Desmoplastic ameloblastoma: analysis of 17 Nigerian cases

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Objective. This study aimed to add to existing knowledge on 90 cases of desmoplastic ameloblastoma (DA) previously reported in the scientific literature and analyze data that could help speculate on its biologic profile.
Study design. From 330 cases of ameloblastoma (pooled from 573 histologically diagnosed odontogenic tumours) 17 cases of DA were retrieved and analyzed for estimated mean growth rate (EMGR) and histologic variants. EMGR for DA was compared with EMGR for conventional ameloblastoma (CA), as recorded over the same period of 38 years.
Results. Desmoplastic ameloblastoma had predilection for mandible (81.2%), posterior mandible being the most commonly affected, contrary to scientific literature reports of anterior maxillary predilection. Simple DA (88.0%) and DA with osteoplasia (12.0%) were the histologic variants observed. EMGR for DA (0.36 ± 0.44 cm/mo) was significantly less than EMGR (0.71 ± 1.16 cm/mo) for CA ($P = .000480$).
Conclusion. This study speculates that DA tends to be less biologically aggressive than CA and has predilection for posterior mandible in Nigerians. **(Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011;111:e27-e31)**

Ameloblastoma may be described as a family of diseases that possess diverse biologic behavior. It has over time been regarded as a benign tumor that may be locally aggressive, owing to its potential for invasion of surrounding soft tissue and bone.[1-10] Not all ameloblastomas behave this aggressively and it is therefore important to distinguish between clinical types of ameloblastoma to deliver qualitative treatment to patients. The following 3 main clinical types, based on clinical behavior, have been identified in the scientific literature: 1) conventional ameloblastoma, which is usually radiographically multilocular; 2) unicystic ameloblastoma, which is radiographically unilocular and commonly related to impacted teeth; and 3) peripheral ameloblastoma, which is basically a soft tissue tumor present on the gingiva and clinically simulates a reactive soft tissue swelling.[1,4,5] Conventional ameloblastoma has at least 6 subtypes, of which desmoplastic ameloblastoma (DA) is one. DA has been observed with a tendency for surrounding bone penetration.[1,3,4,6,11-15] Investigators have observed that recurrence in cases with DA is almost as high as with the conventional ameloblastoma.[12,14] Radical therapy therefore, has been indicated for the treatment of DA.[12-14] Eversole et al.[16] first described DA as a lesion with unique histologic pattern characterized by presence of islands of ameloblastic columnar cells surrounding spindle-shaped stellate reticulum–like cells in a stroma with marked desmoplasia. Waldron and el-Mofty,[17] in a later study, reviewed 116 cases of ameloblastoma, 14 of which were reported as DA. Perusal of the scientific literature shows that DA is a rare variant of ameloblastoma, with 145 cases reported in the scientific literature[13-24] before the present study. In spite of the fact that 145 cases of DA had been reported so far in the scientific literature, the true biologic profile of DA is still not well understood. The aim of the present study was to add new cases of DA to the scientific literature and attempt to further speculate on the possible biologic profile of the tumour.

MATERIALS AND METHODS

Biopsy records of 573 histologically diagnosed cases of odontogenic tumours during the period from January 1969 to December 2007 (38 years) were retrieved from the files of the Oral Biopsy services of the Department of Oral Biology and Oral Pathology, Lagos University Teaching Hospital, Lagos, Nigeria. Hematoxylin and eosin (HE)–stained sections of the 573 cases were retrieved and reviewed to confirm the diagnosis of specific odontogenic tumors. After the review, a total of

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330 cases confirmed as ameloblastoma were categorized into the following 4 subtypes, based on the presenting clinicohistologic features: 1) conventional ameloblastoma; 2) unicystic ameloblastoma; 3) peripheral ameloblastoma; or 4) desmoplastic ameloblastoma. HE-stained glass slides of all cases histologically diagnosed as DA were retrieved for further evaluation. The following clinicohistologic data on each case of DA were pooled: age, gender, size (largest diameter at hospital presentation), site of lesion, duration, histopathologic characteristics, and radiographic presentation. The pooled data were subsequently analyzed statistically using Epi-Info 6 and SPSS. Histologically, the DA cases were grouped into the following 2 variants: 1) simple desmoplastic variant; and 2) desmoplastic with osteoplasia variant. The simple desmoplastic variant showed histologic features of extensive stromal desmoplasia, whereas the desmoplastic with osteoplasia variant showed histologic characteristics that exhibit features of the simple desmoplastic variant and additional calcific structures.

The biologic profile of DA in this study was speculated based on estimated tumor growth rate (ETGR) of the lesion, which was computed from the largest diameter of the tumor and the duration of the tumor at presentation (tumor growth rate = largest diameter at presentation/duration of tumor [in months] at presentation). Estimated mean growth rate for DA was subsequently computed and compared statistically with estimated mean growth rate for non-DA ameloblastomas, which were pooled in the present series.

