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Case Report

DESMOPLASTIC AMELOBLASTOMA OF MANDIBLE - A RARE VARIANT

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ABSTRACT

Ameloblastoma is a benign and locally aggressive epithelial odontogenic tumor, most commonly located in the mandible mainly in the molar and the ramus region. It exhibits histological variation which includes follicular, acanthomatous, granular cell, desmoplastic, basal cell, keratotic, calcifying, osteoplastic and ossifying types.

Desmoplastic Ameloblastoma is a rare variant of ameloblastoma with specific clinical, histological and radiological features. Desmoplastic Ameloblastoma (DA) was first reported by Eversole et.al in 1984 as an unsual variant of ameloblastoma characterized histologically by desmoplastic stroma.

Keywords: Ameloblastoma, Desmoplastic Ameloblastoma, Benign Fibro-Osseous Lesion, Odontogenic Tumor, Desmoplastic, Stromal Desmoplasia.

INTRODUCTION

Ameloblastoma is a tumor arising from the odontogenic epithelium. It is said to be true neoplasm of enamel organ. Robinson defined it as "usually unicentric, nonfunctional, intermittent in growth, anatomically benign and clinically persistent". Ameloblastoma accounts for about 1% of all cysts and tumors of the jaw and about 18% of various odontogenic neoplasms. Histologically it is classified as follicular, plexiform, acanthomatous, granular cell, basal cell and desmoplastic variants.

DesmoplasticAmeloblastoma is one of the histological variants of ameloblastoma. The first detailed report on the desmoplastic variant was given by Eversole et al in 1984 and they called it "ameloblastoma with pronounced desmoplasia." DesmoplasticAmeloblastoma was included in WHO classification of Head and Neck Tumors(WHO-2005) as a variant of ameloblastoma.[9]

DA strikingly differs from other forms of ameloblastoma in its anatomical location, morphology and radiographic appearance. DA usually occurs in 2nd to 5th decade of life with an almost equal gender ratio, same as that of ameloblastoma. Clinically DA appears as a solid mass with a predilection for occurrence in the anterior or premolar region of maxilla and mandible which is an unusual position for ameloblastoma. It is also more complicated to be treated because of its tendency for penetrating the surrounding bone.

Radiographically it presents as mixed radio opaque-radiolucent lesion which resembles a fibro-osseous lesion with well-defined or poorly defined borders.

Histologically it is characterized by extensive stromal collagenization/desmoplasia which is surrounded by compressed small or irregular islands of odontogenic epithelium.

This case report presents a case of DA in a 50year old male patient in the left mandibular premolar region.

CASE REPORT

A 50 year old male reported to the Dept. of Oral and Maxillofacial Surgery with a chief complaint of an asymptomatic swelling of spontaneous origin in the lower left premolar region which he noticed while shaving. The swelling gradually increased in size for 2 years and then remained stable for 6-7 months. The patient reported a history of a slowly enlarging mandibular mass, without bleeding, pain or sensory changes.

The physical examination revealed facial asymmetry (Fig 1) due to swelling on the left side of the face at left corner of mouth in the canine region extending upto lower border of mandible which was oval in shape. The skin over the swelling

appeared normal. It was non tender on palpation. No cervical lymph nodes were palpable.

Figure 1: Clinical Apperance

The intra-oral examination disclosed a large, hard non-tender mass of the anterior mandible covered by an intact overlying mucosa(Fig 2). It extended from the distal of canine till the mesial of 1st molar with obliteration of buccal vestibule(Fig 3). The swelling was ovoid in shape measuring about 2cm*5cm. Expansion was also seen in the lingual cortical plate(Fig 4). Canine and 1st premolar showed displacement. The involved teeth were vital and exhibited no mobility.

