ISSN: 0975-833X

Available online at http://www.journalcra.com

INTERNATIONAL JOURNAL OF CURRENT RESEARCH

International Journal of Current Research
Vol. 13, Issue, 03, pp.16497-16500, March, 2021

DOI: https://doi.org/10.24941/ijcr.40968.03.2021

RESEARCH ARTICLE OPEN ACCESS

AN AGGRESSIVE PLEXIFORM AMELOBASTOMA OF MANDIBLE

[1,*]Dr. Koyel Mukherjee, [2]Dr. Rudra Prasad Chatterjee, [3]Dr. Mousumi Pal, [4]Dr. Sk. Abdul Mahmud and [5]Dr. Mehebuba Sultana

[1]Post Graduate Student, Department Of Oral and Maxillofacial Pathology, Guru Nanak Institute Of Dental Sciences and Research, Kolkata, India
[2]Redear, Department Of Oral and Maxillofacial Pathology, Guru Nanak Institute Of Dental Sciences and Research, Kolkata, India
[3]Professor & HOD, Department Of Oral and Maxillofacial Pathology, Guru Nanak Institute Of Dental Sciences and Research, Kolkata, India
[4]Professor, Department Of Oral and Maxillofacial Pathology, Guru Nanak Institute Of Dental Sciences and Research, Kolkata, India
[5]Post Graduate Student, Department Of Oral and Maxillofacial Pathology, Guru Nanak Institute Of Dental Sciences and Research, Kolkata, India

ARTICLE INFO

Article History:
Received 11th December, 2020
Received in revised form
20th January, 2021
Accepted 14th February, 2021
Published online 17th March, 2021

Key Words:

Aggressive,
Ameloblastoma,
Plexiform.

ABSTRACT

Ameloblastoma is a locally aggressive, benign odontogenic neoplasm of ectodermal origin comprising of about 1% of the tumors and cysts of the jaws commonly appearing in the third to fifth decades of life. The most common site of occurrence is mandibular molar-ramus region. Despite being a benign entity, the tumor frequently shows local invasion with occasional metastasis. Amelobastoma in young persons are thought to be a rare entity which includes approximately 10-15% of all ameloblastomas. Plexiform amelobastoma with an unusual large size in a 20years old female is presented here, with its clinicopathological, radiological and histological features along with brief review of literature.

Citation: Dr. Koyel Mukherjee, Dr. Rudra Prasad Chatterjee, Dr. Mousumi Pal, Dr. Sk. A. and Dr. Mehebuba Sultana "An Aggressive plexiform amelobastoma of mandible". 2021 *International Journal of Current Research, 13, (03), 16497-16500.*

INTRODUCTION

WHO in 2017 classified ameloblastoma as a benign, yet locally aggressive tumor with a very high rate of relapse; which is comprises of 1% all oral tumors & 11% of odontogenic tumors (1). The pathogenesis of the tumor is from remnants of odontogenic epithelium, specifically rests of dental lamina or due to the neoplastic change of a non-neoplastic Odontogenic keratocyst.

Corresponding author:** ***Dr. Koyel Mukherjee,
Post Graduate Student, Department Of Oral and Maxillofacial Pathology, Guru Nanak Institute Of Dental Sciences and Research, Kolkata, India.

Most common site of occurrence is mandibular molar-ramus region (70%), followed by premolar region (20%) (2) and 10% are seen to be confined in the anterior tooth bearing region, without prominent sex predilection. This tumour commonly occurs in 3rd to 5th decades of life. Clinically, ameloblastoma appears as a slow growing asymptomatic tumor which might be associated with distortion, expansion and perforation of regional cortical plates. Conventional radiographs show mostly multilocular, seldom unilocular radiolucency either in honeycomb or soap-bubble appearance of varying size causing extensive destruction of the half of the mandible to only a small growth confined to the alveolar process. It is divided into three subtypes i.e. Solid Multicystic Ameloblastoma (SMA) comprising 86%, Unicystic Ameloblastyoma (7%) and Peripheral ameloblastoma (7%).

