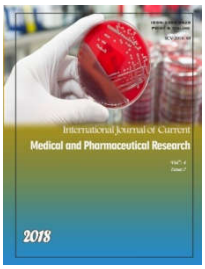
CASE REPORT

INTERNATIONAL JOURNAL OF CURRENT MEDICAL AND PHARMACEUTICAL RESEARCH

ISSN: 2395-6429, Impact Factor: 4.656
Available Online at www.journalcmpr.com
Volume 4; Issue 7(A); July 2018; Page No. 3434-3436
DOI: http://dx.doi.org/10.24327/23956429.ijcmpr20180479



RECURRENT DESMOPLASTIC AMELOBLASTOMA OF MAXILLA- A RARE CASE REPORT

Surabhi Gupta*., Ankur Mudgal., Pragya Singh and Pooja Agrawal

Department of Radiotherapy, S.N. Medical College, Agra (UP) 282005

ARTICLE INFO

Article History:
Received 2nd April, 2018
Received in revised form 10th May, 2018
Accepted 25th June, 2018
Published online 28th July, 2018

Key words:
desmoplastic ameloblastoma, recurrence, radiotherapy, chemotherapy

ABSTRACT

Ameloblastoma is benign, locally aggressive, polymorphic neoplasm of proliferating odontogenic epithelial origin. Desmoplastic variant account for 4-5% of all ameloblastomas. The present study describes the case of a patient that experienced numerous recurrences in the maxilla and also discusses recommendations for treatment. A 18 year old female underwent left maxillectomy for complain of swelling over left cheek. Histopathology report revealed it to be a desmoplastic ameloblastoma. Patient than received external beam radiotherapy to head and neck region 60Gy in 30 fractions and then the patient had recurrence four years later with huge mass seen on left side of hard palate with extension from soft palate. Patient received chemotherapy and then planned for radiotherapy 20 Gy in 5 fractions after which wide local excision was done. Now patient is asymptomatic and on follow up.

INTRODUCTION

Desmoplastic variant of ameloblastoma is a rare , benign, slow growing but locally aggressive tumor.[1] It is a polymorphic neoplasm originating from proliferating odontogenic epithelium.[2] Common sites involved are mandible (80%) and maxilla.[3] Due to less number of cases there are no specific guidelines for the management of the tumor except surgical excision.[4] In this case report, we are highlighting the role of adjuvant radiotherapy and chemotherapy in the management of recurrent desmoplastic ameloblastoma as well as close follow up of these patients due to high recurrence rate.

Case Report

A 18 year old female referred to our opd in September 2011 from the department of ENT where she underwent left maxillectomy 15 days back for complain of swelling over left cheek, headache, recurrent common cold, difficulty in chewing since 2 months and on examination there was a healthy stitch line present along left lower eyelid from lateral canthus to medial canthus with left infraorbital swelling and a scar mark seen from medial canthus to left ala of nose and in the oral cavity left maxilla was absent with a healthy stitch line. Pre op CT PNS showed soft tissue densities in left maxillary sinus with extensive bony destruction and histopathology report revealed it to be a desmoplastic ameloblastoma

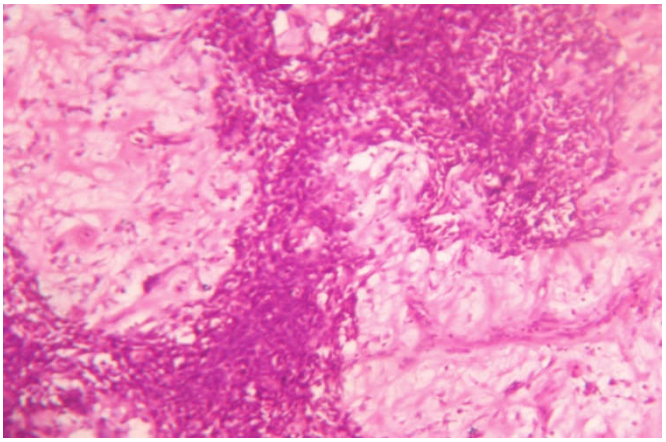


Figure-1

After ruling out metastasis, patient was planned for external beam radiotherapy to head and neck region 60Gy in 30 fractions in October 2011 after which patient was kept on monthly follow up. Patient remained asymptomatic till December 2013 after which the patient got defaulted for one year and reported in May 2015 with complains of blood stained sputum and difficulty in swallowing since two months. On examination a huge mass seen in left side of hard palate with extension from soft palate. CECT PNS showed soft tissue mass lesion 3.8 x 3.8 cm in post maxilectomy area extending into left nasal cavity, oral cavity and left side of tongue.

