



CASE STUDY

DESMOPLASTIC (HYBRID) AMELOBLASTOMA- AN ENIGMA

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ABSTRACT

Ameloblastoma is the most commonly encountered clinically significant odontogenic tumour and its incidence appears to equal or exceed the combined incidence of all other odontogenic tumours exclusive of odontomas. Various histopathologic subtypes of ameloblastomas are recognised: follicular, acanthomatous, granular cell, basal cell, desmoplastic, plexiform and unicystic. Most tumours are found to be composed purely of one histopathologic subtype but sometimes mixtures of different patterns can be observed. The desmoplastic variety was first observed as a distinct variant of ameloblastoma by Eversole *et al* in 1984 with a marked predilection to occur in the anterior-premolar region, distinctive histologic features and radiologic characters mimicking benign fibro-osseous lesions. We report a unique case of a hybrid ameloblastoma containing both desmoplastic and plexiform histology with unusual clinical and radiological presentation.

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INTRODUCTION

Ameloblastoma is the most commonly encountered clinically significant odontogenic tumour and its incidence appears to equal or exceed the combined incidence of all other odontogenic tumours exclusive of odontomas. Various histopathologic subtypes of ameloblastomas are recognised: follicular, acanthomatous, granular cell, basal cell, desmoplastic, plexiform and unicystic (Waldron *et al.*, 1987). Most tumours are found to be composed purely of one histopathologic subtype but sometimes mixtures of different patterns can be observed. The desmoplastic variety was first observed as a distinct variant of ameloblastoma by Eversole *et al* in 1984 with a marked predilection to occur in the anterior-premolar region, distinctive histologic features and radiologic characters mimicking benign fibro-osseous lesions (Eversole *et al.*, 1984). We report a unique case of a hybrid ameloblastoma containing both desmoplastic and plexiform histology with unusual clinical and radiological presentation.

Case report

A male patient aged 23 years reported to the Department of Oral and Maxillofacial Surgery with a history of progressive

swelling and mild associated pain on the right side of the face since 6 months. The swelling measured around 4x3 cms, obliterated the right lower vestibule and extended from right lower canine to the distal of 2nd molar. The swelling was firm and mildly tender on palpation. The overlying mucosa was normal in colour and consistency. There was mobility and mild displacement of the right lower premolars and second molar. There was no paresthesia present on the right side of the mandible. On radiographic examination, the orthopantomogram revealed a mixed radiopaque-radiolucent lesion present in the region of the right first and second molars which gradually merged with a well circumscribed unilocular radiolucency present in the premolar region. Root resorption was evident in the premolars. The lower border of the mandible was intact. Computed tomography demonstrated an expansile multiloculated lesion in the right body region of the mandible measuring approximately 4cms x2.8cmsx1.5cms. Aspiration from the anterior part of the lesion yielded a turbid brown fluid whereas no fluid was aspirated from the posterior aspect of the lesion, which led to the tentative diagnosis of benign tumour with cyst formation. Incisional biopsy from the lesion provided the provisional diagnosis of plexiform ameloblastoma. All laboratory investigations were carried out and were found to be within normal limits before the surgical procedure. The patient refused to give his consent for resection as is the accepted treatment, hence it was decided to follow a conservative treatment option. Taking into consideration the

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young age of the patient and the fact that the patient was explained the need for long follow up and second surgery in case of recurrence, complete surgical excision of the lesion was done followed by peripheral osteotomy to obtain healthy bony margins. Lingual plate was intact and bony curettage was carried out for it. Chemical cauterization was done with Carnoy's solution as an additional measure to prevent recurrence. The associated premolars and second molar were also extracted. Surgical wound was packed with Iodoform gauze subsequently till the wound healing was complete.

