



Published in final edited form as:

*Oral Oncol.* 2009 September ; 45(9): 752–759. doi:10.1016/j.oraloncology.2009.01.016.

## Desmoplastic ameloblastoma – A review

Zhi-Jun Sun<sup>a,b,\*</sup>, Yan-Ru Wu<sup>b</sup>, Ning Cheng<sup>b</sup>, Roger A Zwahlen<sup>c</sup>, and Yi-Fang Zhao<sup>a,\*</sup>

<sup>a</sup>Department of Oral and Maxillofacial Surgery, School and Hospital of Stomatology, Wuhan University, Wuhan, Hubei, China

<sup>b</sup>Key Laboratory of Oral Biomedical Engineering (Wuhan University), Ministry of Education, Wuhan, Hubei, China

<sup>c</sup>Discipline of Oral and Maxillofacial Surgery, Faculty of Dentistry, University of Hong Kong, 34 Hospital Road, Hong Kong (SAR), China

### SUMMARY

Among the ameloblastomas, the desmoplastic variation is rare. The desmoplastic ameloblastoma (DA) is characterized by specific clinical, imaging, and histological features. The here presented retrospective analysis investigated the clinicoradiographic features of an overall of 115 DA-cases, having been reported in literature from 1984 to 2008. DA showed a nearly equal male to female ratio (55/59) with a prevalence within the fourth and fifth decades. Sixty-two lesions occurred in the mandible and fifty-one lesions in the maxilla. Clinically, a painless swelling with buccal extension was the most common presentation being found in 48 cases. Radiologically, the lesion often presented multilocular (49.3%; 36/73), mixed radiolucent/radiopaque (55.6%; 50/90) and with ill-defined borders (64.0%; 48/75). Whereas enucleation provided a recurrence rate of 21.1%, resection reduced this rate remarkably to 3.1%. The average period until recurrence was 36.9 months. Histologically, scattered epithelial nests and extensively desmoplasia were prominent features of DA. In conclusion, these retrospective results confirm the statement that DA is a variation among ameloblastomas. DA present clinicoradiographic and histologic distinct features, when compared with “conventional ameloblastomas”.

### Keywords

Desmoplastic ameloblastoma; Mandible; Maxilla; Histology; Radiology; Treatment

### Introduction

Ameloblastomas, although locally invasive, are considered to be benign neoplasms deprived from odontogenic epithelium. The term “ameloblastoma” includes several clinicoradiographic and histological different types. DA is rare, accounting for approximately 4% to 13% of ameloblastomas.<sup>1–3</sup> It was first described by Eversole et al.<sup>4</sup> in

\*Corresponding authors. Address: Department of Oral and Maxillofacial Surgery, School and Hospital of Stomatology, Wuhan University, 237# Luo Yu Road, Wuhan 430079, Hubei, China. Tel.: +86 27 87686125; fax: +86 27 87873260.

Conflict of Interest Statement  
None declared

1984 as a new type of ameloblastoma which affected different mandibular areas, presenting a unique histopathological pattern and clinicoradiographic findings. This variation has been included in the World Health Organization's histopathological classification.

Histologically, it is characterized with extensive stromal collagenisation or desmoplasia with small nests and strands of odontogenic epithelium (Fig. 1).<sup>5</sup> Additionally "hybrid" lesions showing some microscopic features of the desmoplastic variant together with typical areas of follicular or plexiform ameloblastoma have been described.<sup>1</sup> Most diagnoses are made related to the histological specimen after the DA was already removed.

Radiologically, the DA frequently presented as diffuse, mixed radiolucent – radiopaque lesion, apting to be misdiagnosed as fibro-osseous lesion.<sup>6</sup> Both surgeons and radiologists may be aware of clinicoradiographic features of the "common" ameloblastomas, however, may ignore this variation. The purpose of this article was to review the DA within the English literature in order to provide diagnostic tools for this rare variation of ameloblastomas.

A research on MEDLINE for adequately documented cases of DAs was performed in the English literature. Criteria for inclusion in the here presented study were a confirmed histopathological diagnosis of DAs with a detailed clinicoradiographic description. Thus, a total of 115 cases from 35 published papers<sup>1-4,6-36</sup> have been analyzed and the clinicoradiographic data were summarized in Table 1. The 115 cases of DA including 12 cases of so-called "hybrid lesion of ameloblastoma where areas of intraosseous ameloblastoma coexist with areas of DA. The data was analyzed by SPSS 11.5 software program. The  $X^2$  test was used for the CROSSTABS procedure and exact test was used when needed. In all analyses, the significance level was set at a level of  $P = 0.05$ .