Radiographic examination of the mandible revealed a diffuse ill-defined mixed radiolucent/radiopaque lesion extending from the distal surface of canine upto mesial of 1stmolar(Fig.5). Small flecks of radiopacities were seen within the lesion. There was loss of lamina dura around the involved teeth. The lesion caused displacement of roots of canine,1st and 2nd premolars without any signs of root resorption(Fig.6). Expansion of buccal cortical plate was evident on mandibular occlusal radiograph(Fig.7). Areas of calcification were seen within the lesion giving it a soap bubble appearance.[17-22]

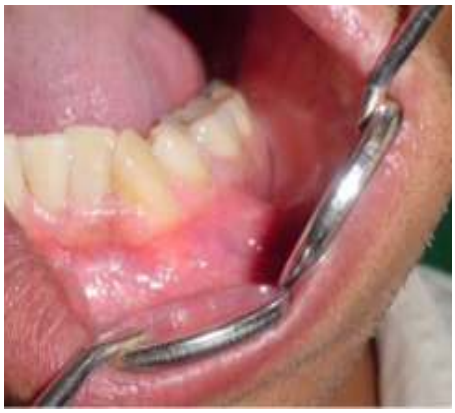

Figure 2: Intact overlying mucosa

Figure 3: Obliteration of buccal vestibule

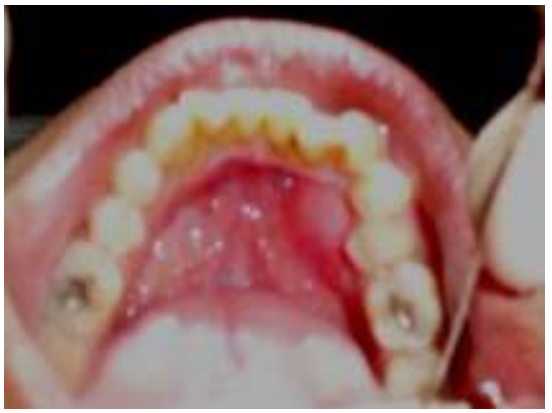

Figure 4: Expansion of lingual cortical plate

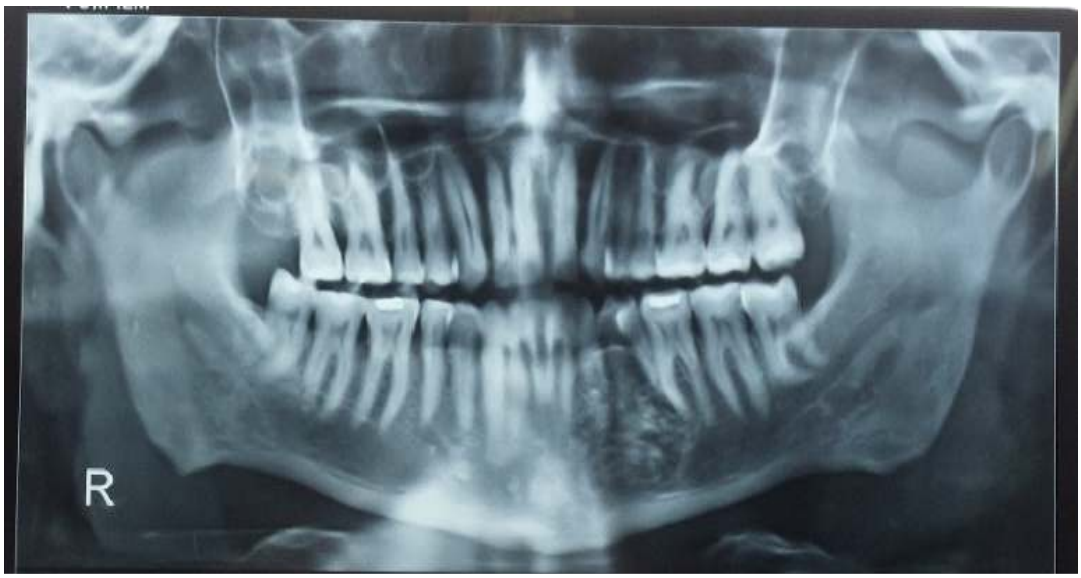

Figure 5: OPG showing the lesion in area of 33-36.

