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CASE REPORT

DESMOPLASTIC AMELOBLASTOMA- A CASE REPORT

G.S.Sangeetha, C.Ganesh , G.Maragathavalli

1.Past . Post graduate student, Department of Oral Medicine and Radiology, Saveetha Dental College, 162 Poonamallee high Road, Velappanchavadi, Chennai, Tamilnadu
2. Senior Lecturer, Department of Oral Medicine and Radiology, SRM Kattankulathur Dental College, Potheri,Chennai, Tamil Nadu.
3. Professor and Head, Department of Oral Medicine and Radiology, Saveetha Dental College, 162 Poonamallee high Road, Velappanchavadi, Chennai, Tamilnadu.

Corresponding Author: Dr.G.S.Sangeetha, MDS, 12/108, Kannagi Street, Kumananchavadi, Chennai-56, Tamil Nadu.

Abstract
Desmoplastic Ameloblastoma (DA) is a rare variant of ameloblastoma. 96 cases of desmoplastic ameloblastoma have been reported so far in literature. We are presenting a rare case of desmoplastic ameloblastoma involving the body of mandible with regards to its clinical and radiographical viewpoints. Reviewing the literature we found that DA most commonly occurs in males in the 3rd to 5th decade of life with a high preponderance in the anterior region of either jaw. Radiological appearance was most commonly of a mixed radio-lucent/radio-opaque type with a high incidence of poorly defined borders. However our case differed from the reviewed cases as it was found to be a unilocular lesion with well defined borders. Histopathological findings of our case was consistent with the hisotpathological appearance of the reviewed cases and showed extensive stromal desmoplasia and small tumour nests of odontogenic epithelium scattered in stroma..
Keywords: Desmoplastic ameloblastoma, odontogenic tumor, stromal desmoplasia

INTRODUCTION

Ameloblastomas are common benign epithelial odontogenic tumours that are locally invasive in nature. Ameloblastoma (AM) of the jaw is a neoplasm that accounts for about 1% of all cysts and tumors of the jaws and 18% of the various odontogenic neoplasms[1] Histopathology of ameloblastomas have been extensively reviewed in numerous publications and it is found that AM may manifest with a number of histological patterns, including the follicular, plexiform, acanthomatous, keratinizing, granular cell, basal cell and clear cell types [2]. However in recent years the histomorphological spectrum of AM has expanded to include a desmoplastic variant. This subtype was first identified by Eversole et al., in 1984 and to the best of our knowledge, 96 cases have been reported so far in the literature[3].Clinically this tumor has a predilection for occurrence in the anterior region of maxilla or mandible .DA present radiographically as a unilocular or multilocular radiolucency or as mixed radiolucent / radiopaque areas with either well-defined or poorly defined borders. However in most of the

cases their radiographic appearances are more typical of the fibro-osseous lesions with well-defined or poorly defined borders. Takigawa et al and Uji et al were the early ones to draw attention to this unusual variant characterized by extensive stroma; desmoplasia with small compressed nests and strands of Odontogenic epithelium [4].Immunohistochernical studies suggest that the desmoplasia originate from de novo synthesis of extracellular matrix proteins [5]

The purpose of this article is to present a case of desmoplastic ameloblastoma that has occurred in an unusual site and has a unique radiographic appearance and to provide a brief review

Case report

A 42 year old female patient was referred to our Department of Oral diagnosis and Radiology for evaluation of an asymptomatic slowly enlarging swelling in the left lower jaw since 2 months. There was no history of trauma or any remarkable medical history.