The characteristic histopathological features include nest or cord like arrangement of odontogenic epithelial cells, bounded peripherally by tall columnar cells having hyperchromatic nuclei situated away from the basement membrane mimicking the ameloblasts & the central portion being occupied by loosely arranged cells simulating the stellate reticulum. The intervening connective tissue stroma appears to be loose vascular with minimum cellularity Solid Multicystic Ameloblastoma histologically can be divided into Follicular, Plexiform, Acanthomatous, Basaloid, Granular cell, Papilliferous, Desmoplastic and Hemangioameloblastoma type. In plexiform ameloblastoma, there is presence of network of interconnected anastomosing strands or chords, bounded peripherally by columnar ameloblastomatous odontogenic epithelial cells and centrally placed stellate reticulum like cells. However, the central stellate like cells are scanty in this variant. Areas of cystic spaces are also noted. Ameloblastic carcinoma, by definition is a primary odontogenic malignant tumor having ogical characteristics of a benign ameloblastoma with cellular atypia comprising of only 2% of the cases.

CASE REPORT

A 20years female patient from semi-urban area reported to the Department of Oral & Maxillofacial Pathology, Guru Nanak Institute of Dental Science and Research, Panihati, Kolkata with a chief complaint of swelling on the right lower third of the face since last one year. Extra-oral examination revealed the presence of a large swelling involving the chin and cheek upto the posterior teeth of the right side[fig no.-1] of the mandible which was intitially small, but achieved a grotesque size within a period of 1year.

Fig no.1 Extra-oral photograph of the patient showing swelling over the right cheek

On palpation it was slightly tender, firm to hard in consistency, with fixation to the overlying skin. Regional lymphadenopathy, local rise of temperature, was also evident but parasthesia/anesthesia of the lower lip was absent.

Intra-orally, a large, diffuse, bosselated, expansile growth with multiple projecting nodules on the surface specially on mandibular anterior region extending from the right second premolar, crossing the midline upto the left canine region with obliteration of the buccal sulcus was evident [Fig no. 2]. Marked expansion, destruction and perforation of the buccal and lingual cortical plate and egg-shell crackling was also noted.

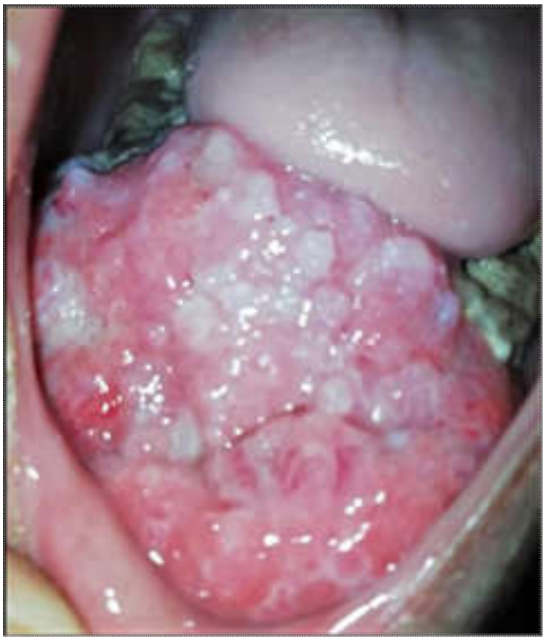

Fig. 2. Intra-oral photograph of the patient showing a huge bosselated lesion with surface nodularity involving the anterior part ofmandible

The CBCT images show a central lesion in the mandible, crossing the midline with ill-defined margins and ragged border[Fig no.-3] extending from right mandibular second pre-molar[Fig no. 5] to the left mandibular canine[Fig no.-4]. Destruction and perforation of the buccal cortical plate was evident. Aspiraion was performed which yielded a reddish yellow blood tinged fluid.[Fig no.-5]

