OSTEOLYTIC LESIONS OF THE MANDIBLE: A TEN-YEAR RETROSPECTIVE ANALYSIS

¹Brig S K Roy Chowdhury, ²Col Vivek Saxena, ³Lt Col Rajkumar K, ⁴Maj Prasun K Dubey ¹Professor & Head of the Department, ²Associate Professor, ³ Associate Professor ¹Oral & Maxillofacial Surgery Dept of Dental Surgery and Oral Health Sciences, ¹Armed Forces Medical College, Pune, India

Abstract: **Introduction:** Various osteolytic lesions affecting mandible have overlapping clinical and radiographic presentation making the diagnosis difficult. Osteolytic lesions of the mandible like odontogenic keratocyst and ameloblastoma have a plethora of treatment modalities with varying rates of success. Central giant cell granuloma is a relatively rarer tumour of the jaws but should be considered as a differential diagnosis in younger age groups. **Materials and Methods:** Retrospective analysis of 50 cases of osteolytic lesions treated surgically over a period of past 10 years with minimum 6 months follow-up was carried out using operative and follow-up records. 38 patients were identified as cases of odontogenic keratocyst, 10 of ameloblastoma and 2 of central giant cell granuloma. Patients were categorised as per lesions and treatment modality used, and outcome was recorded **Results**: Employment of adjunctive therapies like application of Carnoy's solution and peripheral ostectomy showed lesser recurrence in cases of odontogenic keratocyst. More aggressive surgery, with inclusion of 1-2 cm of healthy bone margin along with excision of tumour produces satisfactory results in ameloblastoma. Additional therapy using calcitonin along with surgery in central giant cell granuloma shows progressive healing and lesser recurrence. **Conclusion:** Osteolytic lesions pose a confusion during their diagnosis and many lesoins have very high chances of recurrence. A more radical treatment modality should be adopted in these cases after carefully assessing the individual need, prognosis, post-operative morbidity and functional loss.

IndexTerms – Osteolytic lesions, Odontogenic keratocyst, Ameloblastoma, Central giant cell granuloma

I. INTRODUCTION

Osteolytic lesions of the mandible can be of odontogenic as well as non-odontogenic origin and have a destructive potential which causes bone resorption of varying degrees. Lesions of odontogenic origin are broadly classified as cysts and tumours. Odontogenic tumours represent a group of lesions with a wide spectrum of clinical, radiological and histopathologic presentation. Tumours can be either benign or malignant. Most commonly occurring benign tumours of mandible are different types of ameloblastoma and odontomas. Other tumours like ossifying fibromas and granulomas are encountered less frequently. Malignant tumours involving the mandible include squamous cell carcinomas, osteosarcomas, and metastatic tumours. In addition to these, certain hamartomatous lesions such as haemangiomas and arteriovenous malformations can also involve the mandible. Osteolytic lesions often pose a difficulty in their diagnoses as they have a wide range of histopathologic characteristics but similar and confusing clinical and imaging appearances.

II. PATIENTS AND METHODS

A retrospective review was performed on 50 cases of osteolytic lesions of mandible. Patients who were treated surgically in the Department of Oral & Maxillofacial Surgery, Armed Forces Medical College, Pune between the years 2007 and 2017 were undertaken in order to evaluate the clinicopathologic and radiographical basis for management of osteolytic lesions of the mandible and their outcome. Patients with chief complaints related to parts of maxillofacial skeleton other than mandible were excluded from our study. Also excluded were patients without (or with incomplete) diagnostic (radiological and histopathological) and/or treatment records, patients with recurrent cysts or tumours with previous history of surgery and those with follow-up of less than six months.

The treatment and diagnostic records of these 50 patients were reviewed and the available information was documented. Demographic data, viz., gender and age at the time of presentation were recorded. The patients were then categorised based on the diagnosis using the diagnostic guidelines and classification of cysts and tumours as per WHO. ^[1] The treatment carried out for each lesion was also recorded after reviewing the operative notes.

