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Case Report

Aggressive Giant Desmoplastic Ameloblastoma -A unique case

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ABSTRACT:

Ameloblastoma is one of the most common odontogenic tumors. Desmoplastic Ameloblastoma(DA) is a relatively rare variant. Compared with conventional ameloblastoma, DA shows distinct radiographic features which mimic a fibro-osseous lesion i.e with mixed radiopacity and radiolucency. The local invasive nature and high recurrence rate of this variant necessitates surgical treatment with resection. DA most commonly affects anterior region of the jaws. We present here a unique case of aggressive, giant desmoplastic ameloblastoma in a 50 years old female.

Keywords: Aggressive, Desmoplastic ameloblastoma, Odontogenic tumor, Stromal Desmoplasia

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INTRODUCTION

Eversole[1] in 1984 described desmoplastic variant of ameloblastoma as 'ameloblastoma with pronounced desmoplasia'.WHO,2003 classification of odontogenic tumors includes desmoplastic as a rare variant of ameloblastoma. With respect to age/sex distribution, Desmoplastic ameloblastoma (DA) shows similar features to conventional ameloblastomas. However, site of occurrence viz-anterior or premolar region in either jaws, distinct radiographic & histopathologic features make DA stand as unique, separate entity. Purpose of this article is to present a unique case of huge aggressive desmoplastic ameloblastoma that occurred in an unusual site.

CASE REPORT

50 year old female reported with complaint of facial asymmetry due to long-standing, asymptomatic swelling in left mandible involving anterior and preauricular region since 3 years. History of previous surgical intervention approximately 2 years back following which no change in size of swelling was reported. Past medical & family history was insignificant. Extraoral examination revealed hard, non-fluctuant, non-tender, huge, expansile, diffuse swelling approximately10x5x3cms extending from left preauricular region crossing midline till 45 region. TMJ movements were bilaterally palpable but reduced on left side. Swelling in post-ramal region caused displacement of ear lobule. Overlying skin was tense and shiny. Submandibular and submental lymph nodes could not be palpable. Lips were incompetent. Scar along submandibular region suggested previous surgical intervention.

Intraorally, swelling extended from 45 region to left ascending ramus region with ballooning of buccal and lingual cortices.33,34,35,36 were missing Remaining teeth were displaced & grade III mobile. Overlying mucosa was erythematous, ulcerated due to trauma or indentations from maxillary teeth. Distended blood vessels were visible. X ray examination revealed lesion extends from 46 to left ramus of mandible involving coronoid process & sparing posterior border of ramus and condyle and showing root resorption of 31 to 38. Differential diagnosis of fibro-osseous lesion was considered; patient was advised blood investigations viz-serum calcium, phosphorous and alkaline phosphatase levels, which were within normal limits. Incisional biopsy confirmed pathology as an Ameloblastoma.Surgical resection of tumour mass with disarticulation of left condyle and primary reconstruction with titanium plate was performed.Resected mass was irregular,& distorted measuring approximately 11cmx 6cmx 7cm, non-

encapsulated & firm in consistency. Histopathological features confirmed desmoplastic ameloblastoma.

Fig. 1: Clinical photograph showing massive facial swelling.

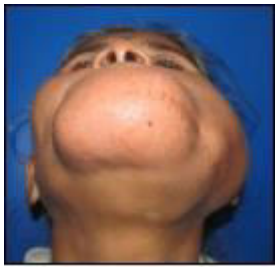

Fig. 2: 3DCT face showing extensive mixed lesion.

Fig. 3: Showing resected specimen.

DISCUSSION

Desmoplastic ameloblastoma is characterized by marked stromal desmoplasia[2] & generally occurs in 3rd–5th decade. Japanese are the most commonly affected race.Clinically,DA commonly occurs in anterior/premolar region of either jaw.Kawai et.al[3] reported a marked predilection for anterior region of both jaws. Effiom and Odukoya[4] reported that 81% of cases showed mandibular predilection, However in our case it involved almost whole mandible sparing right coronoid,ramal and angle regions which is unique in itself. As compared to mandible, maxillary lesions are more dangerous because of easy invasion of adjacent structure due to weak nature of maxillary bone[5].Tooth displacement in 92% and root resorption in 33% cases have been noted[4]. Our case shows similar features. Study by Hideaki et al[6] showed striking difference between DA and other ameloblastomas in anatomic distribution.Due to invasive nature of pathology compounded by delay in treatment, lesion had extended across the midline of mandible to antegonial notch of opposite side.Wide resection followed by primary reconstruction of bony defect should have been ideal choice of treatment.However due to low socioeconomic status and anxiety related to additional surgery for bone graft harvesting,patient insisted only for removal of pathology.Wide resection and placement of reconstruction plate was carried out.Loss of lip support led to functional deficit related to speech and deglutition.However patient persistence with functional exercises compensated for deficit.

In most cases,radiographic appearance is often more typical of fibro-osseous lesions.Mixed radiographic appearance is due to infiltrative growth pattern of tumor cells into marrow space and simultaneous osteoblastic activity leading to bony flecks giving it mixed appearance[7]. These features may also be prognostic significance in predicting tumor behaviour.

Histologically,DA is characterized by extensive stromal collagenization or desmoplasia with small nests and strands of odontogenic epithelium,which were also found in our case. Marx and others reported that tumour extended 2.3–8.0 mm beyond radiographic margin from an analysis of 34 mandibular ameloblastomas.Hence,resection of 1 cm of normal-appearing bone beyond the radiographic margin has been recommended. Because of invasive nature and high recurrence, surgical resection of involved tissue is treatment of choice.Long term follow-up is mandatory.Patient is under observation since 2 years and is satisfied with outcome.

CONCLUSION

Desmoplastic ameloblastoma is relatively rare entity among ameloblastoma variants and due to its aggressive nature necessiates wide resection beyond radiologically evident margins.Conservative treatment such as enucleation which probably must have been employed in this case in past could have been the cause for recurrence.Wide resection with primary reconstruction and long term follow-up is therefore key to successful treatment of desmoplastic ameloblastoma.

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