## Clinical features

### Age and gender distribution

Apart from 1 case without mentioned of age, the age and gender distributions of 114 cases DAs were listed in Fig. 2A. The age at time of primary presentation ranged from 17 to 83 years with a mean age of 41.9 years (males: 44.6 years, females: 39.0 years) and a median age of 42.0 years. The age prevalence was in the 3th to 5th decades. Whereas there were two peaks among females, in the 3rd and in the 4th decades, the males presented only a single peak in the 5th decade. Among the 114 DA patients, there were 59 (51.8%) females and 55 (48.2%) males. However, there is no statistically significant gender distribution as in other types of ameloblastomas. In the fifth decade the incidence rate is a significantly higher in males than females ( $p < 0.05$ ). Although no difference between genders has been reported<sup>14</sup> in the onset of this disease, people in the 4th and 5th decade are most commonly affected.

### Race

The incidence of DA among ameloblastomas ranges from 0.9% to 12.1% in different races.<sup>1,26,31,35,37</sup> Concerning the race, 69 out of 115 cases DAs with detailed description, of which 25(36.2%) were Japanese, 15(21.7%) were Chinese and 10(14.5%) were Indian. Data from different geographical regions seem to suggest a biogeographical pattern in that the relative

frequency of DA is slightly higher in Asian population. However, more systematic studies on DA are needed to verify such suggestions.

### Size

Regarding the tumor size of 3.0 cm in diameter, 32 cases (44.4%) of the DAs were larger, 36 cases (50.0%) smaller and only 4 cases (5.6%) equal to 3.0 cm at the initial presentation. In a review of 3677 ameloblastomas, Reichart et al.<sup>38</sup> reported that the average tumor size at the initial presentation was 4.3 cm (median, 3.0 cm). Therefore it may be assumed that DA may be smaller than the “normal” type of ameloblastomas. There were neither statistical significance between the tumor size and localization, nor the recurrence ( $p > 0.05$ ).

### Location

Anatomical localization was described in 113 cases. 62 cases occurred in mandible and 51 cases in the maxilla. Detailed tooth location was reported in 61 cases, being shown in Fig. 2B. The anterior regions until the first premolar were frequently involved. In the maxilla, 42(82.4%) lesions were in the anterior region and 34(54.8%) in the mandible. The maxillary sinus was involved in nine and the ramus in two cases. Whereas most lesions were one-sided, a total of 18 crossed the midline.

### Presentation

The clinical presentation of 56 cases DAs was summarized. Forty-eight (85.7%) patients presented with a painless swelling or a bone expansion,<sup>2-4,6-9,11-18,21,24-28,30-32,34,36</sup> which was the most conspicuous clinical manifestation of the affected area. Six patients presented with a tender swelling.<sup>10,20,23,31,33</sup> In case of maxillary sinus involvement, nasal<sup>27</sup> and pharyngeal obstruction<sup>4</sup> occurred, however with a very small probability (1.8%).

Among these 56 cases, 16 (69.6%) patients showed a buccal (Fig. 3), one (4.35%) a palatal/lingual and six (26.1%) both a buccal and a palatal/lingual expansion. Only 36 out 114 reports of DAs provided information about tooth resorption and 27 cases provided information about tooth displacement. In nine cases (25.0%) tooth resorption was reported. In 24(88.9%) out of 27 cases tooth displacement was observed (Fig. 4).

### Radiographic features

The radiological information available for each reviewed case was not uniform. Radiographically, 48(64.0%) out of 75 cases with detailed border description presented poorly defined borders, whereas well defined borders were detected in 27 (36.0%). The lesions appeared mixed radiolucent / radiopaque in 50(55.6%) out of 90 case, and radiolucent in 40(44.4%) cases. According to locularity, 36(49.3%) out of 73 cases with detailed description presented multilocular (Fig. 4A), 19(26.0%) cases were unilocular (Fig. 4B) and 18 (24.7%) cases were not loculated. Computed tomography was performed in 20 cases usually detecting ill-defined, mixed radiolucency–radiopacity with buccal expansion (Fig. 4C).<sup>2,3,6-9,11-19,25,26,28,29,39</sup>

## Histopathology

Histologically, scattered epithelial nests and extensively desmoplasia were prominent features of DA (Fig. 1).<sup>40</sup> Most tumors contain cords of odontogenic epithelium, but these tumors tend to lack the more typical follicular pattern. Typical ameloblastic columnar cells may be scant, and peripheral pallisading may be absent. In the focal area, the islands have a swirled hypercellular appearance. Osteoplasia may also be present. The surrounding stroma accompanied the cells islands are characterized for significant collagen proliferation. However, in some juxtaepithelial area, loose myxoid changes can be found.

