Case report

Plexiform Ameloblastoma with Dentinoid Induction: A Rare Case Report

Mishra G, Ramesh G, Seth R, Chaubey S

ABSTRACT: Ameloblastoma is regarded as homogenous group of neoplasm with locally invasive character. It generally does not show induction of dental hard tissue formation except in few cases. Dentinoameloblastoma is a rare odontogenic tumor characterized by classic ameloblast-like areas with unusual induction of dentinoid by the neoplastic odontogenic epithelium. In this article we aim to report a tumor that occurred in the right posterior mandible of an 18-yr old female patient with histopathological resemblance to plexiform ameloblastoma, showing formation of hard tissue. These changes emphasize the differentiation potential of neoplastic odontogenic epithelium and add interesting parameters to the study of tissue reactions associated with this common odontogenic tumor.

Key words: Ameloblastoma; Dentinoid; Plexiform; Odontogenic; Tumor; Enucleation.

INTRODUCTION:

Ameloblastomas are one of the most common odontogenic tumors of the maxillofacial region. Small and Waldron had pointed out that the tumor has a slow growth rate, which generally starts to develop around early childhood and young adulthood.[1] Biological behavior and histogenesis of these tumors is still unexplored as there is lack of relevant studies and long follow-up of these patients.[2] The lesion, referred to as odontoameloblastoma has been defined as "a very rare neoplasm characterized by the presence of enamel, dentin and an odontogenic epithelium resembling that of an ameloblastoma, both in structure and in behavior".[3]

The purpose of this paper is to report this rare case of dentinoameloblastoma (DA) showing evidence of induction of dentinoid by tumor cells but without concomitant formation of enamel, in young female patient involving posterior region of mandible.

CASE REPORT:

A female patient of age 18 yrs, presented with pain and swelling in mandibular right posterior region of 2 months duration. Patient had visited some local dental practitioner for swelling with pain and was under medication, but with no relief from pain. On extraoral examination, an expansile lesion over right body of mandible was evident. The lesion was tender and firm to palpation with no obvious change in color of overlying skin. Intraoral examination revealed an obvious swelling of reddish-pink in color, measuring about 5x2cm in size extending from 43 to 46 region. On palpation, the swelling was tender and hard in consistency. There was both buccal and lingual expansion. All teeth were present except 38.

Radiographs showed the presence of a large radiolucent lesion which was essentially multi-locular extending from mesial surface of first molar to the distal surface of the canine. The enucleation was done for the lesion and submitted for histopathological evaluation. The gross specimen received was a solid well encapsulated, round to oval in shape, creamish to brown in color on the surface. Grittiness was felt while sectioning the tumor.

Histopathological examination with hematoxylin and eosin stained sections revealed a well-encapsulated lesion with stroma showing interconnected odontogenic epithelial cells with tall columnar to cuboidal shaped cells with reverse polarized hyperchromatic nucleus at periphery and central stellate reticulum like cells. These tumor cells were extending into the connective

tissue stroma in thin cords and interlacing strands like pattern, interconnected with each other, giving a plexiform arrangement (fig 1).

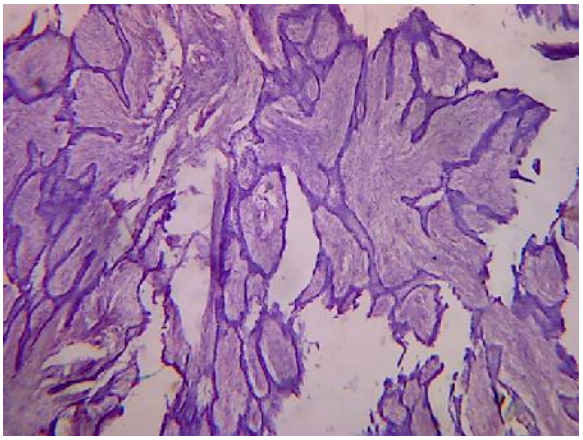

Fig 1- H&E stained section (10x magnification) shows the interconnected strands of odontogenic epithelium giving a plexiform pattern.

The odontogenic epithelium was surrounded by connective tissue stroma with dense collagen bundles and spindle shaped fibroblast. One area of the tumor tissue shows induction of dentin-like structure. Although the material appeared to be lying within the epithelial masses, careful examination of the lesion confirmed that this dentinoid material with tubular structure was a connective tissue product closely surrounded by epithelial cells (fig 2). Based on these findings the diagnosis of Dentinoameloblastoma (DA) was given.