Extraoral examination revealed gross asymmetry towards the left side of the face(fig:1) .Intraoral examination disclosed a solitary swelling of size 4x3 cms in the edentulous area(missing 35 and 36) of the third quadrant extending anteriorly from the distal aspect of 34 to the mesial aspect of 37 posteriorly, Buccolingually extending from buccal vestibule to the floor of the mouth. The mucosa over the swelling is erythematous andlobulated with well defined margins.(fig:2) On palpation the swelling was non-tender and firm in consistency. The surrounding structures were normal. No Lymphadenopathy or fistula present. Panoramic radiograph showed an unilocular radiolucency with impacted 35.rom the distal aspect of 34 to the mesial aspect of 37 .Impacted -38,48; Missing in relation to 16,17,26,36;Root stumps in relation to 18,Root resorption in relation to37.(fig:3) Aspiration of the lesion was unproductive. From clinical and radiographic findings a possible diagnosis of dentigerous cyst and ameloblastoma was made. To obtain a correct diagnosis excisional biopsy was taken. Section showed collagenous stroma with epithelial component reduced to marrow compressed strands of epithelium.The epithelial cells in the periphery of the strands and cords are flattened or cuboidal with hyperchromatic nuclei in certain areas the strands are broadened to form large islands with peripheral rim of cuboidal cells and acompact centre in which spindle shaped cells assuming whorl like pattern.Connective tissue shows dense collagenous stroma with occasional tumor islands and it is hyalinised and hypocellular suggestive of Desmoplastic ameloblastoma(fig:4) All laboratory investigations done before the surgical procedures were found to be in normal limits. It was decided to completely enucleate the lesion. Extraction of impacted 35,38 and GradeII 37 was done simultaneously .

