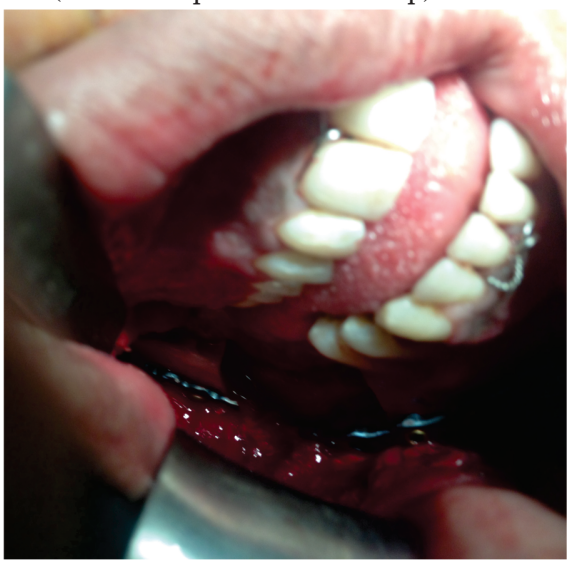

Fig. 20: Recurrence Right Mandibular Ameloblastoma (Second Aggressive Surgery)

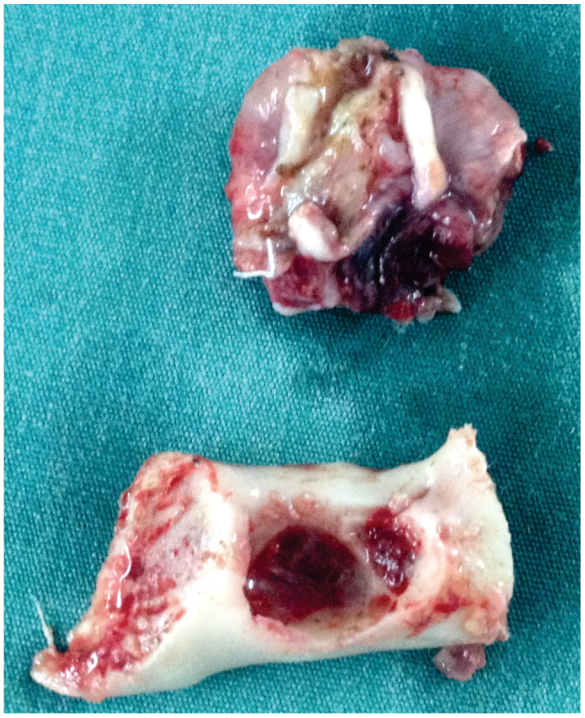

Fig 21: Recurrence Right Mandibular Ameloblastoma (Second Surgery resection)

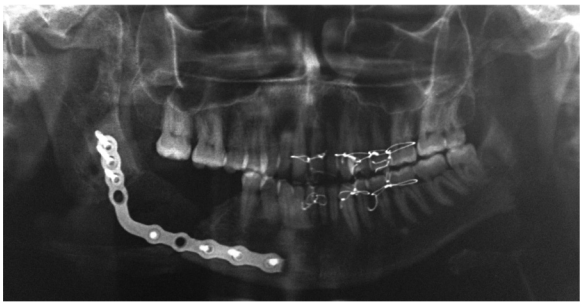

Fig 22: (Postoperative OPG) Recurrence Right Mandibular Ameloblastoma

DISCUSSION

The management of ameloblastoma is surgical and remains controversial worldwide.[15] One school of thought advocates major segmental or en bloc resection for ameloblastoma, based on a requirement of 1 to 1.5 cm of clinically or radiographically normal bone to ensure uninvolved margins.[16] The other view in the literature clearly shows management of ameloblastoma by curettage (conservative) only.[17] The selection and success of surgical options depend on the careful patient's evaluation, accurate history, radiographs & special imaging (CT), good pre-surgery histopathological reporting. Although the radiographic evaluation remains the single most important tool in planning surgery but oral surgeon's interest & experience may be important factor in determining the extent of surgical intervention.[18] The aggressive surgery in multicystic (solid) ameloblastoma with minimum 10mm radiographic or clinical marginal clearance to ensure all micro cysts removal, showed extreme promising results[19] whereas

Robinson & Martinez showed 13% to 15% recurrence rate in their studies and recommended follow-up was essential as most recurrences presented within the first 5 years after surgery.[20] Similar results had been also reported by others[21,22] whereas, in this study recurrence rate was not observed in Group C' even after 5 years of average follow up. We feel that careful segmental or en bloc resection with safe margins could be responsible for these favorable results.

In unicystic ameloblastoma after the removal of lesion, the cavity was painted with Carnoy's solution for three minutes before it was toileted, and long-term clinical and radiological follow-up appreciated the surgical results. Although similar results were not advocated in other studies[23,24] as in our (recurrence rate 08.69%) whereas, the study had strict postoperative vigilance for minimum three years in all unicystic cases. Though the surgical excision with peripheral ostectomy is not preferred treatment for every ameloblastoma but it had saved a bulk of the bone at the site of tumour and neurovascular bundle also whereas, the peripheral 2 to 3mm bone was trimmed after the removal of tumour.[25] In our study, all young patients (less than 30 years) were presented with unicystic type of ameloblastoma, were treated in Group A' & B' and aim was to save the maximum architecture of the jaw bone in these young patients.

