"Hybrid" ameloblastoma: a report of two cases

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Abstract
Ameloblastoma is the most common odontogenic tumour. The tumour has been described as a benign but locally invasive polymorphic neoplasm. Hybrid lesions have been described, which combine histological features of desmoplastic and conventional ameloblastoma. The hybrid ameloblastoma is rare and only few cases have been reported worldwide. We present two cases seen from a review of 195 ameloblastoma cases seen over a ten year period. The cases presented with clinical features of the conventional ameloblastoma such as bucco-lingual bone expansion and multilocular radiololucency. Larger clinical series of hybrid ameloblastoma need to be reviewed in order to better characterize the clinical behaviour, aggressiveness and prognosis of this rare variant of ameloblastoma.

Keywords: *Hybrid ameloblastoma; Ibadan; oral pathology*

Résumé
L'ameloblastome est une tumeur odontogenique le plus commun. La tumeur a été décrit comme bénigne mais localement un néoplasme polymorphique invasive. Les lésions hybrides ont été décrites, laquelle combine les caractéristiques du demoplastique et de l'ameloblastome conventionnelle. L'hybride d'ameloblastome est rare et seulement sauf quelques cas ont été rapports dans le monde entière. Nous présentons deux cas diagnostiqués d'une revue de 195 cas d'ameloblastome vus pendant 10 ans. Les cas présentaient des signes cliniques de l'ameloblastome conventionnelle telle que l'expansion de l'os bucco-lingual et de radiololucence multiloculaire. Un nombre important de cas clinique d'hybride d'ameloblastome doit être revu afin de mieux caractériser le comportement Clinique, agressivité et le pronostic de ce cas rare d'ameloblastome.

Introduction

Ameloblastoma is the most common odontogenic tumour. It is benign but locally invasive. It is polymorphic [1] in nature with several histological variants described such as follicular, plexiform, acanthomatous, granular cell, basal cell and desmoplastic [2]. The various histomorphological patterns do not appear to significantly affect the biological behaviour or prognosis of these tumours, with the possible exceptions of unicystic and desmoplastic types [3].

Hybrid lesions combining histological features of both desmoplastic and conventional ameloblastoma variants were first documented by Waldron and El-Mofty in 1987 [4]. The hybrid variant of ameloblastoma is rare, with only nine cases reported as at 2001. The desmoplastic ameloblastoma was initially described by Eversole *et al* [3] in 1984. This variant often occurs in the anterior maxilla and shows unique radiographic features that appear as a mixed radiolucent-radiopaque lesion similar to a benign fibro osseous lesion [4,5]. Histologically, it presents as small nests and strands of odontogenic epithelium, which are surrounded or compressed by a dense collagenous stroma [6,7,8].

We present a report of two cases of hybrid ameloblastoma from a review of cases of ameloblastoma seen at the University College Hospital Ibadan with the view of adding to the scarce literature on hybrid ameloblastoma.

Case description

Case 1

A 50 year old woman was referred to the University College Hospital Ibadan with a two year history of a slow growing, painless anterior mandibular swelling. Examination revealed a swelling extending from (36) to (46) with bucco-lingual bone expansion and obliteration of the floor of the mouth. The lesion was bony hard with an isolated eggs hell cracking consistency felt on palpation. There was associated poor oral hygiene and dental anarchy. Medical history was not contributory. Radiographic examination showed a multilocular radiolucency extending from the lower first molars on both sides. A clinical impression of ameloblastoma was made.

Histological examination showed the conventional ameloblastoma component comprised of interconnecting epithelial strands giving the

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plexiform pattern of ameloblastoma (fig 1a). Fig 1b shows high collagenisation and desmoplasia of the stroma and tumour nests compressed by the desmoplastic stroma resulting in narrow strands and cords that lack peripheral palisading of columnar cells but are rimmed by cuboidal or flattened cells. The border between the desmoplastic and plexiform areas was relatively indistinct (fig 1a).

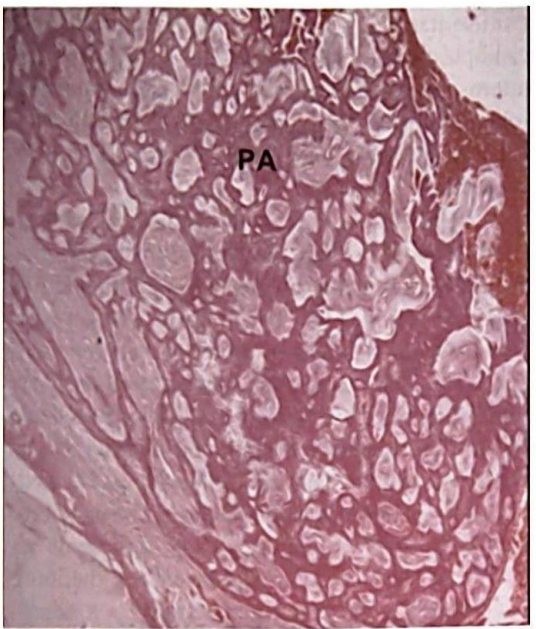

Fig. 1a: Photomicrograph showing anastomosing cords of epithelial cells consistent with plexiform ameloblastoma (PA) (H&E X40)