## Management

Treatment methods were mentioned in 83 out of 115 cases. Most cases of DA were treated by resection (77.1%) and some cases were treated by enucleation and/or curettage 19 (22.9%). Only 69 DA provided an available postoperative follow-up period between 2 months and 20 years. Eleven patients suffered from recurrence (15.9%) with an average recurrence period of 36.9 months.<sup>1,12,22,26,29,31,39</sup> Among these patients, one underwent revision and showing thereafter no sign of recurrence after 6 years. Among these 11 patients, two suffered from recurrence after resection and four after enucleation, the treatment methods of other five cases were not mentioned. The remaining five remained unknown. The recurrence rate through the treatment of enucleation was significantly higher than that of resection ( $p < 0.01$ ).

## Discussion

Clinically, DA may develop in all ages, however, people of the 4th and 5th decade are more prone to be affected.<sup>14,17</sup> No gender predilection of DA has been reported. Usually the DA variation is smaller in size than other types of ameloblastoma. DA occurs in the anterior or premolar regions of the jaws and there is not any difference in prevalence between the maxilla and mandible.

This represents a contrast with classic ameloblastomas, which are usually found in the posterior mandibular regions presenting a mandible to maxilla ratio of 5:1.<sup>38</sup> The main symptom at first clinical examination of DA is a painless swelling, with frequently buccal expansion. Tooth displacement were frequently been seen.<sup>32</sup>

Radiologically, there is not any agreement among DA reviews as to whether it deals with a radiolucent or mixed radiolucent– radiopaque lesion.<sup>14,32</sup> In a review, only 1 of the 15 cases of DA had a radiolucent appearance.<sup>32</sup> Whether this is due to limited data or a specific patient population is uncertain. However, in the here presented retrospective analysis, the content of the lesion was mixed radiolucent/radiopaque in 55.6% cases. Radiographically, other types of ameloblastomas are classically described as a unilocular or multilocular radiolucences with well-defined borders,<sup>41</sup> whereas DAs are usually described as poorly defined in most cases. In the series by Reichart et al.<sup>38</sup>, 51.1% of the tumors were described as unilocular, whereas 48.9% were described as multilocular or multicystic. When applying this criterion to the here presented study, 24.7% did not show any locularity, 26.0% were unilocular, and 49.3% were multilocular. On examination of the radiographic borders of 75

reported cases of desmoplastic ameloblastomas in the here presented study, poorly defined borders were reported in 48 cases (64.0%) and well defined borders were seen in only 27 cases (36.0%). The DA usually appears in the anterior and premolar regions as a mixed radiolucent and radiopaque lesion sometimes mimicking a benign fibro-osseous lesion.<sup>1</sup> Approximately half of the reported lesions showed diffuse radiological borders<sup>32</sup> and mimicking fibro-osseous lesions or malignant tumors. The lamina dura also was involved.<sup>34</sup> The radiographic appearance may indicate that this tumor is more aggressive than other variants of ameloblastoma. According to Philipsen et al.,<sup>33</sup> radiographically ill-defined borders suggest an infiltrative process with propensity to recur. Thus the pronounced stromal reaction characteristic of DAs can be viewed as a defensive response of the host to the “aggressive” tumor. This, along with the fact that recurrences of DAs have been documented by more than one author, suggests potentially aggressive biologic behavior.<sup>29,31,39</sup> The ill-defined borders of DAs reflect the desirability for a relatively radical approach to treatment.

