AN UNUSUAL EPITHELIAL ODONTOGENIC TUMOUR:
AMELOBLASTOMA WITH INDUCTION OR AN AGGRESSIVE ADENOMATOID ODONTOGENIC TUMOUR-A CASE REPORT
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ABSTRACT
Objectives: To highlight the variations in the histological presentations of odontogenic epithelial tumours especially ameloblastoma and adenomatoid odontogenic tumour; the deficits in existing classifications and proposed modifications.
Design: Case report
Setting: outpatient department of dental school
Subject: 21 year old Indian male with a swelling of palate.
Main outcome measure and results: Clinically aggressive lesion that invaded the antrum was variably diagnosed as aggressive AOT or ameloblastoma with inductive dentinoid. The lesion was treated surgically and a ten year follow-up revealed no recurrence.
Conclusion - It is important to note the variations in the clinical and histological presentations of odontogenic tumours and treatment should be guided by the clinical behaviour rather than histological nature only.
Key words: odontogenic, ameloblastoma, adenomatoid, AOT, dentinoid

INTRODUCTION
Odontogenic tumours often show the presence of matrix components resembling dentinoid, osteoid or a mixture of both. These components presumably arise from inductive interactions between epithelium and connective tissue. These interactions form the basis of the WHO classification of odontogenic tumours but cannot be regarded as rigid (Kramer et al 1992). Thus, increasingly, cases are being reported in the literature where the presence of dentinoid, osteoid, dentine or enamel is seen in so called “potentially non-inductive” epithelial odontogenic tumours like ameloblastoma (Slabbert et al 1992, Orlowski et al 1991, Scott and Wood 1989, Tajima et al 1992). As commented upon by Slabbert et al 1992, the presence of calcified structures in epithelial odontogenic tumours generates the question as to whether this group of lesions has the potential for laying down tooth matrix. If such a potential is recognised and agreed upon, then there arises a need for a new look at the classification of these lesions based on the histopatho-

Fig1: Right maxillary occlusal radiographic view showing large radiolucent lesion extending from the alveolar margins of the premolars and molars to the midline.

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logical observations and biologic potential. The purpose of this article is to present a case of an unusual epithelial odontogenic tumour with excessive stromal hyalinization that resembled dentinoid in places and features of ameloblastoma with some parts resembling an adenomatoid odontogenic tumour. The invasiveness of the tumour and the presence of clear cells further compounded the diagnostic dilemma.

CASE REPORT
A 21 year old Indian male presented in Oct 1980 to the Department of Oral and Maxillofacial Surgery of the Government Dental College and Hospital, Bombay with an unilateral, rapidly enlarging palatal swelling and pain involving the maxillary right first molar of 4 months duration. Extraoral examination revealed the swelling extending up to the infraorbital ridge and zygomatic arch on the right side. Intraorally the palatal swelling was 4 cm x 3 cm in size extending from teeth 14-17 region to the midline, bony hard in consistency, and with mobility of teeth 15,16 and 17. The right maxillary third molar was clinically missing.

Intraoral radiographs maxillary occlusal view (Fig. 1) and periapical (Fig. 2) revealed a radiolucent lesion extending from 14-17 region to the midline with root resorption of the first molar. A PA Waters projection showed haziness of the right maxillary antrum. Based on the clinical and radiological features, a provisional diagnosis of ameloblastoma was entertained.

An incisional biopsy revealed features of an ameloblastoma. A subtotal maxillectomy on the right side was performed. On raising the flap and gaining access to the antrum the tumour mass was seen to extend throughout the antrum but no resorption of the infraorbital ridge was noted. The right maxillary segment was resected in toto along with the mass and sent for histopathological examination. The surgical wound was closed and post operative healing and recovery were uneventful. The patient was discharged after 10 days but advised to report to the department for periodic follow-ups.
Histopathological examination of the hematoxylin and eosin stained sections revealed a predominantly epithelial tumour composed of columnar cells with hyperchromatic nuclei arranged in a peripheral palisading manner resembling ameloblasts. These enclosed areas of stellate cells, which sometimes exhibited a whorled arrangement (Fig. 3). In other places, an adenoid pattern was discernible with a central lumen lined by two layers of cuboidal cells with nuclei polarised away from the basement membrane (Fig. 3). These adenoid structures contained eosinophilic material that stained positive with Periodic acid Schiff. A few of the whorled epithelial structures showed clear cells (clear cytoplasm and a small nucleus) (Fig. 4). The stroma showed prominent hyalinization with areas of an eosinophilic extracellular homogenous material interpreted as “dentinoid” especially in relation to the epithelial cells (Fig. 3). This material stained positively for collagen (van Gieson and Masson’s trichrome) and negatively for amyloid (Congo red). Focal calcifications of basophilic nature were distributed sparsely in the stroma. The lesion was non-encapsulated and invasion of muscle was observed; margins of the resected specimen were however free of tumour tissue.

The histopathological features were suggestive of an ameloblastoma with excessive induction or an adenomatoid odontogenic tumour that behaved aggressively. The histopathological slides were circulated to a total of six oral and three general histopathologists and varying opinions were encountered. There was an equivocal opinion between the above stated diagnoses.

An immunohistochemical assay of the keratin profile of the lesion including Type IV collagen and Laminin using standardised avidin-biotin-complex techniques was carried out. In all the immunohistochemical procedures, antigen retrieval was carried out by microwaving the sections at 900C for 10 min in citrate buffer at pH 6.2. An electron microscopic evaluation of the paraffin embedded tissue was also done.