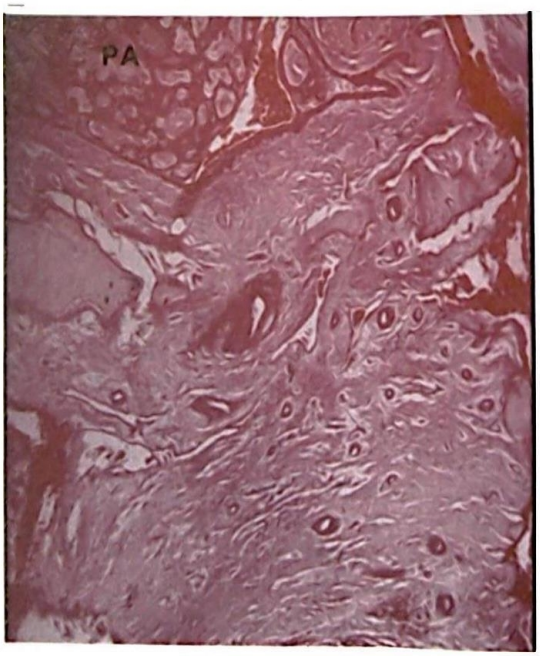

Fig.1b: Photomicrograph showing prominent stromal desmoplasia in another area of the tumour.

Case 2

A 29-year-old man presented with a one and a half year history of slow growing mandibular swelling. Examination revealed a mandibular swelling extending from the (33) to (47). There was associated bucco-lingual bone expansion. The 43, 44 and 45 were mobile and the 31 and 41 were labially displaced. Medical history was not contributory. Plain radiographs showed a multilocular radiolucency extending from 37 to 42 with thinning of cortex and root resorption of 36, 35 and 34. A clinical impression of ameloblastoma was made and a biopsy was taken. Histologically it also showed dense connective tissue stroma and the conventional ameloblastoma portions showed acanthomathous differentiation with squamous metaplasia of the stellate reticulum-like central areas and foci of keratinization, similar to Case 1.

Discussion

Waldron and El-Mofty [4] reported that hybrid ameloblastoma accounted for 4.3% of all ameloblastomas, while Higuchi *et al* [10] reported a figure of 3.4%. In this study, we found 2 hybrid ameloblastoma cases out of a total of 195 cases seen over a period of 10 years. This representing a relative frequency of 1.5% and is similar to the 1.1% reported from a study in Japan [9].

Both of our cases were in the anterior mandible not extending posterior to the first molar teeth. Takashi *et al* [9] reported that five of their reported nine cases were located in the posterior mandible and three in anterior to posterior areas of mandible. No information on anatomical site was available in one of the cases [9]. Waldron and El-Mofty [4] reported that hybrid lesions had a predilection for the posterior portion of the mandible, although neither radiographic nor clinical details were available in their cases. However, most authors agree that the desmoplastic variant had predilection for anterior part of the jaws [4,5,9] with Takashi *et al* [9] reporting that 70% of their cases occurred in the anterior/premolar regions of the jaw and about half of the cases were located in the maxilla. Although no definite explanation has been given for desmoplastic ameloblastoma having a predilection for anterior jaws, Higuchi *et al [10]* suggested that ameloblastoma in the tooth-bearing area has a tendency to have an abundant stroma and to be desmoplastic. Many cases of desmoplastic ameloblastoma are said to be associated with prominent osteoplasia, which explained the mixed radiopaque/radiolucent appearance in these tumours [7,11], however, most cases of hybrid ameloblastoma presented as multilocular radiolucency similar to those

seen in conventional ameloblastoma [9]. This is consistent with our findings.

The correlation between desmoplastic ameloblastoma and the conventional ameloblastoma and the reason why they sometimes coexist have not been clearly established. It is possible that desmoplastic changes occur in the stroma of pre-existing conventional ameloblastomas or areas in a primary desmoplastic ameloblastoma transform to one of the conventional types. Another possibility is that the Hybrid ameloblastoma is actually a collision tumour [8] where the conventional and desmoplastic ameloblastoma variants develop simultaneously.
The radiological and histological appearance of the hybrid ameloblastoma suggests that it is worthy of separate study. However, more cases need to be examined to ascertain its clinical behaviour and prognosis *vis-a-vis* other variants of ameloblastoma.

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