MANAGEMENT OF LARGE AMELOBLASTOMA: A CASE REPORT

Rudagi B M*, Halli R*, Mahindra U*, Asnani S*

Abstract

Ameloblastoma, first recognized by Cusack in 1827, is a neoplasm of odontogenic epithelium, especially of enamel organ-type tissue that has not undergone differentiation to the point of hard tissue formation. It accounts for about 1% of all oral tumors and about 9-11% of odontogenic tumors. Ameloblastoma in the mandible can progress to a great size and cause facial asymmetry, displacement of teeth, malocclusion, and pathologic fractures. A case of large ameloblastoma of mandible in a 39 year old male patient is being reported. Patient had large swelling on left lower side of the jaw since 2 years. Swelling was approximately 15 X 10cm in size. Intraorally the swelling involved buccal and lingual cortical plates and was obliterating the vestibular space. Patient did not seek medical attention because swelling was painless. The patient was investigated and treated with surgical excision and was provided with prosthesis to correct the post operative sequale. The patient showed uneventful recovery with no recurrence after a follow up of one year.

***Key Words**: Large ameloblastoma,Mandible,Reconstruction.*

Introduction

Ameloblastoma is a neoplasm of odontogenic epithelium, especially of enamel organ-type tissue that has not undergone differentiation to the point of hard tissue formation.[1] It accounts for about 1% of all oral tumors and about 9-11% of odontogenic tumors. It was first recognized in 1827 by Cusack.[2] It was designated as an adamantinoma in 1885 by the French physician Louis-Charles Malassez.[3] It was finally renamed ameloblastoma in 1930 by Ivey and Churchill.[4] It is generally a slow-growing but locally invasive tumour. Its peak incidence is in the 3rd to 4th decades of life and the male to female ratio is 1:1. Eighty percent of ameloblastomas occur in the mandible and majority are found in the angle and ramus regions. They are classified as unicystic, multicystic or solid. Eighty six percent of cases are multicystic ameloblastomas.

Ameloblastoma in the mandible can progress to great size and cause facial asymmetry, displacement of teeth, malocclusion, and pathologic fractures. Tumor size may range from 1 to 16 cm at presentation.

Case report

A 39 year old married male patient presented with the chief complaint of swelling of left lower side of jaw since 2 yrs. Patient gave history of exfoliation of two teeth in lower left posterior region of jaw one year back. Initially swelling was very small and hence he neglected it. Rapid growth to present size occured over last four months.

On extraoral examination, a large, diffuse swelling was seen on left side of face (15 X 10 cms). Superiorly it was extending upto lower margin of left orbit and zygomatic arch; inferiorly it was extending below inferior border of mandible and involving left submandibular and

***** Dept of Oral and Maxillofacial Surgery, RDC, Loni

Address for correspondence:
Dr Sanjay Asnani, Post Graduate, Department of Oral and Maxillofacial surgery, Rural dental college, Loni. (bk), Taluka: Rahata, dist. Ahmednagar.

submental region, anteriorly it was extending upto left corner of the mouth, posteriorly it was extending upto posterior border of ramus of mandible. Overlying skin was normal in appearence. It was soft to firm in consistency and tender on palpation (Fig 1).

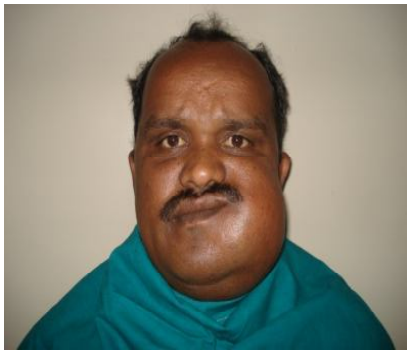

Fig 1:Preoperative photograph of the patient

On intra oral examination a large swelling was seen extending from 32 to 38 region. Expansion of buccal and lingual cortical plates and obliteration of left vestibular space was observed. Mucosa over the alveolar ridge was necrotic. Mucosa adjacent to alveolar ridge was erythematous, showing black necrotic patches in some areas. Mucosa over remaining part of swelling was pink in colour and 35,36, were missing. It was firm in consistency and tender on palpation. The 34, 37 and 38 had grade III mobility, 37 and 38 were displaced superiorly near buccal mucosa, 33 had grade II mobility. A provisional diagnosis of benign tumour of mandible was made.