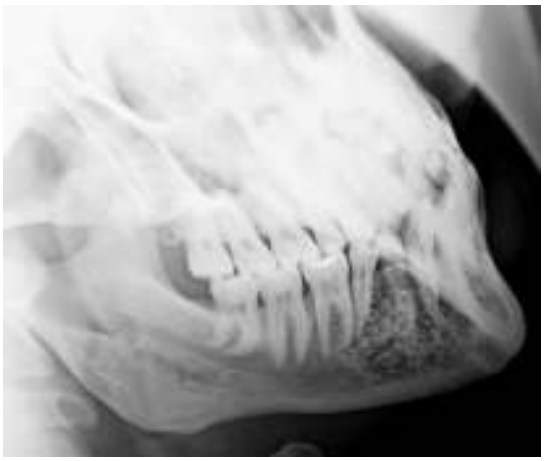

Figure 6: Lateral oblique radioraph

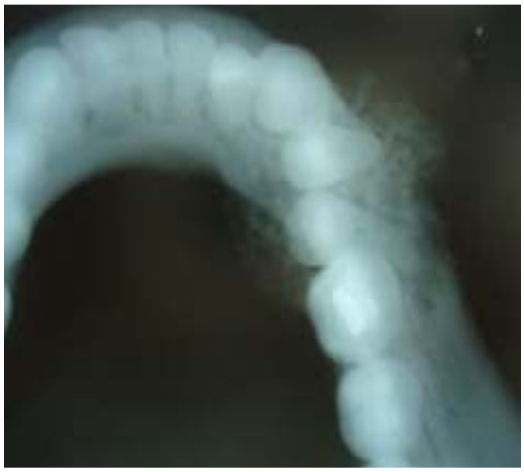

Figure 7: Occlusal radiograph

Based on clinical and radiographic appearnace a provisional diagnosis of odontogenic tumor with a differential diagnosis of cemento-osseous dysplasia, cemento-ossifying fibroma, traumatic bone cyst was made.

An incisional biopsy was performed under LA to establish definitive diagnosis.

Histopathological examination revealed dense collagen stroma with odontogenic epithelial cells arranged in form of long thin strands. Hyperchromatic epithelial cells were observed. The prolifertaion was surrounded by dense stroma which confirmed the diagnosis of of desmoplastic ameloblastoma(Fig 8, Fig 9).

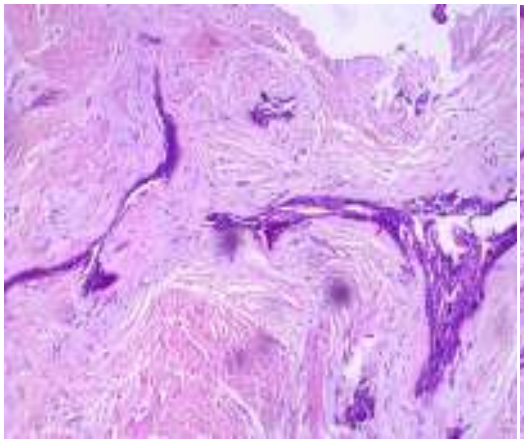

Figure 8: Presence of hyperchromatic cells

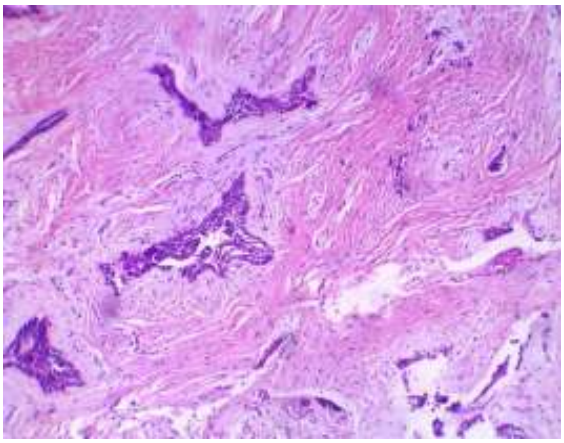

Figure 9: Dense stromal proliferation

The patient was investigated pre-operatively,fitness certificate was obtained from physician and patient was prepared under usual surgical pattern for surgery under general anesthesia. IAN block was given on the left side. A trapezoidal incision starting from left lateral incisor along the crevicular margins till distal of left 2nd molar was taken. Mucoperiosteal flap was reflected, mental nerve was located and part of the nerve crossing the lesion was seperated carefully(Fig 10). Segmental resection from canine to 1st molar was carried out, maintaining the lower border and the lingual plate intact(Fig 11). Wound closure was done using 3-0 vicryl. The postoperative course was uneventful.

