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Hybrid Lesion of Ameloblastoma and Adenomatoid Odontogenic Tumour (AOT): Report of Two Cases from a Tertiary Referral Hospital in Sub-Saharan Africa

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Authors' contributions

This work was carried out in collaboration among all authors. Authors OMA, OAF and ROB designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Author FAA managed the analyses of the study and the literature searches. All authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Introduction: Hybrid lesions are lesions showing the combined histopathological characteristics of two or more previously recognized odontogenic tumours and /cysts of different categories. Hybrid lesions do exist because of close interrelationship of several odontogenic lesions and also because odontogenic tumors and cysts can arise at any stage of odontogenesis. The objective of this study was to present 2 cases of hybrid odontogenic tumour that is composed of adenomatoid odontogenic tumour (AOT) and ameloblastoma.
Case Reports:
Case 1: A 33year old female patient with a bucco-lingual swelling in the left mandibular premolar-molar-ramus regions of 13years duration. The lesion measured about 15x5x3cm, it is non tender. Surgical specimen revealed hybrid lesion of granular cell type ameloblastoma and AOT.

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Case 2: An 18year old female patient with a painless right mandibular molar-ramus swelling of 10years duration. Mandibulectomy specimen measured about 11 cm x 8.5 cm x 5 cm and was found to be hybrid lesion of acanthomatous ameloblastoma and AOT.
Conclusion: Both cases had a very long clinical duration and showed more buccal bone expansion with barely noticeable lingual bone expansion. With such clinical scenarios, a suspicion of hybrid tumour should be made.

Keywords: Hybrid tumour; ameloblastoma; odontogenic.

1. INTRODUCTION

Odontogenic tumours and cysts present with a high degree of diversity because of the complexity and prolonged period of odontogenesis [1-6]. The first internationally agreed classification of this diverse lesions were done by WHO in 1971. [6] Further re-classification has emerged to accommodate other evolving entities in the recent 2017 WHO classification of odontogenic cysts and tumours. [7] However, there have been unique lesions that shows histological patterns of two or more distinct previously recognized lesions by the WHO classification. These lesions further compound the diversity of odontogenic tumours and make diagnosis cumbersome [1]. This group of tumours have been termed hybrid tumours and have been defined as lesions showing the combined histopathological characteristics of two or more previously recognized odontogenic tumours and/or cysts of different categories [1]. Different combinations of hybrid tumours have been reported in the literature, however non has reported histological subtypes of ameloblastoma with other epithelial tumours. We report 2 cases of hybrid lesions of granular cell ameloblastoma and adenomatoid odontogenic tumour (AOT) & acanthomatous ameloblastoma and AOT.

2. CASE REPORTS

2.1 Case 1

A 33year old female presented with 13years history of painless, progressive left lower jaw swelling which she first noticed as a swelling in relation to the last tooth in the lower left quadrant with an estimated size of a peanut. She had incisional biopsy done and thereafter was lost to follow up. She presented again with the same swelling which had increased progressively to its present size. Between presentation and time of operation, she gave history of 2 episodes of rapid increase in size of the swelling, with associated pain, but no discharge and no history paraesthesia. There was no history of similar swelling in other part of patient's body.

Extra oral examination revealed discrete left lower jaw swelling measuring 10 x 5 x 3 cm, extending from 2 cm of the left commissure of the mouth to the left angle of the mandible. Skin overlying the swelling was intact and with the same colour of the surrounding skin. Swelling was not tender and of mixed consistencies ranging from bony hard, ping pong and egg cracking sensations with expansion of the inferior border of the mandible on the left (Fig. 1a). Intra-orally, there was no limitation in mouth opening but there was indentation of the teeth 26,27,28 on the lower mucosa which was not erythematous. There was marked buccal and mild lingual expansion of cortical plates extending from 34 to the ascending ramus of the mandible with obliteration of buccal sulcus (Fig. 1b). The swelling was not tender and also had mixed consistency of bony hard, ping pong and egg cracking sensations.

A provisional diagnosis of Ameloblastoma of the mandible was made with odontogenic myxoma, calcifying epithelial odontogenic tumour, dentigerous cyst, keratocysticodontogenic tumour and central giant cell granuloma as differentials.