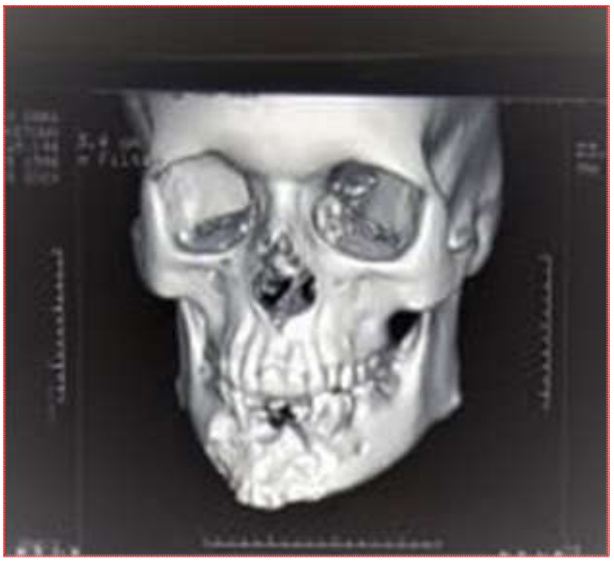

Fig no. 3 CBCT image showing gross destruction of the buccal cortical plate in the anterior mandible crossing the midline

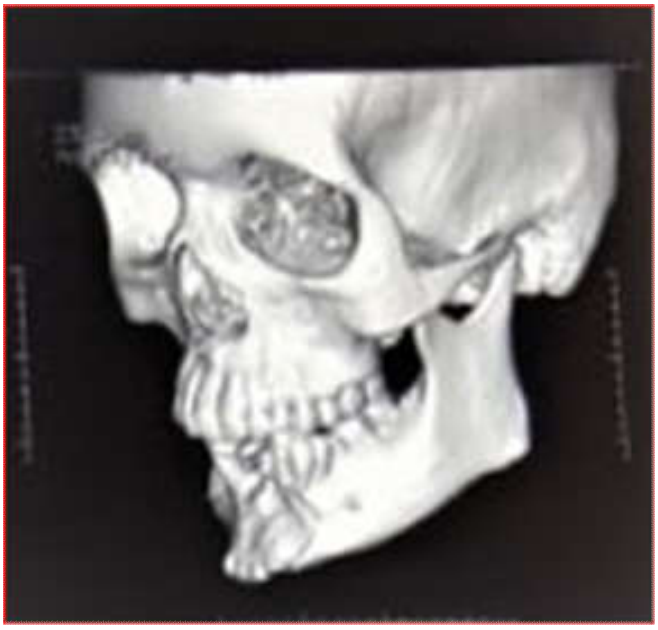

Fig no.4 CBCT image showing the extension of bone destruction upto left mandibular canine

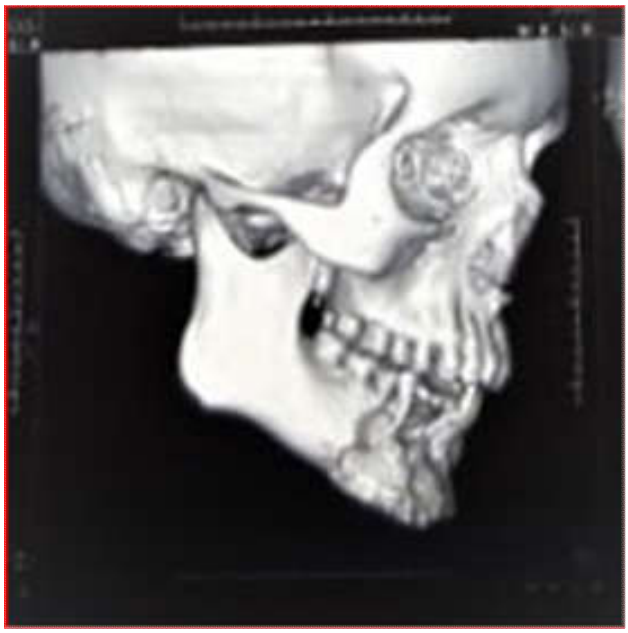

Fig no.5 CBCT image showing the extension of bone destruction beyond mandibular second premolar

The preoperative haematological examination revealed that the patient was severely anaemic at the time with a Hb% 7.6 gm/dl. Patient was admitted and was transfused with 2units of B+ve blood 2days prior the biopsy procedure. Histopathology revealed the presence of anastomosing chords of ameloblastomatous epithelium in a loose fibro-vascular connective tissue stroma.[Fig no.6]