RESULTS

During the period of 1969-2007, 573 odontogenic tumors were histologically diagnosed and recorded in the Biopsy Services of the Department of Oral Pathology of the Lagos University Teaching Hospital. The most frequently occurring odontogenic tumour was ameloblastoma (57.59%, 330 cases). Among the 330 cases of ameloblastoma observed in the series, 299 were conventional, 19 unicystic, and 3 peripheral. A total of 17 conventional types were DA (Table I).

Age range in cases with DA was 17-73 years (mean 38.3 ± 18.197, mode 22.00, median 32.00) with peak incidence between the third and fifth decades (Table II). DA was observed to occur slightly more in men (56%) than in women (44%), with a male:female ratio of 1.27:1. A total of 88.0% of the lesion were the simple desmoplastic variant (Fig. 1), and 12.0% presented with osteoplasia (Fig. 2).

Record of size based on the estimated largest diameter was recorded in only 14 cases of DA. The rate of growth of DA based on the estimated largest diameter per month varied from 0.18 1.67 cm (mean 0.3653 ± 0.4417). The estimated monthly tumor growth rate observed was statistically significantly higher in the conventional type (0.7080 ± 1.1645 cm/mo) compared with DA (0.3653 ± 0.4417 cm/mo); variance between samples = 9.65; residual variance = 0.78; F statistic = 12.44; P = .000480; Table III).

The majority of DA (82.4%) presented as radiolucent lesions on radiograph, and 3 cases (17.6%), which included 1 case of simple DA and 2 cases of DA with osteoplasia, presented as a combination of radiolucency and radiopacity. A total of 81.3% of DA had a mandibular site predilection, with the posterior mandible being the most commonly affected site (56.3%) compared with the anterior mandible (Fig. 3). Estimated duration of growth at presentation ranged from ~3 to 192 months.

Table I. Frequency distribution of types of ameloblastoma in the series

Malignant ameloblastoma	*n*	*%*
Follicular	199	60.30
Plexiform	83	25.15
Desmoplastic	17	5.15
Peripheral	3	0.91
Unicystic	19	5.76
Malignant ameloblastoma	4	1.21
Ameloblastic carcinoma	4	1.21
Unspecified	1	0.30
TOTAL	330	99.99

Table II. Frequency distribution of age range in patients with desmoplastic ameloblastoma

Age range (y)	*n*	*%*
0-10	0	0.00
11-20	2	11.76
21-30	5	29.41
31-40	3	17.65
41-50	3	17.65
51-60	1	5.88
>60	3	17.65
TOTAL	17	100.00

Age range = 17-73 years. Mean age = 38.353 years. Standard deviation = 18.197 years. Median = 32.000 years. Mode = 22.000 years.

DISCUSSION

Observation from this study showed that DA represented 5.15% of 330 cases of ameloblastoma in this series. This observation is consistent with a global incidence of 0.9%-12.1% of DA relative to ameloblastoma in general.[18-22] Our observation of 2 histologic variants of DA, with simple DA being predominant (88.0%) and DA with osteoplasia being rare (12.0%), is

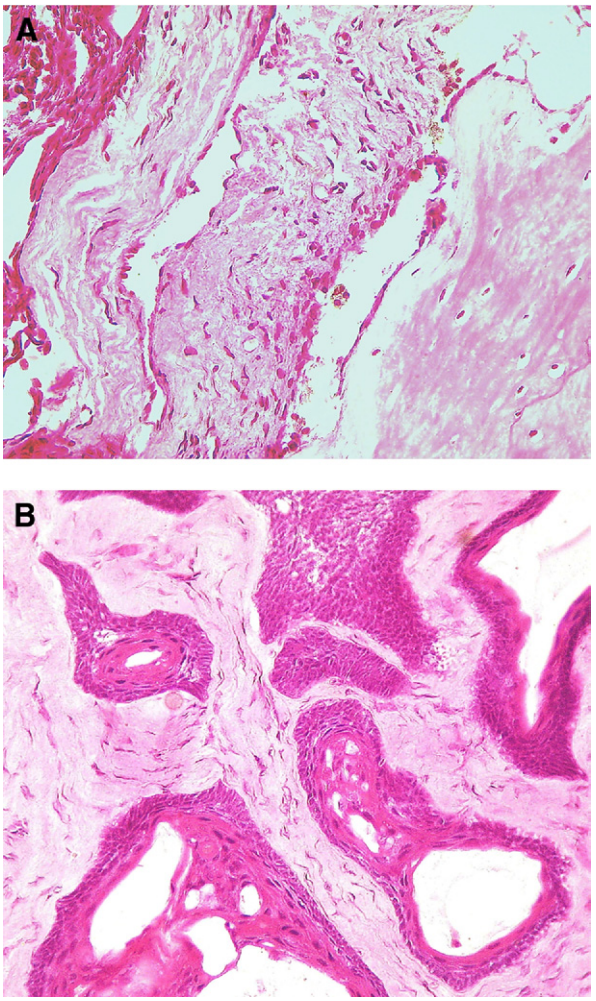

Fig. 1. **A,** Photomicrograph of desmoplastic ameloblastoma, showing follicles of ameloblastoma tumour cells with peripheral ameloblast-like cells and central stellate reticulum–like cells as well as some focal areas of cystification. The stroma consists of desmoplastic fibrous connective tissue. Hematoxylin-eosin (HE) stain, ×400. **B,** Photomicrograph of desmoplastic ameloblastoma, in which acanthomatous changes and cystification are seen in the tumor cells. HE stain, ×200.