Concerning the biological behavior of DA, it is mentioned in the WHO classification of odontogenic tumors that DA, like unicystic ameloblastoma and peripheral ameloblastomas, possibly have a lower recurrence rate than other ameloblastomas.<sup>5</sup> In contrast to that statement of the WHO, reviewing the literature provided the information that DA showed a similar recurrence rate (15.9%) with the other types of ameloblastomas. Keszler et al.<sup>29</sup> even reported a higher recurrence rate (21.4%) than the other type (10.1%) of ameloblastoma. The reason for this may be somewhat hypothetical: First, radiographically, DAs are apt to be mismatched with fibro-osseous lesions. The accurate diagnosis of a DA is hard to achieve before the operation; Second, DA frequently present with ill-defined border making it difficult to investigate the exact interface of the lesion with normal bone; third, the more common location in the maxilla may produce an early invasion of adjacent structures. In view of the paucity of DA case series and the only limited understanding of its biologic behavior and prognosis, the proper treatment strategies for DA are not entirely defined so far. Whether the recurrence is due to the nature of the tumor or due to the incomplete surgery remains speculative. Prospective studies with regular and long term follow-up are needed to provide the necessary information before any conclusions in this aspect can be drawn. According to the average relapse time in 11 patients, which was 36.9 months, the follow-up period should be more than three years.

## Conclusions

Clinically, DA usually presents with a painless buccal swelling predominantly located in the anterior of the maxilla and mandible. Radiologically, most DAs present with a multilocular mixed radiolucent/radiopaque lesion with ill-defined borders. Tooth displacement was frequently found and root resorption was discovered in 25% of the reported cases. Resection and enucleation are the main treatment modalities of DAs. The recurrence rate is similar to the “normal” ameloblastomas. The recurrence rate after enucleation is significantly higher than the one after resection.

## Acknowledgments

This work is supported by a grant from National Natural Science Foundation of China (No.: 30600712) to Dr. Z.J. Sun.

## Appendix

Data on 115 cases of desmoplastic ameloblastoma.