Corresponding author:* **Surabhi Gupta
Department of Radiotherapy, S.N. Medical College, Agra (UP) 282005

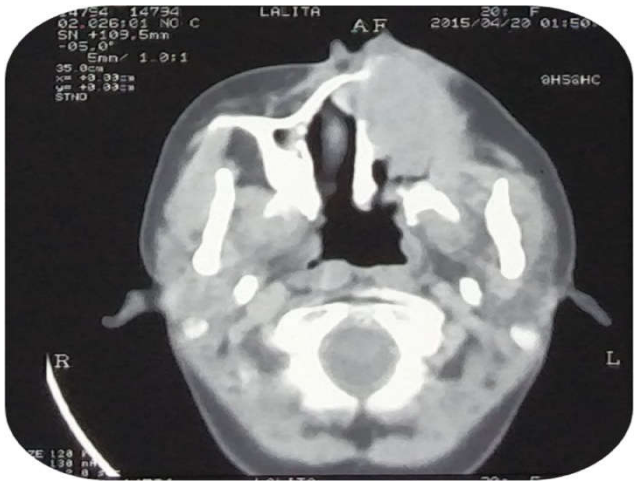


Figure-2

Patient received 2 cycle chemotherapy (paclitaxal+cisplatin+5 FU) and than referred to higher centre for debulking surgery but patient got defaulted again and reported in sep 2015 with progressive disease which was protruding through the mouth.

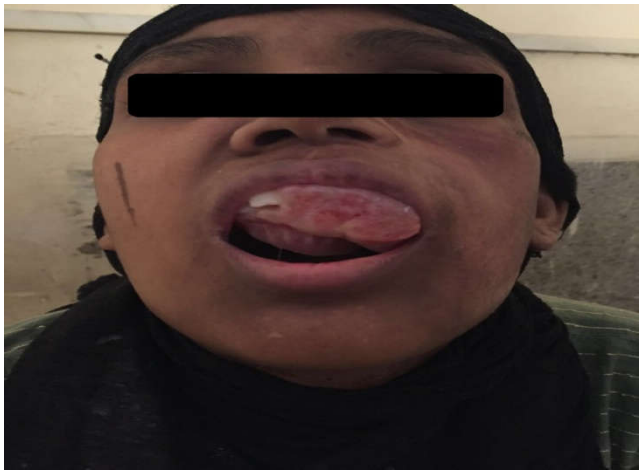


Figure-3

Surgical opinion was sought but surgery was not feasible due to very huge mass so patient was planned for palliative radiotherapy 20 Gy in 5 fractions after which patient got some symptomatic relief than patient received 3 more cycles of chemotherapy(PCF) and then she underwent wide excision in april 2016 after good tumour debulking and histopathology report revealed it to be desmoplastic ameloblastoma . Patient kept on Methotrexate weekly with Geftinib and received 5 cycles till September 2016.

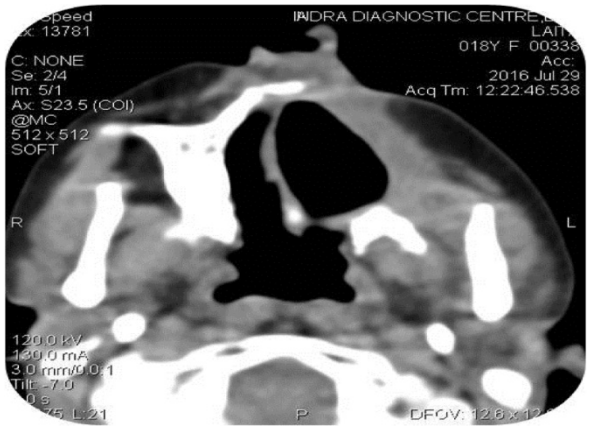


Figure-4

Patient then developed swelling of right orbital region and on examination there was a globular mass in right nasal cavity which was then evaluated with CECT PNS which revealed fronto ethmoidal sinusitis with obstructed right osteomeatal unit with DNS towards right side.

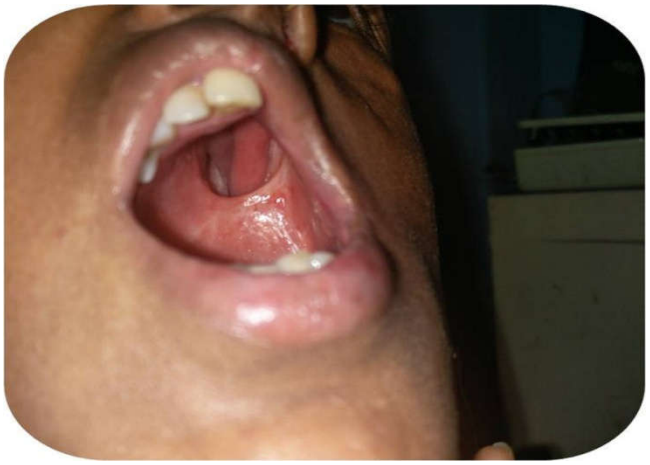


Figure-5

Patient then shifted to (doxorubicin + cisplatin) and received 6 cycles till February 2017 . Now patient is asymptomatic and was kept on monthly follow up .