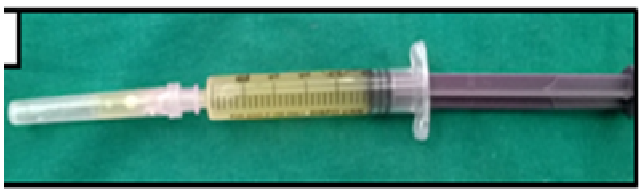

Fig no.6 Aspiration yielded a reddish yellow blood tinged fluid

Higher magnification shows the presence of peripheral tall columnar cells with reverse polarity along with scanty central stellate-reticulum like cells[Fig no.7]. Definite features of malignancy like loss of peripheral palisading, replacement of stellate cells with basaloid cells were not present. Not much cellular atypia or cellular pleomorphism was noted. A confirmatory histopathological diagnosis of Plexiform Ameloblastoma was established and the patient was reffred to the Oral Surgery Department for further treatment procedures.

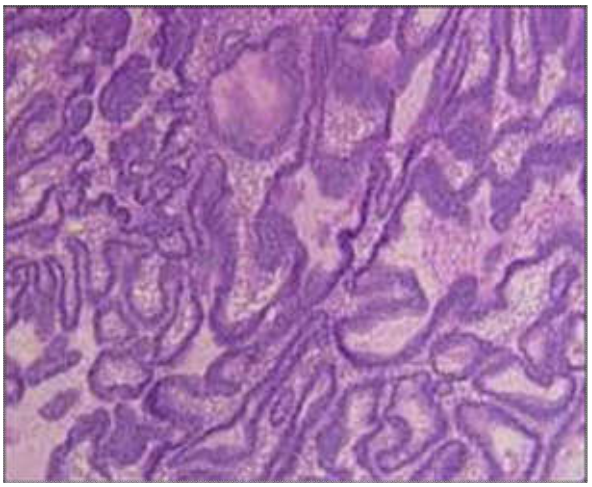

Fig.7. H&E stained section under low power(10X) view showing anastomosing strands and chords of odontogenic epithelial cells

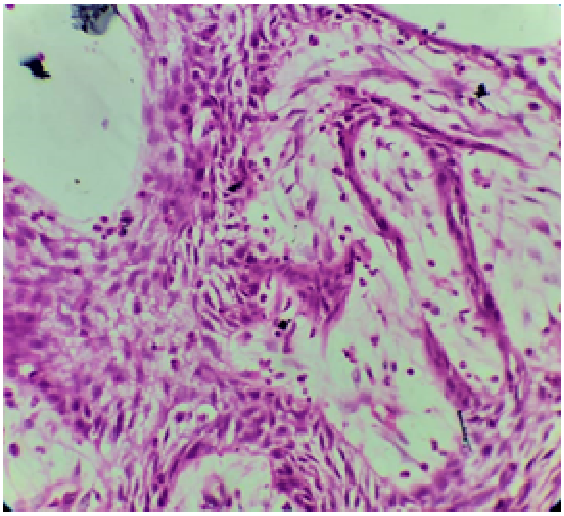

Fig . 8. H&E stained section under high power view(40X) showing peripheral ameloblast like cells like cells and central polygonal stellate like cells

DISCUSSION

The term ameloblastoma was coined by Churchill in 1879, the first detailed description of this lesion was given by Falkson in 1933 (3). Ameloblastoma is a benign epithelial odontogenic tumor but is often locally aggressive and destructive, with the capacity to attain great size, erode bone and invade adjacent normal structures (4). According to various authors stated that ameloblastoma arises from the epithelium of dental hard tissue during odontogenesis (5). This tumor commonly affect males in their 3rd to 6th decade of life with posterior mandible being the most frequent site of occurrence.