FIG -1:Swelling in the left side of the face

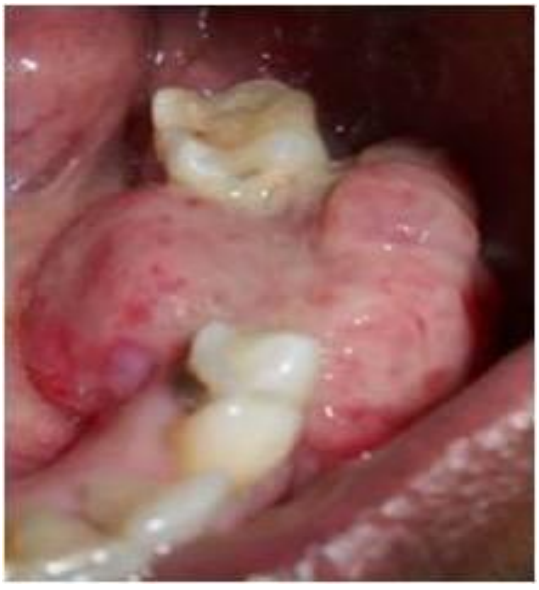

FIG -2: Intraoral view the lesion

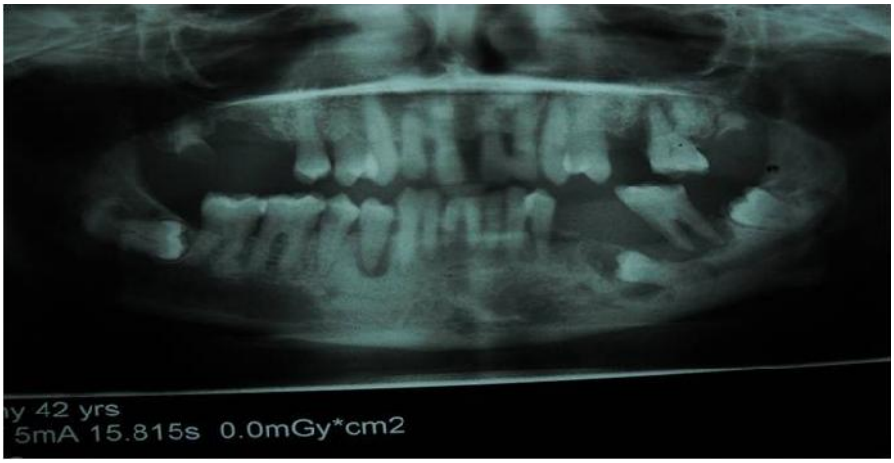

FIG -3 OPG reveals unilocular radiolucency with impacted left premolar

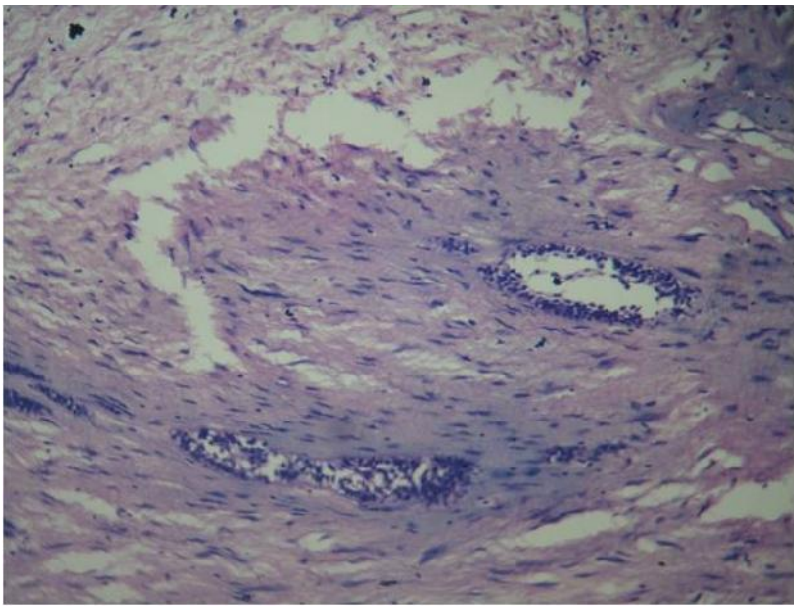

FIG-4 Histopathological view of the lesion.

DISCUSSION

Desmoplastic ameloblastoma (DA) is a rare variant of ameloblastoma (AM). And have been widely reported in the last two decades since its first description given by Eversole. It constitutes 0.9% to 12.1% of all AMs[6]

The mean age at initial presentation is 42.3 years (range 17-70 years) . Males are more commonly affected than females. Demographic studies have revealed that the highest incidence of this tumor is in patients of the Japanese race [7]
The study of 17 cases by Kezler *et al* [8]also showed mandible to be involved in 83% and maxilla to be involved in 17% cases. According to our review, predilection of occurrence were seen in anterior region of either maxilla or mandible as also reported by Kawai *et al*[9] However our case differed considerably because of its occurrence in the posterior region of the mandible.

Leon Barnes has categorized AMs into four types on the basis of the behavioral pattern, anatomical location, histological features and radiographic appearance; these are the solid (multicystic), unicystic, desmoplastic and peripheral varieties. The first three are intraosseous (central), while the last is extraosseous (mucosal). DAs are described as poorly defined in 88% of cases.

As per the reviewed cases majority of desmoplastic ameloblastomas showed mixed radiolucent/radioopaque appearance which can be due to infiltrative growth pattern of tumour cells into surrounding marrow spaces and simultaneous vigorous osteoblastic activity leading to number of bony flecks which gives a mixed radiolucent/radioopaque appearance [9] ,this was in contradiction to our case with unilocular radiolucency and well defined borders. Root resorption is a common finding in DA which was present in our case [10]
Desmoplastic ameloblastoma should always be considered in the differential diagnosis of mixed radiodense radiolucent lesion with diffused borders. Other possibilities are fibro-osseous lesions, Cemento ossifying fibroma, Cementoma(intermediate stage) cementoma, calcifying odontogenic cyst.
Histopathology showed extensive stromal desmoplasia and small tumor nests of odontogenic epithelium .The epithelial cells about the periphery of the epithelial islands are usually cuboidal and occasionally hyperchromatic ,however occasional islands showing columnar peripheral cells with reversed nuclear polarity.

CONCLUSION:

The lesion in the present case deviates from the usual desmoplastic variant of ameloblastoma in terms of site and radiological appearance.Proper diagnosis and treatment of neoplasm should be done,As the majority of DAs present with radiographic features resembling that of fibroosseous lesions, a definitive diagnosis should always be based on the histology and not the radiographic and clinical findings

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