Treatment performed as per the lesion were also recorded. This included, simple enucleation with the use of curettes, marsupialisation, marginal resection, segmental resection and hemi-mandibulectomy. Adjuvant treatment in combination with the above procedures like use of Carnoy's solution, Calcitonin spray, and peripheral ostectomy, if used, were documented. A note was made if any reconstruction was carried out after resection. If yes, then the reconstruction material/modality used, was alo documented.

III. RESULTS

Out of the fifty patients meeting the inclusion criteria, 38 patients (76%) were diagnosed with Odontogenic Keratocyst (OKC), 10 cases (20%) were diagnosed with Ameloblastoma and 2 patients (4%) were diagnosed with Central Giant Cell Granuloma (CGCG). [Figures 1 & 2]

Out of 38 cases of OKC, 23 were male and 15 were female with an age range of 18-72 years. Mean age in our study for OKC was 39 years, which is nearly similar to the findings of Brannon et al. ^[2] who found the mean age of occurrence as 38 years. [Table 1]. Most commonly involved region was the molar-angle region with a total of 24 cases (63.1%). 10 patients were treated by enucleation alone (26.3%), 9 with enucleation followed by application of Carnoy's solution (23.6%) and 8 with enucleation followed by peripheral ostectomy (21.0%). 7 patients were treated by marsupialisation (18.4%). Resection of the mandible, without continuity defect, was carried out in 4 patients (10.5%). [Table 2] Recurrence rate was nil with resection and use of peripheral ostectomy and lower in enucleation followed by application of Carnoy's solution (Recurrence rate: 11.1%) as compared to enucleation alone (Recurrence rate: 30.0%). [Figure 3]

Amongst the 10 cases diagnosed with ameloblastoma, all ten were males with an age range of 22 to 75 years. Mean age at the time of presentation was nearly 39 years which is in concurrence with the findings of Sampson et al. ^[3] The lesion in its largest diameter varied from 5 to 11.5 cm. 1 patient was identified with tumour of anterior mandibular region whereas remaining patients had tumours of body-ramus-angle region. In four of the patients with ameloblastoma of the posterior mandible, the lesion crossed the mandibular midline. All patients were treated with the resection of mandible with inclusion of 1-1.5 cm of unaffected bone margin. Complete disarticulation was carried out in 2 patients (20%). High profile reconstruction plate was used in all the patients (100%).

Two children, both male, aged 12 and 15 years were diagnosed with central giant cell granuloma (CGCG) in the symphysis region. One individual was treated surgically by marginal resection of the mandible, without continuity defect. Another individual was treated by enucleation of the lesion followed by regular application of whitehead varnish pack for 02 months. Both patients were given Calcitonin nasal spray, 100 IU BID for 12 months, using a can which delivers metered dose of 100 IU per spray. An acrylic removable partial denture was used for functional rehabilitation.

IV. DISCUSSION

Development of teeth and associated structures is a complex process involving different types of tissues. Odontogenic cysts and tumours originate as a result of deviation from the normal pattern of odontogenesis. ^[4] Classification of odontogenic cysts is approved as per 2017 WHO guidelines. ^[5]

Often, patients with cystic lesions do not have presenting problems even in advanced forms of disease. As per Morgan et al., ^[6] significant number of patients (42.5%) with cystic lesions are diagnosed incidentally during routine radiography or during radiographical examinations for unrelated reasons. In symptomatic cases, clinical presentation and thorough history of the disease is important in establishing a diagnosis.

Mandibular lesions with different pathologic features may have similar and overlapping appearances on the radiographs. Hence, clinical parameters like patient age at manifestation, prevalence of the disease in local population, site of the lesion within the mandible, appearance, border contour, and effects of the lesion on adjacent structures and clinician's familiarity with embryologic characteristics are crucial in diagnostic considerations. ^[7] Even with all this data, it is sometimes impossible to provide a diagnosis without a biopsy.