In mandible, 70% of the tumors are located in the area of the molars or the ascending ramus, 20% in the premolar region, and 10% in the anterior region(1). In maxilla too the lesion mostly occurs in the posterior region. About 10-15% of ameloblastomas are associated with a non-erupted tooth. This disease process is characterised by expansion, destruction and perforation of the cortical plates with displacement of the adjacent teeth. The clinico-radiological features were in accordance with the conventional ameloblastomas recorded in published literature. Histopathologically, plexiform ameloblastoma is characterised by proliferation of odontogenic epithelial cells as anastomosing strands or cords of varying length, within a collagenised connective tissue stroma (4). In higher magnification, the cells of the strands are tall columnar type consisting of hyperchromatic, round to ovoid elongated nuclei resembling pre-secretory ameloblast cells. Some polygonal stellate reticulum like cells are seen to be proliferating around the chords or strands. In general,almost $1/3^{rd}$ of the amelobastomas have plexiform pattern. These histopathological features were similar to the obsevations made by various researchers.

Distinctive features of the tumor were it's growth rate as it achieved a grotesque size within a period of 1year and it's occurrence in a young adult female patient both of which appears to be unusual. But, Definite features of malignancy like loss of peripheral palisading, replacement of stellate cells with basaloid cells were not present. Not much cellular atypia or cellular pleomorphism was noted. Unique biological behavior of ameloblastoma is a high incidence of relapse and possible malignant alteration. Various treatment modalities for Plexiform Ameloblastoma include enucleation followed by application of Carnoy's solution,(6) marsupialization followed by surgery, and segmental resection(7). Amelobastomas tend to extend through intact cancellous bone which causes the actual margin of the tumor to often extend beyond the actual clinical and radiographic margin. The recurrence rate after curattage alone is highest (50-90%) while resection results in the lowest recurrence rate. The radical resection of jaws, extending macroscopically 1.5 to 2 cm into the healthy tissue, is a surgical method of choice in the treatment of these ameloblastomas (8,4) which was performed in this case. Annual follow-up for atleast 10 years was recommended to the patient.

Conclusion

Ameloblastoma is a benign, locally aggressive odontogenic neoplasm frequently occurring in the mandibular molar ramus region which shows high rate of recurrence. Owing to their high prevalence and recurrence rate among odontogenic tumors, ameloblastomas should be should be handled carefully with early diagnosis and proper surgical intervention.

Acknowledgements

The authors gratefully acknowledge the contributions of Professor (Dr.) S. Kundu, Dr. Neha Shah, Dr.Sanjeet kr.Das, Dr. Swagata Gayen for theier valuable guidance in every step.

REFERENCES

1. Adebiyi KE, Ugboko VI, Omoniyi-Esan GO, Ndukwe KC, Oginni FO. 2006. Clinicopathological analysis of histological variants of ameloblastoma in a suburban Nigerian population. *Head & Face Medicine*., Dec;2(1):1-8.
2. Sehdev MK, Huvos AG, Strong EW, Gerold FP, Willis GW. 1974. Ameloblastoma of maxilla and mandible. Cancer. 33(2):324-33.
3. Iordanidis S, Makos C, Dimitrakopoulos J, Kariki H. 1999. Ameloblastoma of the maxilla. Case report. *Aust Dent J.,* 44:51-5.
4. Varkhede A, Tupkari JV, Mandale MS, Sardar M. Plexiform ameloblastoma of mandible: Case report.
5. Rajendran R. Shafer's textbook of oral pathology. Elsevier India; 2009, page no.- 271"
6. T. J. Li, R. M. Browne, and J. B. Matthews, 1995. "Epithelial cell proliferation in odontogenic keratocysts: a comparative immunocytochemical study of Ki67 in simple, recurrent and basal cell naevus syndrome (BCNS)-associated lesions," *Journal of Oral Pathology & Medicine*, vol. 24, no. 5, pp. 221–226.
7. Saku T, Shibata Y, Koyama Z, Cheng J, Okabe H, Yen Y. 1991. Lectin histochemistry of cystic jaw lesions: an aid for differential diagnosis between cystic ameloblastoma and odontogenic cysts. *Journal of oral pathology & medicine*. Mar;20(3):108-13.
8. Lau SL, Samman N. 2006. Recurrence related to treatment modalities of unicystic ameloblastoma: a systematic review. *International journal of oral and maxillofacial surgery*. 1;35(8):681-90.

****