Investigations

Orthopantamogram(OPG), left lateral oblique view of mandible, PA mandible, mandibular occlusal view, CT Scan of mandible were obtained.

Orthopantamogram (Fig 2) revealed a large mutilocular radiolucency (soap bubble appearence) extending anteriorly upto 33, posteriorly upto a region of 2 cm. in front of left angle of mandible, superiorly extending into the part of ramus, just above third molar region; inferiorly below the inferior border of mandible. External root resorption and superior displacement of 34,37,38 was seen and 16,17,28,45 were carious.

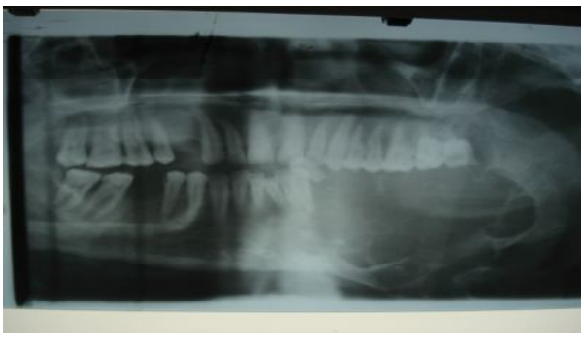

Fig 2:OPG showing multilocular radiolucency on left side of mandible

CT Scan (Fig 3) showed a large 8.5 x 7.5 x 5.1cm, multilocular, well circumscribed, expansile, heterogenous solid lesion which was devoid of cystic component in left body of mandible. It was mildly enhancing on post contrast study. Few non enhancing areas suggestive of necrosis were seen .

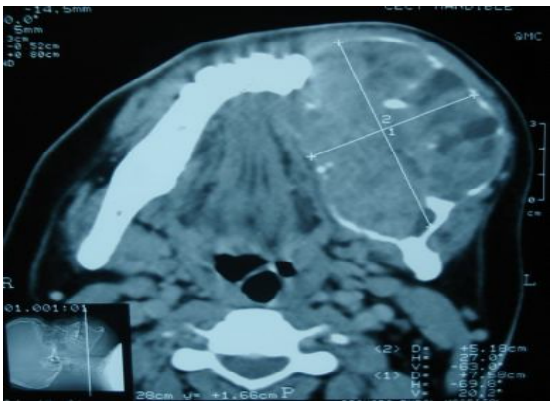

Figure 3:Section of CT Scan of mandible showing the extent of the lesion

Diagnosis

A differential diagnosis for multilocular lesion was considered which included ameloblastoma, odontogenic myxoma and central giant cell

granuloma. Histopathology of incisional biopsy showed para keratinised stratified squamous epithelium with proliferation, suggestive of oral epithelium. The connective tissue was fibrocellular and loosely arranged in certain areas. The tumour was composed of odontogenic epithelium with massive proliferation forming large follicles and plexes. The follicles and plexes were lined by tall columnar ameloblasts like cells containing stellate reticulum cells with areas of cystic change. The supporting stroma was made up of fibrocellular connective tissue with abundance of blood vessels and few inflammatory cells. Overall features were suggestive of ameloblastoma undergoing cystic changes (Fig4).

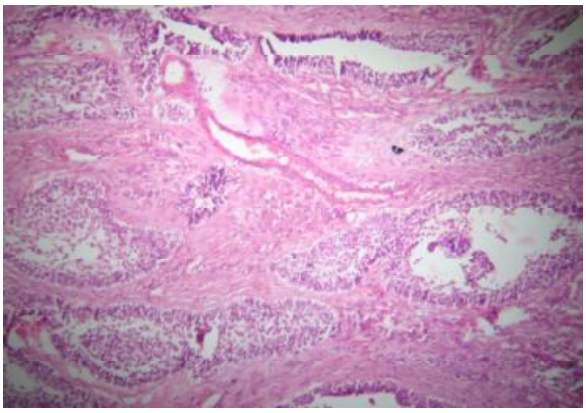

Fig 4:Histopathological slide showing ameloblastoma undergoing cyst changes

Management

Under general anaesthesia excision of the tumour mass was done via lip split incision. Segmental resection of the mandible was carried out from distal of left central incisor upto sigmoid notch, maintaining the condyle in the glenoid fossa. The tumour was resected en bloc and reconstruction was done immediately with the help of reconstruction plate.The mass was sent for histopathological examination and the surgical site was sutured. Ryles tube was inserted for feeding for 2 weeks. Patient was discharged and was kept on follow up. On follow up, it was found that the mandible deviated to the opposite side due to muscle pull and scar tissue, causing altered occlusal relationship. Physiotherapy was adviced and a mandibular guiding splint with a maxillary ramp was constructed to guide the mandible to as close as centric position as possible, thus reducing the facial disfigurement. Patient was kept on follow up for a period of one year and no signs of recurrence was observed (Fig 5).