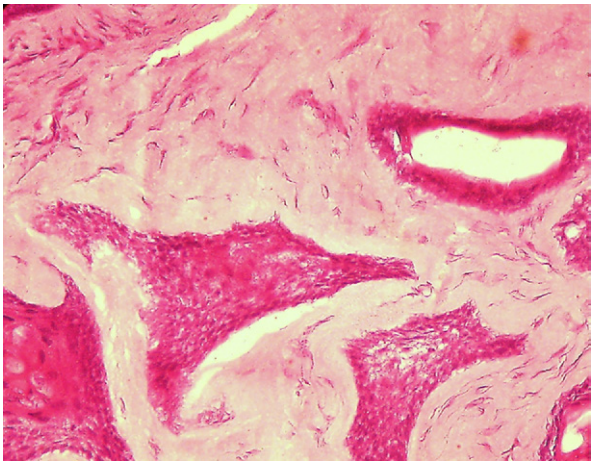

Fig. 2. Photomicrograph of desmoplastic ameloblastoma with osteoplasia. Tumor cells are to the left of the photomicrograph and bone formation to the right of the photomicrograph. Hematoxylin-eosin stain.

Table III. Growth pattern of ameloblastoma

Type	*EMGR (cm/mo)*	*Variance*	*SD*
Conventional	0.7080	1.356	1.1645
Desmoplastic	0.3653	0.195	0.4417

EMGR, Estimated mean growth rate.
Variance between samples = 9.65. Residual variance = 0.78. F statistic = 12.44. P = .000480.

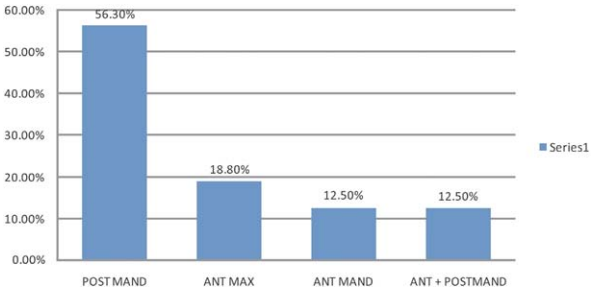

Fig. 3. Site distribution of desmoplastic ameloblastoma. *Post*, Posterior; *Mand*, mandible; *Ant*, anterior; *Max*, maxilla.

worthy of note, because most of the reports in the scientific literature have reported occurrence of only the simple DA. However, Philipsen et al.,[11] 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. The veracity of this could be clarified in future biologic studies.

Observation from this series that DA, like the conventional ameloblastoma, occurred more frequently in the posterior mandible differs from the anterior maxillary/mandibular site predilection previously reported in the scientific literature.[11-14,18,19,23-25]

Observation of a mean age of 38.3 ± 18.20 years in this series is slightly lower than 42.9 and 41.2 years as reported by Philipsen et al.[12] and Rejeev et al.,[18] respectively, but falls within the age range of third and fifth decades recently reported by Sivapathasundharam et al.[14] Regarding gender prevalence, our observation in this series of male:female ratio of 1.27:1 for DA is similar to other studies in the literature.[12,13,15,18,23]

Multilocular radiolucency, accounting for 82.4% of cases in this series, is the predominant radiographic presentation. The remaining 17.6% presented as a mixed radiolucent and radiopaque radiographic appearance, thereby mimicking a fibro-osseous lesion.

Further observation in this series of 0.36 ± 0.44 cm/mo EMGR of DA is significantly lower than 0.71 ± 1.16 cm/mo EMGR for conventional ameloblastoma ($P = .000480$), suggesting that DA might be less biologically aggressive than conventional ameloblastoma. In an earlier Nigerian study using the EMGR, the conventional ameloblastoma was reported to have a higher growth rate than unicystic ameloblastoma.[26] It could be speculated that the desmoplasia in DA might act as a limiting barrier for local spread of the DA tumor cells compared with the situation in conventional ameloblastoma where such barrier is absent.

It could be argued that duration of lesion, information obtained from the patient to compute EMGR, is unreliable. However, the use of EMGR is regarded to be an estimate of growth rate rather than the actual growth rate. This could be argued as a limitation of this study, but it does not invalidate the essential findings. Our Oral and Maxillofacial Surgery Department treats all cases of ameloblastoma, except unicystic ameloblastoma, by surgical resection. Both DA and conventional ameloblastoma in the present series were therefore treated by surgical resection, and there is no report in our record of recurrence of any of these lesions after this mode of therapy. Further study with close monitoring and long-term follow-up of such cases of ameloblastoma is in the pipeline.

In 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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