Initial imaging can be carried out in the form of Orthopantomogram (OPG). However, Computed tomography imaging is considered to be the gold standard for diagnosis of such lesions. Cone Beam and regular fan beam Computed tomography are superior to OPG in terms of facilitating accurate measurement of the extent of the lesion in the bone and the adjoining structures. 3-dimensional volume rendering further helps the surgeon to plan out the operative procedure. [Figure 4] With the advent of 3-dimensional printing, stereolithographic models [Figure 5] are extremely advantageous in planning the osteotomy/resection. Digital surgeries can be carried out and, the unaffected bone can be mirrored onto the affected side so as to predict the most probable anatomy of a healthy mandible for the individual. Such digitally corrected models further help in precise adaptation of reconstructive plates [Figure 6], hence saving the operative time. This modality is regularly utilised in this institution for extensive lesions of mandible.

Radiographic imaging cannot be solely relied for diagnosis of osteolytic lesions. Imaging doesn't always provide a specific diagnosis, but it helps to narrow down the possible differential diagnosis hence helping to formulate treatment guidelines.^[8] Before proceeding for surgical treatment, it is important to establish a microscopic diagnosis by histopathological examination. Needle-aspiration biopsy is a quick and simple technique in order to differentiate the type of lesion, whether cystic or solid with further confirmation by incisional/punch biopsy.

Ever since OKC was first described by Philipsen in 1956, management of OKC is a subject of constant debate. A plethora of treatment modalities are available, all aiming to minimise the recurrence which ranges from 2.5% to 62.5% in various studies. ^[9] Meiselmann ^[10] concluded that enucleation or curettage with or without a "peripheral ostectomy" was adequate provided that care is taken to carefully excise the entire specimen. The lining of OKC, which is extremely thin and friable, poses a significant challenge to the attempts of removal of the cyst, in toto. The recurrence rate of OKC is found to be lower after chemical cauterisation by

Carnoy's solution is used as an adjunct procedure after removal of the cyst. ^{[11] [12]} Use of the same is also advocated by Blanas et. al ^[13]

Akin to OKC, ameloblastoma is also a controversial lesion when it comes to its management and has been discussed extensively in numerous literature. Conservative management of ameloblastoma by enucleation and curettage has been known to have a recurrence rate as high as 55 - 95 % ^[14] ^[15] ^[16] Once the tumour has recurred, the management becomes more difficult and challenging. Once detected, a radical approach should be selected for ameloblastoma during the first surgery itself. In our institution, ameloblastoma is treated with resection of the tumour along with healthy bony margin up to 1.5 - 2 cm in multi cystic and intraosseous lesions and up to 1 - 1.5 cm in uni-cystic ameloblastoma as advocated by Carlson et al. ^[17]

Central giant cell lesions, first described by Jaffe^[18] are relatively rare lesions and constitute about 7% of all the jaw tumours. Standard treatment for such lesions is enucleation/curettage or en bloc resection.^[19] With surgical treatment, a recurrence of 15 – 20% is seen.^[19] Pogrel conducted a study in which ten patients received calcitonin therapy for CGCG. He observed that in 8 cases, the lesions resolved completely after 19 to 21 months.^[20] Rationale of using calcitonin for CGCG lies in the fact that there are histological similarities between the giant cell lesion and the tumours of hyperparathyroidism. Calcitonin can be administered either as subcutaneous injections or in a lesser invasive form of nasal spray. Efficacy of nasal spray has been described in literature.^[21] Use of calcitonin nasal spray after surgical excision of the lesions in our study, showed satisfactory results and progressive healing.