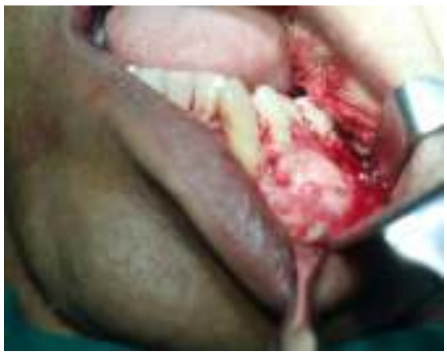

Figure 10: Surgical exposure of the lesion

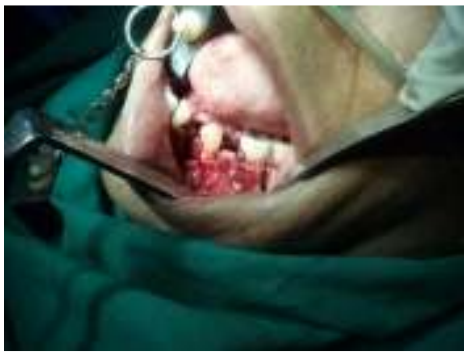

Figure 11: Segmental resection of the lesion

DISCUSSION

Desmoplastic Ameloblastoma exhibits important differences in anatomic distribution, histologic appearance and radiographic findings compared with other types of ameloblastomas. Until now approximately 150 cases are reported in the literature.[8] There is no sex prediliction observed, it occurs in 3rd to 5th decade of life with equal male to female ratio. A painless swelling or bony expansion is the conspicuous feature of DA.

DA constitutes 0.9% to 12% of all ameloblastomas. It has been suggested that DA arises from the periodontal membrane of the related tooth. Some suggest DA might also arise from the epithelial cell rest of Malassez in the periodontal membrane.[8]

DA is usually occur in the anterior or premolar region which is in contrast to the location of classic types of ameloblastoma, which are usually found in the posterior area of mandible. Maxillary lesions are more insidious than mandibular tumors owing to the proximity of vital structures and also maxillary sinus.[5]

DA exhibits a more aggressive behaviour as compared to other ameloblastomas[2]. It may be due to:

1. Its potential to grow to a large size.
2. Common location in maxilla leading to an early invasion of adjoining structures.
3. Diffuse radiographic appearance of the lesion.
4. Invasion into the bone (Histological Finding)[8]

   The most common feature of DA is tooth displacement around 92% in all the cases and root resorption around 33%.

   Radiographically DA usually appears as a mixed radiolucent and radiopaque lesion mimicking a benign fibro-osseous lesion. The mixed radiographic apearance is due to osseous metaplasia within the dense fibrous septa that characterizes the lesion.

A conformatory diagnosis is made by histopathological evaluation. The features seen are:

1.Stromal Desmoplasia

2.Islands of different shapes in the epithelial component.

3.Peripheral layer of cuboidal cells.

4.Hyper-cellular central area composed of spindle shaped epithelial cells[5-15].

Ng & sia using various histochemical techniques demonstrated that the tumor cells of DA showed variable expression of S-100 protein and desmin[5,8]. It also exhibited a strong positive reaction for collagen type VI. This was interpreted as an active de novo synthesis of extra cellular matrix protein, hence ruling out scar tissue.

Enucleation or curretage alone of the lesion may lead to recurrence as there is indistinct bone border between the tumor and normal bone tissue. Therefore complete resection is treatment of choice.

CONCLUSION

Due to unique features, the biologic behaviour of DA is still not fully understood. For proper understanding of such cases, more in depth analysis and long term follow-up is required. The clinician has to be alert regarding correct diagnosis and correct treatment of the lesion. The definitive diagnosis requires histopathological examination due to its potential for recurrence.

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