Fig 5:Post operative photograph of the patient after one year

Discussion

Cases of ameloblastoma that advanced to the size of an infant's head have been reported from time to time, but in recent years, after the introduction of panoramic radiographs in routine dental practice it is rare to find a large ameloblastoma of the mandible.[5,6] Many cases of large ameloblastomas of the mandible associated with hypoprotenemia have also been reported. Hypoprotenemia results from leakage of fluid from the lesion and also due to anaemia. Large ameloblastomas can cause facial disfigurement, loss of occlusal function and difficulty in ingestion of food. It can also cause haemorrhage due to ulcerations. As the diet is affected in such cases the condition of the patient can further deteriorate and life may be endangered by pulmonary edema.[7] In our case there was loss of occlusal function. The tumour

protruded into the oral cavity, with pathological migration of the teeth. There were no signs of malnutrition or hypoprotenemia in the patient since his intake of nutrition was not affected.

The malignant (metastasizing) ameloblastoma is exceedingly rare. The reported incidence is approximately 2%. Most metastasis(75%) involve the lung, pleura, or hilar lymph nodes. Less than 15% of the reported cases involve cervical lymph nodes. It also has been observed that the interval between the diagnosis of the tumour and appearance of metastasis is 9 years, with a survival time of 2 years after metastasis.[8] The underlying mechanism of metastasis is not clear. Aspiration, haematogenous or lymphatic spread are postulated as possible mechanisms. Malignancy in our reported case was ruled out by histopathological examination.

Large ameloblastomas are usually managed by single stage procedures and the same was done in our case. It reduces hospital stay with its attended morbidity.

Acknowledgements

We would like to acknowledge our thanks to Dr Yogesh Kini, Reader; Dr Viraj Kharkar, Senior lecturer, Department of Oral and Maxillofacial Surgery,Rural Dental College,Loni for their contribution during surgery. We appreciate the support given by Dr Jitendra Kalburge, Professor, Department of Oral Pathology, Rural Dental College, Loni, Dr Vikrant Kasat (MDS) Senior lecturer, Department of Oral Medicine and Diagnosis and Dr Parmeet Banga (PG Student), Department of Prosthodontics their help and support.

References

1. Woo S, Smith-Williams JE, Sciubba JJ, Lippers S: Peripheral Ameloblastoma of the Buccal Mucosa: Case Report and Review of the English literature. *Oral Surg, Oral Med Oral Pathol*, 1987, 67:78-84.
2. J.W. Cusack "Report of the amputations of the lower jaw". 1827, Dublin Hosp, Rec 4: 1-38.
3. L. Malassez. "Sur Le role des debris epitheliaux papdentaires". Arch Physiol Norm Pathol 1885; 5: 309-340, 6:379-449.
4. R.H. Ivey, H.R. Churchill, "The need of a standardized surgical and pathological classification of tumors and anomalies of dental origin," Am Assoc Dent Sch Trans 1930; **7**: 240-245.
5. Tokio Osaki, Kazuo Ryoke,Terumasa Nagami,Takatsugu Ogawa and Takeshi Hamada "Ameloblastoma with hypoprotinemia due to protein leakage" Int. Journal of Oral Surgery 1985:14:302-306.
6. H.Hata,M.Ebihara,T.Onitsuka,M Nakagawa,Y Kitagawa, Y.Ota: Large amelopblastoma of mandible with hypoprotenemia.Int. Journal of Oral and Maxillofacial Surgery. 2008;37: 866-869.
7. Shigeki Nakasato, Satoru Okamura,Keigo Kudo,Yasunori Takeda Journal of Oral and Maxillofacial Surgery 1991:49;764-767.
8. Laughlin EH: Metastasizing ameloblastoma. Cancer 64: 776,1989.
9. Glen Houston et al. Malignant (Metastatic) Ameloblastoma: Report of a case. J oral Maxillofac Surg. 51: 1152-1155, 1993.