V. CONCLUSION

Osteolytic lesions of the jaws are a great challenge to the surgeon as well as the radiologist. Due to rarity of certain lesions, they are not generally included in differential diagnoses which may further affect the treatment planning. Majority of lesions have notoriously high recurrence and are a reason of increased patient discomfort. Hence, it is imperative that the treatment should be meticulously planned and tailored after considering the behaviour of the lesion, its effects on the adjoining structures and the individual's general health. Of all the modalities, the treatment should be planned in such a way, so as to achieve complete removal of the lesion with minimum possibilities of recurrence, post-operative morbidity and functional loss.

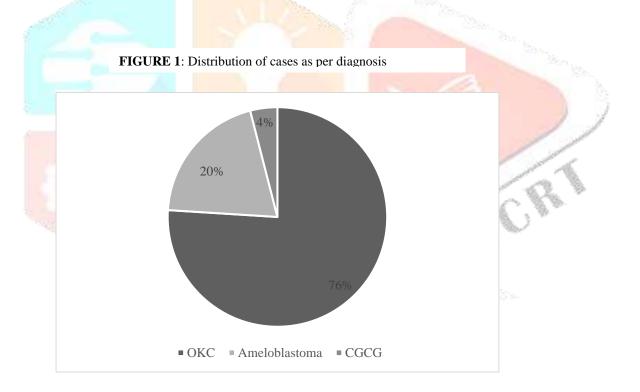
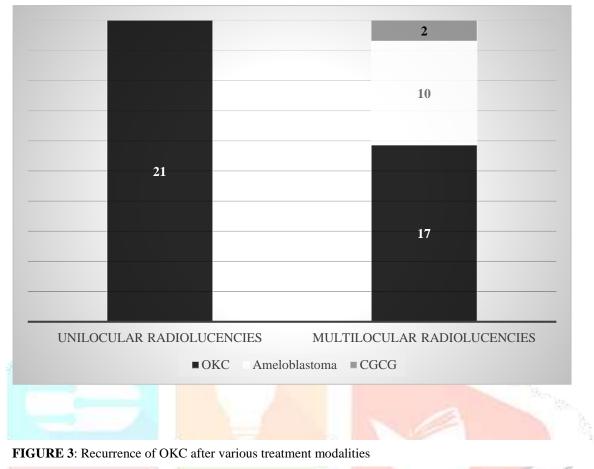


FIGURE 2: Radiographic presentation of osteolytic lesions



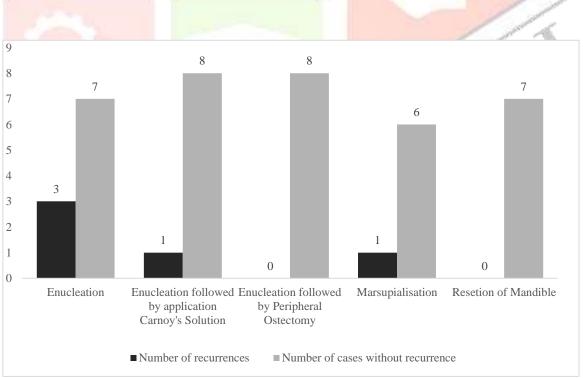




FIGURE 4: Use of CT with Volume rendering



FIGURE 5: Stereolithographic model of a patient with ameloblastoma

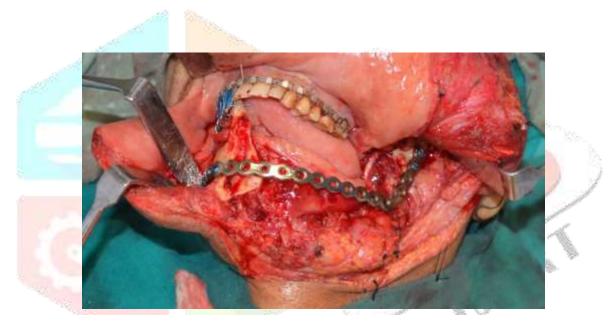


FIGURE 6: Fixation of recon plate that has been pre-adapted on stereolithographic model

	No. of patients
Gender	Total = 38
Male	23
Female	15
Age	18-72 years
range	

TABLE 1: Demographic distribution of OKC

Treatment	Number	of	
		patients	
Enucleation		10	
Enucleation followed	by	9	
application of Carnoy's Solution			
Enucleation followed	by	8	
peripheral ostectomy			
Marsupialisation	7		
Resection of mandible	4		

TABLE 2: Treatment modalities used in OKC

REFERENCES

Reichart PA, Philipsen HA. Odontogenic Tumours and Allied Lesions. London: Quintessence; 2004.
P.18.