The marsupialization followed by enucleation had not been routine treatment option and was safely and vigilantly used in this study. The recurrence rate in this case was 13.33% whereas all selected patients were younger age in this group. Very little literature is available regarding this treatment option and mainly case reports are only available with relatively high recurrence rate.[26,27]

This study has been carried out at a tertiary care centre by using three different techniques to treat two varieties of locally aggressive odontogenic ameloblastomas. The long term follow-up was highly encouraging towards its conservative management in unicystic variety comparative to solid type, where segmental or enbloc resection with clear safety margins may lead to loss of jaw bone and disability resulting in psychosomatic trauma.

REFERENCES

1 Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: biological profiles of 3677 cases. Oral Oncol Eur J Cancer 1995; 31: 86-99.

2 Butt FMA, Guthua SW, Awange DA & et al. The pattern and occurrence of ameloblastoma in adolescents treated at the university teaching hospital, in Kenya: 3 year study. J. Cranio Max Fac Surg 2012; 40: 39-45.

3 Mendenhall WM, Werning JW, Fernandes R & et al. Ameloblastoma. Am J Clin Oncol 2007; 30: 645-648.

4 Dunfee BL, Sakai O, Pistey R, Gohel A. Radiologic and pathologic characteristics of benign and malignant lesions of the mandible. Radiographics. 2006; 26: 1751-68.

5 Gardner DG. Controversies in Oral and Maxillofacial Surgery. W.B. Saunders; 1994: 301-314.

6 Chapelle KAOM, Stoelinga PJW, de' Wilde PCM & et al. Rational approach to diagnosis and treatment of ameloblastomas and odontogenic keratocysts. Br J Oral Max fac Surg 2004; 42: 381-390.

7 Bataineh A. Effect of preservation of the inferior and posterior borders on recurrence of ameloblastomas of the mandible. Oral Surg Oral Med Oral Path Oral Rad End 2000; 90: 155.

8 Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: a clinicopathological study of 57 cases. J Oral Path 1988; 17: 541-546.

9 Leider AS, Eversole LR, Barkin ME. Cystic ameloblastoma: a clinicopathological analysis. Oral Surg Oral Med Oral Path Oral Rad End 1985; 60: 624-630.

10 Rosenstein T, Pogrel MA, Smith RA, Regezi JA. Cystic ameloblastoma: behaviour and treatment of 21 cases. J Oral Max fac Surg 2001; 59: 1311-1316.

11 Nakamura N, Higuchi Y, Mitsuyasu T & et al. Comparison of long-term results between different approaches to ameloblastoma. Oral Surg 2002; 93: 13.

12 Philipsen PH, Reichart PA, Nikai H & et al. Peripheral ameloblastoma: biological profile based on 160 cases from the literature. Oral Oncol 2001; 37 (1): 17-27.

13 Ide F, Mishima K, Miyazaki Y & et al. Peripheral ameloblastoma in-situ: an evidential fact of surface epithelium origin. Oral Surg Oral Med Oral Path Oral Rad End 2009; 108 (5): 763-767.

14 Gilijamse M, Leemans CR, Winters HAH & et al. Metastasizing ameloblastoma. Int. J Oral Max Fac Surg 2007; 36: 462-464.

15 Williams TP. Management of ameloblastoma: A changing perspective. J Oral Max Fac Surg 1993; 51: 1064.

16 Thompson LDR. Head and neck pathology. "Foundations in diagnostic pathology series". Philadelphia: Elsevier; 2006.

17 Olaitan AA, Adekeye EO. Unicystic ameloblastoma of the mandible: Long terms follow up. J Oral Max Fac Surg 1997; 55: 345-348.

18 Gardner DG. Some current concepts on the pathology of ameloblastomas. Oral Surg Oral Med Oral Path Oral Rad End 1996: 82 (6): 660-69.

19 Hammarfjord O, Roslund J, Abrahamsson P, et al. Surgical treatment of recurring ameloblastoma: are there options? Br. J. Oral Max Fac. Surg. 2013; 4095: 1-5.

20 Robinson L, Martinez MG. Unicystic ameloblastoma: a prognostic distinct entity. Oral Cancer 1977; 40: 2278-2285.

21 Jones AV, Franklin CD. An analysis of oral and maxillofacial pathology found in adults over a 30-year period. J Oral Path Oral Med 2006; 35 (7): 392-401.

22 Hong J, Yun P-Y, Chung I-H, Myoung H & et al. Long-term follow up on recurrence of 305 ameloblastoma cases. Br J Oral Max Fac Surg 2007; 36: 283-288.

23 Lau SL, Saman N. Recurrence related to treatment modalities of unicystic ameloblastoma: a systemic review. Int J Oral Max Fac Surg 2006; 35: 681-690.

24 Li TJ, Wu YT, Yu SF, Yu GY. Unicystic ameloblastoma: a clinicopathological study of 33 Chinese patients. Am J Surg Path 2000; 24: 1385-1392.

25 Nagata T, Uchiyama Y, Miyazaki H & et al. Treatment of a large mandibular ameloblastoma by marsupialization and marginal resection preserves the mandibular inferior border. Asian J Oral Max Fac Surg. 2009; 21: 38-42.

26 Norifumi N, Yoshinori H, Hideo T, Masamichi O. Marsupialization of cystic ameloblastoma. J Oral Max Fac Surg 1995; 53 (7): 748-754.

27 Dolanmaz D, Etoz OA, Pampu A & et al. Marsupialization of unicystic ameloblastoma: a conservative approach for aggressive odontogenic tumors. Indian J Dent Rad 2011; 22 (5): 709-712.