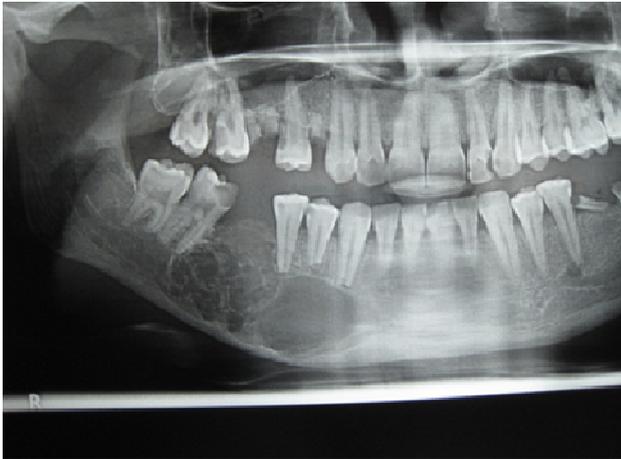


Fig.1. Orthopantomogram showing the extent of the ameloblastoma



Fig.2. Intraoperative picture showing clinical extent of the lesion

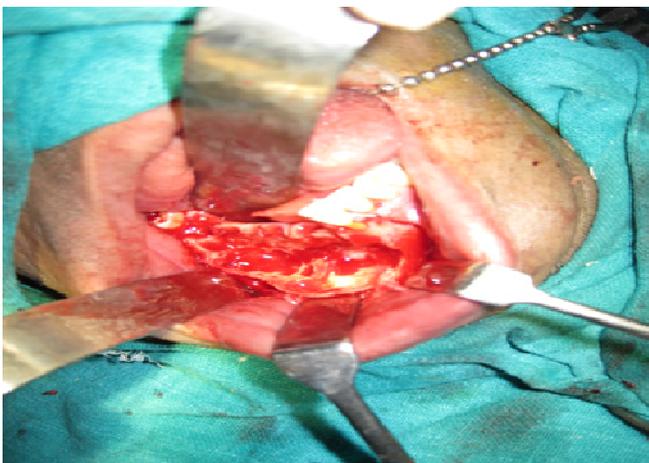


Fig.3. Excision of tumour with lingual plate intact



Fig.4. Tumour mass and resorption of roots of involved posteriors



Fig.5. 3 yrs post op OPG showing excellent healing on the right side of the mandible

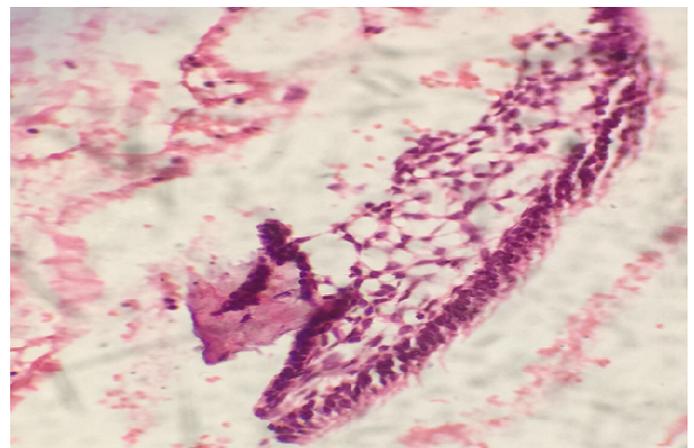


Fig. 6. Magnification 40x- showing tall columnar ameloblasts and stellate reticulum

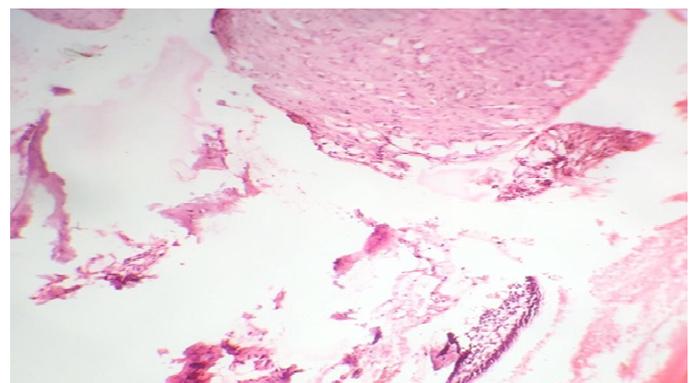


Fig.7. Magnification 40x- showing fibrous stroma

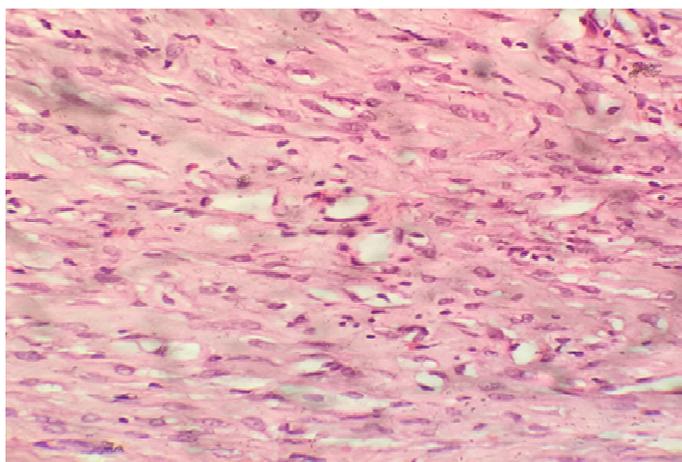


Fig.8. Desmoplastic component

Follow up at the end of one year demonstrated uneventful healing and no signs of recurrence were seen till 4years postoperatively.

Microscopic examination: The sections processed for examination showed hybrid morphology. At one end were cystic spaces in which were projecting nodes of ameloblastoma arising from the cystic lining into the lumen of the cyst completely filling it, suggestive of plexiform ameloblastoma. Separated from these cystic areas of ameloblastic epithelia were areas of dense mature collagenised stroma containing variable islands and cords of ameloblastic epithelium suggestive of desmoplastic ameloblastoma. Hence the histopathological findings seem to be those of desmoplastic pattern of ameloblastoma with intraluminal type of plexiform pattern ameloblastoma at the periphery.