Plain radiographs showed a multilocular radiolucency extending from the 34 to the condyle with a distomolar (Fig. 1c). Loculation of varying sizes with septae of different thicknesses were seen within the tumour. Following aspiration of straw coloured fluid, incisional biopsy and mandibulectomy, a histological diagnosis of hybrid lesion of granular cell ameloblastoma and AOT was made (Fig. 2a and 2b).

2.2 Case 2

An 18year old female patient presented with 2years history of painless, progressive left lower jaw swelling which he first noticed as a swelling in relation to the last tooth in the lower left

quadrant with size barely noticeable. She had incisional biopsy done and thereafter was been followed up. She represented with the same swelling which had increased progressively to its present size. Between her last clinic visit and present, she gave history of rapid increase in size of the swelling, with associated pain, however no history of any form of discharge and no history paraesthesia. There was no history of swelling in other part of patient's body.

Extra oral examination revealed discrete right lower jaw swelling measuring 8 x 5 x 5 cm, extending from the left commissure of the mouth to mandibular region angle-ramus posteriorly (Fig. 3a). Skin overlying the swelling was intact and of same colour with the surrounding skin. Swelling was not tender and of mixed consistencies ranging from bony hard, ping pong and egg cracking sensations with expansion of the inferior border of the mandible on the left. Intra-orally, there was no limitation in mouth opening but there was marked buccal and mild lingual expansion of cortical plates extending from 44 to the ascending ramus of the mandible with obliteration of buccal sulcus. The swelling was not tender, and also had bony hard consistency. The occlusion was deranged.

A provisional diagnosis of ameloblastoma of the mandible was made with dentigerous cyst, keratocystic odontogenic tumour and central giant cell granuloma as differential diagnosis.

Plain radiograph showed multilocular radiolucency in the region of 44-48 (Fig. 3b). Following incisional biopsy and mandibulectomy (Fig. 3c), a histological diagnosis of hybrid lesion of acanthomatous ameloblastoma and AOT was made in the two specimens (Fig. 4a and 4b).

3. DISCUSSION

Hybrid odontogenic tumors including two or more different histologic types have been documented, but their occurrences are very rare. [8-12] Other authors have referred to this lesions as anomalous histo-differentiation and/or morpho-differentiation process rather than hybrid lesions. [1,7].

There have been 3 reported cases of hybrid ameloblastoma and AOT, all of which presented unicystic types as the ameloblastoma components. [13] Yamazaki et al. [13] in 2013 reported a hybrid lesion of AOT and solid ameloblastoma. Although, he was first to report a solid component of the ameloblastous component which comprised of both plexiform and follicular patterns, no histological variant of the ameloblastous component have been reported. We report a granular cell variant of ameloblastoma co-existing with AOT in 33year old Nigerian female and acanthomatous variant of ameloblastoma co-existing with AOT in an 18year old Nigerian female.

Radiographically, ameloblastoma appears as either unilocular for unicystic or multilocular for solid/multicystic types. The periphery of the lesion may be smooth or scalloped. With growth and expansion of the tumor, there may be merging and fusion of the compartments and, as a result, there may be transformation from a multilocular to a unilocular cystic space [14,15].

On the other hand, AOT appears radiographically as a well-circumscribed unilocular radiolucency or mixed radiopaque-radiolucent with well-defined corticated or sclerotic border, usually surrounding an unerupted tooth. They may include multiple tiny variable-shaped calcifications or radiopaque foci, which may appear like a 'cluster of small pebbles'. These calcified deposits are seen in approximately 78% of the lesions. [16,17] However, such calcifications may be seen radiologically as we observed in our cases. [18]

In our series, although there was multilocular appearance, we observed single large locule which was associated with an unerupted tooth a distomolar. This large locule may be cystic degeneration or fusion of compartments as earlier stated. The follicular type of AOT, which is about 70% of AOT is associated with an unerupted teeth [19,20] and we opined that it might be a possibility in our series, however, the extra-follicular type which is not associated with tooth [18,19] could also be a possibility. The behaviour of these individual tumours in a hybrid pattern needs further investigation.

