Case Report

International Journal of Dental and Health Sciences
Volume 01, Issue 05

DESMOPLASTIC AMELOBLASTOMA: A CASE REPORT AND REVIEW OF THE LITERATURE

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ABSTRACT:

Ameloblastoma is a true neoplasm of enamel organ type tissue which does not undergo differentiation to the point of enamel formation.The term ''ameloblastoma'' was suggested by Churchill in 1934 to replace the term 'adamantinoma' coined by Malassez in 1885.The chief histopathological variants of ameloblastoma are the follicular and plexiform types, followed by the acanthomatous and granular cell types. Uncommon variants include desmoplastic, basal cell, clear cell ameloblastoma, keratoameloblastoma and papilliferous keratoameloblastoma.When the desmoplastic type co-exists with other types, it is called as "hybrid" ameloblastoma. Desmoplastic ameloblastoma (DA) was included in the World Health Organization Classification of Head and Neck Tumors (WHO-2005) as a variant of ameloblastoma with specific clinical, image, and histological features. The purpose of this article is to report a case and to review the relevant literature, emphasizing peculiar aspects of this unusual lesion.

Key words: ameloblastoma, odontogenic tumor, desmoplastic ameloblastoma

INTRODUCTION:

Ameloblastomas are tumors arising from the odontogenic epithelium.[1,6] Despite their locally destructive nature, they are considered benign.[2]It is the second most common odontogenic neoplasm and only odontoma outnumbers it in reported frequency of occurance .Excluding odontoma, the incidence ameloblastoma is at least equal to the incidence of all the other odontogenic neoplasms combined.[3] The desmoplastic ameloblastoma is a rare and infrequent tumor that Eversole and colleagues documented initially in literature in 1984 .The knowledge regarding the clinico-radiological presentation and pathology of

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desmoplastic ameloblastomas has led to its categorization as a distinct variant of ameloblastoma in the World Health Organization classification of odontogenic tumors in 2005 .There are significant anatomic, histopathologic, and radiologic differences between desmoplastic ameloblastomas and the classic type. To provide additional clinical-radiographic-histological data about DA, this article aims to describe a case of DA which occurred in the mandible of a 25 year-old man.

CASE DETAIL:

A 32 years old male patient reported in our institution with chief complaint of bony hard swelling in right mandibular anterior region. The swelling had been present since last 5 months, growing slowly and was not associated with any bleeding, pain or any sensory change. The patient's medical history revealed nothing significant. Extra oral examination revealed diffuse swelling involving the lower jaw anterior part causing facial asymmetry on right side of the face. The intraoral examination revealed a well-defined large hard, non-tender lingual swelling of 4x3cm in anterior mandible extending from right central incisor to right second premolar, covered by normal overlying mucosa. No lymphadenopathy or fistulae were present. The involved teeth were vital, displaced slightly and exhibited no mobility.{fig.1}

A panoramic radiograph showed an ill-defined multilocular lesion with a honey comb appearance along with a large radiolucent lesion crossing the mid line extending from right second premolar to left canine region. Although the root of right lateral incisor and right central incisor were displaced by the lesion ,no apparent root resorption seen {fig.2}. Based on the radiological appearances, the diagnostic possibility of either a fibro-osseous mass or an ameloblastoma (desmoplastic variant) was suggested .An incisional biopsy was taken and all the laboratory investigation were carried out before the surgical procedure and were found within the normal limit.

Histopathologically the section showed strands, islands and a few small sheaths of odontogenic epithelium in a dense collagenous fibrous tissue stroma with stretched out kite-tail appearance {Fig.3} . Under high power follicles with typical peripheral pre-ameloblast like cells and central stellate reticulum like cells seen scattered in the connective tissue stroma{Fig.4}.A histological diagnosis of desmoplastic ameloblastoma was made with respect to the above finding

DISCUSSION:

DA has a marked predilection to occur in anterior regions of the jaw particularly the maxilla. Radiographically ,this type seldom suggests the diagnosis of ameloblastoma and usually resemble a fibro-osseous lesion because of its mixed radiolucent-radiopaque appearance. This mixed radiographic appearance is due to osseous metaplasia within the dense fibrous septa, not because the tumor itself is producing a mineralized product [7]however, Philipsen et al[16] in their report of 2 cases

of DA, remarked that some DA present osteoplasia and that this may explain the radiologic appearance of mixed radiolucency and radiopacity in some of the DAs, thereby presenting radiographic features of a fibro-osseous lesion. Philipsen et al. also remarked that DA has a tendency for de novo synthesis of extracellular fibrous protein, which has been attributed to desmoplasia seen in this lesion. Furthermore, the de novo synthesized extracellular protein could serve as nidus for calcification seen in the DA with osteoplasia. This argument again is not acceptable, as the same inductive potential is not observed in solid multilocular ameloblastoma (SMA). Waldron and El Mofty **[8]** described the histologic appearance of DA as small ovoid islands and narrow cords of odontogenic epithelium widely separated by dense, moderately cellular, fibrous, and connective tissue. Although columnar cells with reverse polarity within the epithelial islands are present, they are not the dominant feature. Spicules of mature lamellar bone trabeculae have been reported in intimate contact with the tumor, and invasion has been demonstrated. This histologic finding may indicate the potential for local invasion, and accounts for the diffuse radiographic imaging. DA may exhibit a more aggressive behavior than other types of ameloblastoma. Various facts about this lesion may suggest aggressiveness:

– A potential to grow to a large size [8]

– The common location in the maxilla that may produce an early invasion to adjacent structures;

– The diffuse radiographic appearance and the histologic finding of bone invasion [8]

Since desmoplastic ameloblastomas tend to infiltrate between bone trabeculae, curettage eventually leads to recurrences, therefore block excision is the most widely accepted form of treatment**[10]**.Even after two decades of the first report of DA, the cause for this peculiar histologic appearance is still unclear. Compared with the solid multilocular ameloblastomas (SMA),various immunohistochemical studies have reported DA tumor cells as showing variable expression of S-100 protein and desmin,**[13]** marked immunoexpression of TGF-β,**[12]** high expression of caspase-3 and Fas,13 decreased expression of CK19,[14]and high expression of p63.**[11]**.Compared to the stroma of SMA, the desmoplastic stroma of DA has been reported to show a strong positive reaction for collagen type VI (ruling out the scar tissue)[17] .Immunonegativity for tenascin, and strong immunopositive reaction for fibronectin and type 1 collagen.[18] Kishino et al,[19] demonstrated oxytalan fibres in the stromal tissue of one case, suggesting that the tumor had derived from the epithelial rest of Malassez in the periodontal membrane of a neighbouring tooth.

Desmoplasia of the stromal connective tissue in DA can be argued to be a

maturation change in a SMA, as similar dense collagenization is seen during maturation of longstanding tumors. This argument can be supported by the existence of 'hybrid' tumors, wherein the follicles were present in a desmoplastic background. But the lesser frequency of DA in the posterior mandible compared to SMAs is then unanswered. Firstly, it is probable that the location of the tumor can influence the maturity of the lesion and, hence, the tumors in the anterior jaws may mature sooner than those occurring in the posterior mandible . Many hybrid lesions may have been misclassified, since the presence of typical ameloblastic islands in some areas could have warranted a diagnosis of SMA. Ameloblastomas may thus be common in the anterior and posterior jaw and the tumors in the anterior jaw may mature early, explaining the unique site predilection, mixed radiolucent appearance, and histologic presentation of DAs.

CONCLUSION:

There is need for oral and maxillofacial surgeons to recognize the distinction in the observed biologic profile of DA and conventional ameloblastoma, especially because DA may present a confusing clinical diagnosis in view of its resemblance to a fibro-osseous lesion. However, further studies are necessary to clarify the biologic profile of DA, especially the induction of desmoplasia and occasional osteoplasia observable in this lesion.

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FIGURES:

Fig 1:- Intraoral view :-large intraoral hard Swelling in anterior mandible extending from right central incisor to right first premolar

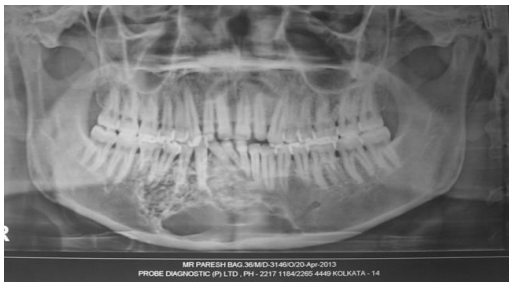

Fig 2 :- The radiographic finding :-ill defined multilocular lesion with honey comb appearance along with large radiolucent lesion crossing the midline with displace - ment of associated teeth.

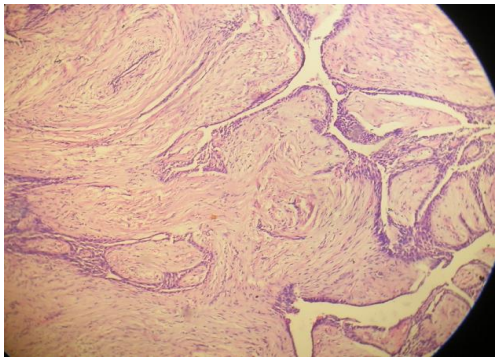

Fig 3:- island of odontogenic epithelium in dense collagenous fibrous tissue stroma with stretched out kite-tail appearance

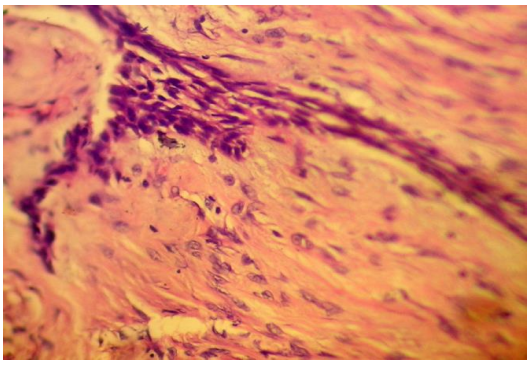

Fig 4:- High power view show peripheral ameloblast like cells and central straitum reticulum cells in fibrous stroma.