Monoclonal antibodies (MAbs) against simple keratins K8 & 18 (CAM 5.2, Beckton & Dickinson, Oxford, UK), two antibodies against K18 ( RCK 106, EuroPath Ltd., Stratton, UK., and Novostra, UK), K19 (Novo-castra, UK) were utilised. K8 and K19 (CAM 5.2) were focally and moderately expressed. K19 was expressed with a greater intensity, predominantly in the peripheral layers of
the islands of epithelial cells (Fig. 5). No expression of K18 could be demonstrated.

Mabs against Type IV collagen and laminin (Sigma Chemicals, Poole, Dorset, UK) were also utilised and a strong expression of both were seen in the lesion. Type IV collagen was prominently demonstrated in the basement membranes of the luminal aspect of the adenoid structures while the cosinophilic structures seen inside the lumen strongly expressed laminin. A variable expression was seen in the blood vessels of the stroma. The hyalinised areas and the dentinoid structures were negative for the antibodies.

For electron microscopy the tissues, recovered from paraffin, were post fixed in osmium tetroxide and processed according to standard protocols. Uranyl acetate and lead citrate was used for staining the grids. The sections were examined using an AEI 6B transmission electron microscope. The fixation was unsatisfactory and epithelial cell architecture was poorly defined. The collagen fibres of the lesion were however adequately preserved and were found to be predominantly 45nm in diameter, exhibiting an interlacing pattern and showed a regular periodicity of 64-68 nm. The dentinoid material consisted predominantly of these fibrils arranged irregularly (Fig 6). In between these fibrils an homogenous, structureless ground substance was present. These areas were very often surrounded by epithelial cells. Focal calcifications represented as dense dark bodies were also seen. The nature of these structures could not be ascertained and probably represented dystrophic calcifications.

The patient was followed up regularly for the first ten years post operatively and no recurrence was observed. He was subsequently lost to follow up.

DISCUSSION

The diagnostic confusion associated with this case was based on two counts. First, the presentation of the epithelial component resembled both an ameloblastoma in some areas and an adenomatoid odontogenic tumour in other parts. If it is to be accepted that the case represents an ameloblastoma then the presence of the adenoid pattern and the inductive stromal changes is perplexing. In recent literature, similar cases of an ameloblastoma with stromal induction interpreted as dentinoid have been labelled as dentinoameloblastoma (Slabbert et al 1992). In their discussion on the case of dentinoameloblastoma, Slabbert et al (1992) postulate that induction, though a rare occurrence in ameloblastomas, is nevertheless real. Distinction between these cases of dentinoameloblastoma and the accepted entity of odontoameloblastoma would be based on the absence or presence of enamel as well as of dentine or “dentinoid”. The presence of enamel formation concomitant with that of dentine (Boyle and Kalnins 1960) or independent of the presence of dentine has also been reported (Fleming, 1952). Such histopathological oddities underlie the confusions that prevail contrary to currently held concepts on tooth embryogenesis; that enamel formation requires the presence of dentine and the latter is a result of interaction of odontogenic epithelium with ectomesenchyme. The biological behaviour of this lesion and its invasive potential justified the diagnosis of an ameloblastoma.

The presence of clear cells in ameloblastomas has been regularly reported. These cells contain abundant glycogen and ultrastructurally may exhibit prominent vacuolization and a paucity of cyttoplasmic organelles. The WHO has classified odontogenic lesions containing a predominance of clear cells as clear cell odontogenic tumour which is stated to be more locally aggressive than ameloblastoma (Kramer et al 1992).

These areas in the tumour which resembled an AOT were rather prominent and exhibited more of a solid (whorling) pattern than the adenoid pattern. This diagnosis cannot be reconciled with the clinical aggressiveness of the tumour and the presence of clear cells, which to the best of our knowledge have not been previously reported in an AOT. If the diagnosis of an AOT was to be accepted then
the unusual aggressiveness could be explained on the basis of excessive cellularity, paucity of adenoid elements and presence of clear cells.

In an excellent analysis of a large collection of adenomatoid odontogenic tumours, Phillipsen et al like the classical ameloblastomas, although these lesions are classified by WHO as epithelial odontogenic tumours without inductive changes. It is conceivable that every epithelial odontogenic tumour has the potential for induction, although this is normally not expressed in ameloblastomas. Thus two subsets of ameloblastomas could arise if the concept of induction as the basis for classification of odontogenic tumours is accepted. Firstly the interaction of the outer enamel epithelium with that of the dental follicle may give rise to epithelial tumours including ameloblastomas where induction is not a phenomenon. Secondly the interaction of the inner enamel epithelium with that of the dental papilla may result in ameloblastomas showing inductive changes as seen in this case.

The keratin profiles of this lesion were interesting. Despite the use of different antigen retrieval techniques keratin 18 was not expressed and keratins 8 & 18 (CAM 5.2) and 19 were moderately and focally expressed. K18 was found to be expressed in seven out of nine ameloblastomas studied by Heikinheimo (1993). This is in contrast with the findings in our laboratory where K18 could not be demonstrated in a series of 10 ameloblastomas, but were interestingly expressed in two out of four AOTs evaluated (unpublished data).

The interesting features of this case illustrate the complexities in the presentations and the diagnosis of odontogenic tumours. We propose that this case be labelled as an ameloblastoma with induction resembling dentinoid (Dentino-ameloblastoma). In view of the increasing numbers of similar cases being reported in the literature, it is suggested that a review of the classification of odontogenic tumours be attempted to include similar cases.

REFERENCES

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