2. Brannon RB. The odontogenic keratocyst: a clinicopathologic study of 312 cases. Part I: clinical features. Oral Surg Oral Med Oral Pathol 1976; 42:54–72.

3. Sampson DE, Pogrel MA. Management of mandibular ameloblastoma: The clinical basis for a treatment algorithm. Journal of Oral and Maxillofacial Surgery. 1999 Sep 1;57(9):1074–7.

4. Shafer, Hine, Levy Text book of Oral Pathology, 7th ed.

5. Speight PM, Takata T. New tumour entities in the 4th edition of the World Health Organization Classification of Head and Neck tumours: odontogenic and maxillofacial bone tumours. Virchows Arch. 2017 Jul 3

Morgan TA, Burton CC, Qian F. A Retrospective Review of Treatment of the Odontogenic Keratocyst.
Journal of Oral and Maxillofacial Surgery. 2005 May 1;63(5):635–9

7. Dunfee BL, Sakai O, Pistey R, Gohel A. Radiologic and pathologic characteristics of benign and malignant lesions of the mandible. Radiographics. 2006 Dec;26(6):1751–68.

8. Neyaz Z, Gadodia A, Gamanagatti S, Mukhopadhyay S. Radiographical approach to jaw lesions. Singapore Med J. 2008 Feb;49(2):165–176

Williams TP, Connor FA. Surgical management of the odontogenic keratocyst. Aggressive approach.
J Oral Maxillofac Surg 1994;52:964–966.

10. Meiselmann E Surgical management of the odontogenic keratocyst: conservative approach. J Oral Maxillofac Surg 1994;52:960-3.

Voorsmit RA, Stoelinga PJ, van Haelst UJ: The management of keratocysts. J Maxillofac Surg 9:228,
1981

12. Stoelinga PJW, Bronkhorst FB. The incidence, multiple presentation and recurrence of aggressive cysts of the jaws. J Craniomaxillofac Surg 1988;16:184-95

13. Blanas N, Freund B, Schwartz M, Furst IM. Systematic review of the treatment and prognosis of the odontogenic keratocyst. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2000 Nov;90(5):553–8.

14. Sehdev MK, Huvos AG, Strong EW, et al: Ameloblastoma of the maxilla and mandible. Cancer 33:324, 1974

15. Shatkin S, Hoffmeister FS: Ameloblastoma: A rational approach to therapy. Oral Surg 20:42 1, 1965

16. Waldron CA: Ameloblastoma in perspective. J Oral Surg 24:331,1966

17. Carlson ER, Marx RE. The Ameloblastoma: primary, curative surgical management, J Oral Maxillofac Surg. 64: 484, 2006.

18. Jaffe HL. Giant-cell reparative granuloma, traumatic bone cyst, and fibrous (fibro-osseous) dysplasia of the jawbones. Oral Surg Oral Med Oral Pathol 1953;6:159-75.

19. Pogrel AM. The diagnosis and management of giant cell lesions of the jaws. Ann Maxillofac Surg 2012;2:102-6.

20. Pogrel MA. Calcitonin therapy for central giant cell granuloma. J Oral Maxillofac Surg. 2003 Jun;61(6):649-653; discussion 53-54.

21. Allon DM, Anavi Y, Calderon S. Central giant cell lesion of the jaw: Nonsurgical treatment with calcitonin nasal spray. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2009; 107:811-8.