Histological examination after incisional biopsy and tumour resection of Case 1 revealed a hybrid odontogenic tumour, showing the histopathological characteristics of granular cell ameloblastoma and AOT in about equal proportions. Cystic cavities were lined peripherally by ameloblast-like columnar cells which were palisaded and had hyperchromatic nuclei and demonstrating reverse polarity and with vacuolization of the cytoplasm within a fibrous stroma. Blue-based arrow shows

neoplastic epithelial island comprised of peripheral tall columnar cells and central stellate reticulum-like cells. Red-based arrow shows central cells that ballooned out and become eosinophilic and granular (Fig. 2a). Also seen are neoplastic ameloblastous island (Blue lines) and sheets of spindle and polygonal cells with dark eosinophilic nuclei (Red lines) with ductal structures interspersed (Fig. 2b).

Histological examination after incisional biopsy and tumour resection in Case 2 revealed a hybrid odontogenic tumour, showing the histopathological characteristics of follicular pattern of acanthomatous ameloblastoma and AOT in about equal proportions. Cystic degeneration in the stellate reticulum-like cells was observed (black pointer) (Fig. 4a). Also seen are areas of squamous metaplasia (green arrow). A depicts a duct which patent while B depicts sheets of spindle or polygonal cells (AOT component) (Fig. 4a). Arrow with red square base shows a ductal component of the AOT (Fig. 4b).

The similarity between the two cases showed a very long clinical duration of between 2-13 years with both having more buccal plate expansion with barely noticeable lingual plate expansion. We opined that such clinical scenarios may suggest a hybrid lesion.

Fig. 1a. Front view of case 1 patient showing the left mandibular swelling

Fig. 1b. Intraoral view showing more of buccal plate expansion than lingual plate (black arrow)

Fig. 1c. Posterior anterior view of the jaws showing the multilocular radiolucency

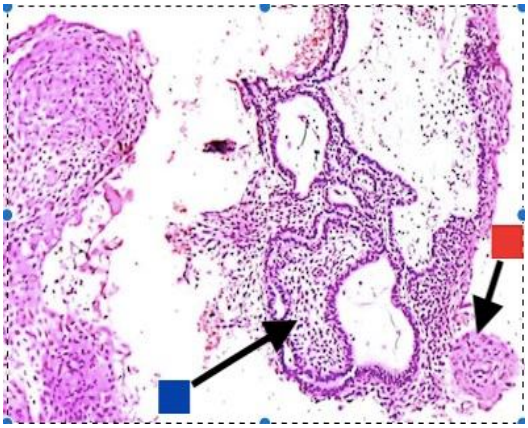

Fig. 2a. Blue-based arrow shows neoplastic epithelial island comprised of peripheral tall columnar cells and central stellate reticulum-like cells. Red-based arrow shows central cells that ballooned out and become eosinophilic and granular (H and E) (Magnification x100)

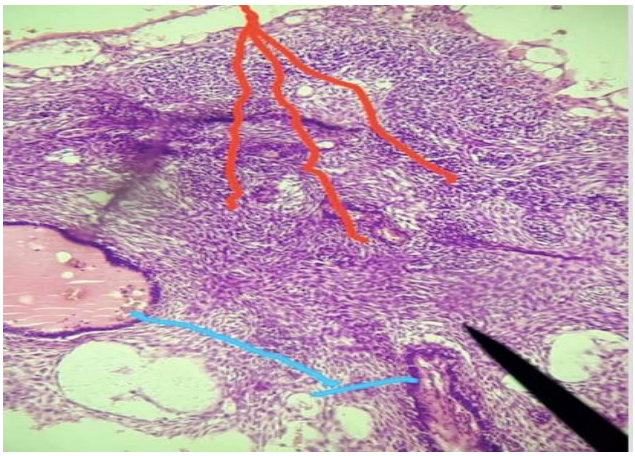

Fig. 2b. Neoplastic ameloblastous island (Blue lines) and sheets of spindle and polygonal cells with dark eosinophilic nuclei (Red lines) with ductal structures interspersed (H and E) (Magnification x100)