DISCUSSION

Desmoplastic ameloblastoma is a rare, benign odontogenic tumor with locally aggressive clinical behavior occurring in around 0.9- 12 % of ameloblastomas. [4] Cases have been reported in patients aged 18 to 70 years with a mean of 41.2 years.[5] No difference between sex has been reported. [2] These tumors generally presents with complain of loose teeth, a swelling, paresthesias, and pain on the face.[4] Histopathologically, desmoplastic ameloblastomas are nonencapsulated tumours with extensive collagenous stroma or desmoplasia containing small islands and nests of ameloblast cells.[3]

Surgery is the primary treatment modality for these types of tumors.[4,5,6] . Various procedures which can be used are wide resection , electrocautery , cryotherapy, enucleation , curettage or marsupialization. Instead of using wide excision these tumors have high propensity of recurrence. The recurrent lesion may be treated by reexcision.

Patients having tumors that cannot be completely resected specifically tumors located in posterior maxilla poses a great treatment challenge.[5] Posterior maxillary tumors are dangerous because of their close proximity to the orbit, pterygopalatine fossa and risk of direct intracranial extension. [7,8]. We also faced similar problem in our case for which we used chemotherapy and radiotherapy . Gardner et al in his study recommended that irradiation can reduce the size of the ameloblastoma, especially the soft-tissue component , these lesions responded well to irradiation , and doses of 50–60 Gy in 5 to 6 weeks had resulted in substantial regression , even of large tumors. [6]Atkinson et al reported a case series of 10 patients in which two patients underwent total excision and postoperative RT and one patient underwent subtotal excision and radiotherapy; all 3 remained alive and disease-free at 27 months, 30 months and 5 years after treatment. [7] Seven patients received radiotherapy alone out of which one patient had stable disease while others responded well and remained locally controlled .[7]

The efficacy of chemotherapy in the management of primary and recurrent ameloblastomas is still being explored as chemotherapy can improve clinical outcomes in non-surgical patients.[6,8,9] Several drug regimens may be used in combination with surgical resection and/or or radiotherapy. These include the combinations of vinblastine + cisplatin + bleomycin; adriamycin + cisplatin + cyclophosphamide; doxorubicin + cisplatin; and gemcitabine + carboplatin .[9] However, there is still a need for more multicenter randomized controlled clinical studies to validate the use of radiation and chemotherapy as treatment options for ameloblastoma.

CONCLUSION

Although Ameloblastomas are benign tumors but they have high rate of recurrence and show aggressive nature in maxillary region. It is evident that surgery is the treatment of choice, however, chemotherapy and radiotherapy can be used before surgery for debulking the tumor.

References

1. Kennedy WR, Werning JW, Kaye FJ, Mendenhall WM. Treatment of ameloblastoma and ameloblastic carcinoma with radiotherapy. *European Archives of Oto-Rhino-Laryngology*. 2016 Oct 1;273(10):3293-7.
2. Pillai RS, Ongole R, Ahsan A, Radhakrishnan RA, Pai KM. Recurrent desmoplastic ameloblastoma of the maxilla: a case report. *Journal-Canadian Dental Association*. 2004 Jan 1;70(2):100-4.
3. Anand R, Sarode GS, Sarode SC, Reddy M, Unadkat HV, Mushtaq S, Deshmukh R, Choudhary S, Gupta N, Ganjre AP, Patil S. Clinicopathological characteristics of desmoplastic ameloblastoma: A systematic review. *Journal of investigative and clinical dentistry*. 2017 Jul 14.
4. McClary AC, West RB, McClary AC, Pollack JR, Fischbein NJ, Holsinger CF, Sunwoo J, Colevas AD, Sirjani D. Ameloblastoma: a clinical review and trends in management. *European Archives of Oto-Rhino-Laryngology*. 2016 Jul 1;273(7):1649-61.
5. Effiom OA, Ogundana OM, Akinshipo AO, Akintoye SO. Ameloblastoma: current etiopathological concepts and management. *Oral diseases*. 2017 Jan 31.
6. Gardner DG. Radiotherapy in the treatment of ameloblastoma. *International journal of oral and maxillofacial surgery*. 1988 Jun 1;17(3):201-5.
7. Atkinson CH, Harwood AR, Cummings BJ. Ameloblastoma of the jaw. A reappraisal of the role of megavoltage irradiation. *Cancer*. 1984 Feb 15;53(4):869-73.
8. Huang CM, Chen JY, Chen CH, Huang CJ. Radiotherapy for a repeatedly recurrent ameloblastoma with malignant transformation. *Head & neck*. 2014 Jan 1;36(1).
9. Philip M, Morris CG, Werning JW, Mendenhall WM. Radiotherapy in the treatment of ameloblastoma and ameloblastic carcinoma. *Journal-hong kong college of radiologists*. 2005;8(3):157.

How to cite this article:

Surabhi Gupta *et al* (2018) 'Recurrent Desmoplastic Ameloblastoma of Maxilla- A Rare Case Report', *International Journal of Current Medical And Pharmaceutical Research,* 04(7), pp. 3434-3436.

****