DISCUSSION:

The age of presentation of ameloblastoma varies with changing locations according to various literature reviews, but the age range usually remains within standard accepted age group of 2nd to 7th decade.[1] The age of patient in our case was 18 years which is not common as according to Z. Chaudhary et al the overall incidence rate of ameloblastoma in age group of less than 20 years has come to 16.9%.[1]

In the exclusive pediatric and adolescent review, correlating the age with histopathological pattern, Takahashi et al reported 66% of the cases with plexiform type which is similar to case reported here.[4]

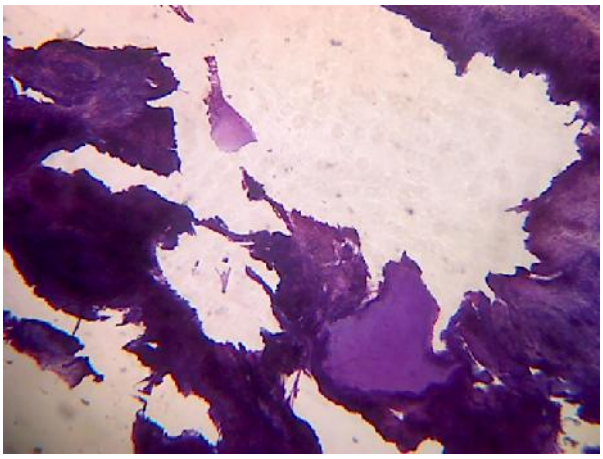

Fig 2- H&E stained section (10x magnification) shows the dentinoid induction within the stroma in close approximation with the odontogenic epithelium.

In our case the lesion was in the body of ramus which is in consistent with Chawla et al[5] report of the predilection of ameloblastoma for the posterior segment as 25.3% and posterior segment and vertical ramus as 40.6%. They mentioned that the unicystic and plexiform variants occurred at a younger age and more frequently involved the body and ramus area of the mandible.

The term dentinoameloblastome (DA) has been in use since it was first defined by the World Health Organization in 1970 and has been frequently confused with ameloblastic odontoma/odontoameloblastoma.[6] DA has been described by Slabbert *et al.*,[3] who observed intimate association of dentinoid like material with odontogenic epithelium in unicystic neoplasm containing typical follicles of ameloblastoma along with psammomatous-type dystrophic calcifications in male patient of Asian ethnic origin. However, the present case showed plexiform ameloblastoma features along with small amount of dentinoid-like material.

Orlowski *et al.*,[7] have described an odontogenic tumor that exhibited features of unicystic plexiform ameloblastoma with

dentinoid-like material but they did not term it as "odontoameloblastoma" or "dentinoameloblastoma" although dental hard tissue formation occurred.

In the cases reported by Slabbert, *et al.*,[3] Orlowski, *et al.*,[7] and in the present report, hard tissue formation was found with tumor diagnosed as ameloblastoma. Bone formation was observed in one neoplasm but in other reports the hard tissue had been interpreted as dentin or dentinoid. Interstitial ossification has been reported in two cases of polycystic ameloblastoma[8] and enamel matrix formation was described in two reports. Our present case revealed presence of dentinoid-like material.

Odontoameloblastoma has been categorized as odontogenic tumor with/without hard tissue formation in WHO 2005 classification.[9] WHO and Philipsen and Reichart[10] have defined it as a neoplasm that includes odontogenic ectomesenchyme in addition to odontogenic epithelium that resembles an ameloblastoma in both structure and behavior. Because of the presence of odontogenic ectomesenchyme, inductive changes take place leading to the formation of dentin and enamel in parts of the tumor.

Papagerakis, *et al.*,[11] demonstrated that ameloblastic epithelial cells in mixed odontogenic tumors expressed gene products normally present in ectomesenchymal cells and resulted in conversion and coexpression of mesenchymal phenotype. Dentinoameloblastoma shows dentinoid induction without concomitant enamel formation while odontoameloblastoma shows enamel and dentinoid formation in ameloblatoma.

Thus, it is probable that neoplastic epithelial cells committed to ameloblastic differentiation could produce the dentinoid which exists in some tumors. It has been seen that induction in ameloblastomas is extremely rare which suggest that attempts to classify odontogenic tumors on the basis of induction will be unsound.[2]

CONCLUSION: Dentinoameloblastoma is a rare odontogenic tumor characterized by classic ameloblastoma-like areas with unusual induction of dentinoid by the neoplastic odontogenic epithelium without any evidence of enamel matrix or tooth formation. Many of these changes emphasize the differentiation potential of neoplastic odontogenic epithelium and add interesting parameters to the study of tissue reactions associated with this common odontogenic tumor. Further case reports on dentinoameloblastomas are expected to shed a light on the biological behavior and nature of this unique tumor.

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