DISCUSSION

Eversole *et al.* were the 1st to describe the desmoplastic ameloblastoma in the English literature as a separate entity having characteristic features of extensive stromal desmoplasia with small nests, cords and strands of odontogenic epithelium². This variant was included in WHO histopathological typing of odontogenic tumours (1992) and has been reported to occur mainly in the anterior region of the jaws and appears radiographically as a diffuse, mixed radiolucent-radioopaque lesion mimicking a benign fibro-osseous lesion (Wakoh *et al.*, 2002; Philipsen *et al.*, 2001) These lesions may occur as diffuse mixed radiolucent-radioopaque lesions or as ill-defined radiolucencies with indistinct borders and containing scattered radiopacities. Waldron and El-Mofty described in their review of 116 cases of ameloblastoma, five cases of hybrid lesions showing features of desmoplastic variant together with typical areas of follicular or plexiform ameloblastoma (Waldron *et al.*, 1987). Till date 19 cases of hybrid desmoplastic ameloblastoma have been reported, only few cases have been reported in the posterior mandible, whereas others have shown a predilection for anterior maxilla/ mandible (Waldron *et al.*, 1987; Wakoh *et al.*, 2002; Philipsen *et al.*, 2001; Sivapathasundharam *et al.*, 2007; Hirota *et al.*, 2005; Yazdi *et al.*, 2009; Ashman *et al.*, 1993). Ashman *et al.* documented a large DA alongwith follicular, plexiform, acanthomatous and granular histology. (Ashman *et al.*, 1993) Desmoplastic ameloblastoma with mucous cell differentiation or cyst formation have also been reported. Very similar to the present case, Kawai reported a

case of DA in the posterior mandible with cyst formation in the posterior portion of the lesion (Kawai *et al.*, 1999). The authors suggested that the cyst was formed by the cystic degeneration of tumour epithelial islands, nests or even microcyst phenomenon seen in this variant as well as other variants of ameloblastoma. Wakoh *et al.* also reported a case of follicular/desmoplastic hybrid ameloblastoma with radiographic features of concomitant fibro-osseous and solitary cystic lesion in the canine- premolar region (Wakoh *et al.*, 2002). Another reported case in literature on desmoplastic ameloblastoma with cystic findings accounting for half of the lesion brings the total number of such reported cases to 4 including our case (Yoshimura and Saito, 1990). The presence of intraluminal plexiform ameloblastoma with DA (as in our case) in accordance with Philipsen and colleagues could be either due to DA arising from plexiform ameloblastoma or vice-versa, hence should be termed as *collision tumour*. (Philipsen *et al.*, 2001) The radiographic appearance of the lesion has been described by many authors as honeycomb or multilocular radiolucency with floccular radioopacities but the vast majority of cases have been described radiographically as mixed radioopaque-radiolucent lesions which have to be differentially diagnosed from benign fibroosseous lesions, odontogenic fibroma or ossifying fibroma (Beckley *et al.*, 2002). This radiographic feature has been attributed to the osteoplasia in addition to desmoplasia in these tumours. Also the infiltrative behaviour of DA may explain the characteristic feature of the tumour, the ill defined border (Philipsen *et al.*, 1992). Yazdi *et al.* have reported a marked immunoeexpression of TGF- β in areas of stromal desmoplasia but was very less in areas of follicular ameloblastoma within the same lesion. (Yazdi *et al.*, 2009) Kawai *et al.* demonstrated the presence of oxytalan fibres in the stromal tissue of the tumour which suggested that the tumour arose from the epithelial rests of Malassez in the periodontal membrane of the related tooth (Kawai *et al.*, 1999). Immunohistochemical analysis by Santos *et al.* postulated that extracellular matrix molecules like tenascin demonstrable in the stroma of follicular part of hybrid lesion and type I collagen and fibronectin seen throughout the lesion may participate in tumoral modulation of hybrid DA. (Santos *et al.*, 2006) Also the TGF- β produced DA tumour cells plays a part in prominent matrix formation. Hence the biological behaviour of DA including recurrence rate still cannot be fully appreciated due to relatively few reported cases with sufficiently long follow up periods (Philipsen *et al.*, 2001). The atypical histopathology, marked stromal desmoplasia, unusual radiographic appearance and striking difference in anatomic predilection in comparison with other solid /multicystic ameloblastomas warrant a separate consideration for this tumour in terms of treatment and followup as they comprise a unique clinicopathologic entity.

Ethical Standards- approved by the institutional ethics committee/ institutional review board.

Conflict of interest- nil

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Informed Consent- obtained

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