Fig. 3a. Clinical photograph of case 2 with right lower jaw swelling

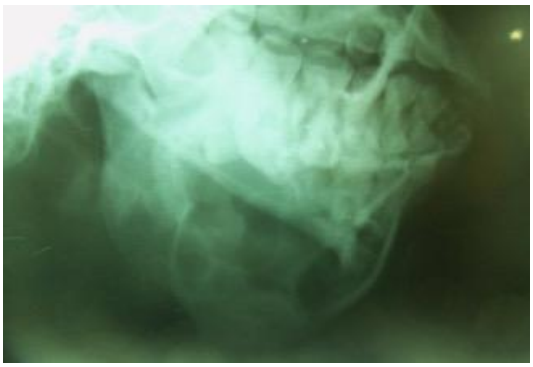

Fig. 3b. Right oblique lateral view of case 2 showing multilocular radiolucency

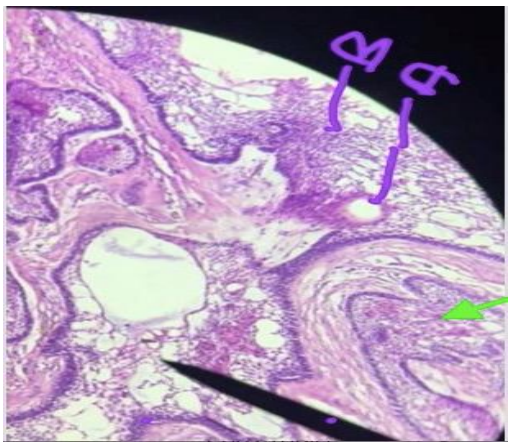

Fig. 4a. Photomicrograph showing cystic degeneration in the stellate reticulum-like cells (black pointer). Also seen are areas of squamous metaplasia (green arrow). A depicts a duct which patent while B depicts sheets of spindle or polygonal cells (AOT component) (H and E) (Magnification x100)

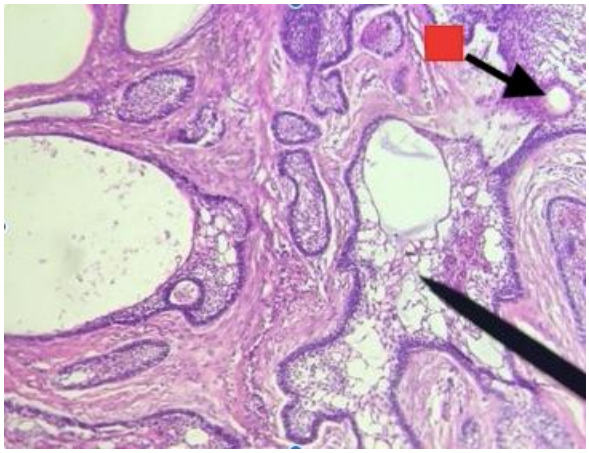

Fig. 4b. Arrow with red square base shows a ductal component of the AOT (H and E) (Magnification x100)

The biological mechanism causing hybrid lesions is still not well known till now. Possible histogenesis that has been documented in literature which includes;

1. **Collision theory:** Collision tumors are lesions that originate in different regions but coalesce in a particular area. They are composed of two different types of neoplasms derived from clearly different origins of two neoplastic clones that have arisen from different cell types in close proximity to each other. [21]
2. **Transformation theory:** This theory suggests that there is transformation from one lesion to another. [10]
3. The Combination theory suggests that both components are derived from a single stem cell that undergoes a divergent differentiation early in the evolution of the tumour.
4. The Conversion Theory suggests the derivation of one tumoural element from another e.g sarcoma deriving from carcinoma during the evolution of the tumour.
5. The Composition Theory suggests that one component arises as a result of stromal reaction to the presence of the other.

In this report the possibility of collision theory as a possible histogenesis was ruled out because the AOT co-existing with the granular cell and acanthomatous variants of ameloblastoma showed equal proportions that was intermingled with each other. We were therefore left to speculate that this lesion might have developed by transformation from one lesion to another. More cases of these hybrid lesions needs to be reported to know their true biologic nature.

4. CONCLUSION

Hybrid lesions do exist because of close interrelationship of several odontogenic lesions and also because odontogenic tumors and cysts can arise at any stage of odontogenesis. Similarly, hybrid appearances of odontogenic tumors illustrate not only the differentiation potential of odontogenic epithelium and ecto-mesemchyme, but also their complex inductive interaction.

CONSENT

As per international standard, patient's consent has been collected and preserved by the authors.

ETHICAL APPROVAL

As per international standard, written ethical approval has been collected and preserved by the author(s).

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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