Author	Age (Y)	Sex <sup>1</sup>	Race	Tumor size (cm)	Tumor Location <sup>2</sup>	Borders <sup>3</sup>	Locularity <sup>4</sup>	Radiology <sup>5</sup>	Tooth resorption	Tooth displacement	Treatment <sup>6</sup>	Follow-up <sup>7</sup>
Smullin et al. <sup>2</sup>	44	F	NA	<3.0	Max.L3-5	W	NA	NA	NA	NA	Res	NR1Y
Curran and Byerly <sup>7</sup>	56	M	black	<3.0	Mand.R4-5	W	NA	NA	No	NA	Res	NR51M
Sivapathasundharam et al. <sup>8</sup>	25	M	Indian	NA	Max.R3-5	W	Uni	Mixed	No	Yes	Res	NA
	40	M	Indian	NA	Max.L1-5	W	Uni	Mixed	No	No	Res	NA
	30	F	Indian	NA	Max.L1-5	I	NA	Mixed	No	No	Res	NA
	32	F	Indian	3.0	Max.L1-4	I	Multi	Mixed	Yes	Yes	Res	NR1Y
<i>a</i>	40	M	NA	NA	Max.L1-6	I	NA	Mixed	No	No	Res	NA
<i>a</i>	31	F	NA	NA	Mand.L2-R5	I	Uni	Mixed	No	Yes	Res	NA
Shashikanth et al. <sup>9</sup>	32	F	Indian	<3.0	Max.L1-4	I	Multi	Mixed	Yes	Yes	Res	NR1Y
Desai et al. <sup>10</sup>	32	M	Indian	>3.0	Mand.Ramus	W	Uni	RL	NA	NA	Cure	NR2Y
Hirota et al. <sup>11</sup>	17	F	Japanese	NA	Max.R3-4, sinus	W	Multi	Mixed	NA	Yes	Res	NR7Y
Pillai et al. <sup>12</sup>	24	F	Indian	<3.0	Max.L3-5, sinus	I	Multi	RL	No	Yes	Cure+BG	R2M
Durmus et al. <sup>13</sup>	68	M	Turkish	<3.0	Mand.R1-3	W	Uni	RL	Yes	NA	Cure	NR32M
Beckley et al. <sup>14</sup>	31	M	Hispanic	<3.0	Mand.L3-4	I	No Loc	Mixed	No	Yes	Res+BG	NR19M
Iida et al. <sup>15</sup>	52	M	Japanese	NA	Max.L2-5, sinus	I	Multi	Mixed	NA	Yes	Res	NR
Mannuel et al. <sup>16</sup>	20	F	NA	<3.0	Max.L3-4	W	Uni	Mixed	No	Yes	Res	NR22M
Mintz and Velez <sup>17</sup>	52	M	NA	NA	Max.L1-8, sinus	I	Multi	Mixed	NA	Yes	Res	NR
	51	F	NA	NA	Mand.R3-L5	I	Multi	Mixed	NA	Yes	Res+BG	NR1Y
Wakoh et al. <sup>18a</sup>	35	F	Japanese	>3.0	Mand. L3-5	W	Multi	Mixed	Yes	Yes	Res	NR
Philipsen et al. <sup>3</sup>	42	F	Caucasian	NA	Mand.L4-R4	I	Uni	Mixed	NA	Yes	Res+BG	NR36M
Kishino et al. <sup>19</sup>	41	M	Japanese	>3.0	Max.R5-L2	W	Multi	RL	No	NA	Res	NR17Y
	28	M	Japanese	>3.0	Max.R1-R4	I	NA	Mixed	No	NA	Res	NR72M
	57	M	Japanese	<3.0	Max.R1-R2	W	NA	RL	No	NA	Res	NR8Y
	58	F	Japanese	3.0	Max.L1-L2	W	Uni	RL	No	NA	Cure	NR16Y
	17	M	Japanese	<3.0	Mand.R6-R7	W	Uni	RL	No	NA	Cure	NR5Y
	50	M	Japanese	>3.0	Mand.L5-R4	I	NA	Mixed	No	NA	Res	NR6Y
	51	M	Japanese	>3.0	Mand.L5-R4	I	NA	Mixed	No	NA	Res+BG	NR23Y
	47	M	Japanese	>3.0	Mand.L4-R2	I	NA	Mixed	No	NA	Res+BG	NR20Y
	42	M	Japanese	>3.0	Mand.L3-R5	I	NA	Mixed	No	NA	NA	NA
Saran et al. <sup>20</sup>	25	F	Indian	<3.0	Max.L2-4	W	Uni	NA	NA	NA	Res	NR10M
Louis et al. <sup>21</sup>	33	M	Black	<3.0	Max.L3-4, sinus	I	NA	Mixed	No	Yes	Res+BG	NR18M
Takata et al. <sup>22</sup>	53	M	Japanese	>3.0	Max.R2-3	W	No Loc	RL	Yes	NA	Res	NR
	33	F	Japanese	3.0	Max.R1-5	I	Multi	Mixed	No	NA	Cure	NR
	51	M	Japanese	<3.0	Max.L3-5	W	Uni	RL	No	NA	Res	NR
	52	M	Japanese	>3.0	Max.L2-5	I	Multi	Mixed	No	NA	Res	NR
	54	M	Japanese	>3.0	Mand.R1-L5	W	No Loc	Mixed	No	NA	Res	NR
	17	M	Japanese	<3.0	Mand.L3-5	I	Multi	Mixed	No	NA	Cure	4R
<i>a</i>	48	M	Japanese	>3.0	Mand.R2-6	I	Multi	Mixed	NA	Yes	Cure	R14M
Kawai et al. <sup>6</sup>	56	M	Japanese	NA	Mand. L4-7	W	Uni	RL	NA	NA	Res	NA
Ludvikoya et al. <sup>23</sup>	50	M	NA	>3.0	Mand.R4-8	I	No Loc	RL	NA	NA	Res	NR
	42	F	NA	<3.0	Mand.L4-5	I	No Loc	RL	NA	NA	Res	NR3Y
Sakashita et al. <sup>24</sup>	60	F	Japanese	>3.0	Max.L1-4	I	Multi	RL	NA	NA	Res	NR
Lee et al. <sup>25</sup>	83	F	Asian	<3.0	Mand.L2-3	W	Multi	Mixed	NA	Yes	Res	NR
Lam et al. <sup>26</sup>	64	M	Chinese	>3.0	Max: A, PM, M	I	No Loc	RL	NA	NA	Res	R2Y
	18	M	Chinese	>3.0	Max: A	I	Multi	RL	NA	NA	Res	NR53M
	68	F	Chinese	<3.0	Max:PM	I	Multi	RL	NA	NA	Cure	R74M
	37	F	Chinese	<3.0	Mand: A	I	Multi	RL	NA	NA	Res	NR77M
	37	M	Chinese	<3.0	Mand: A	I	No loc	RL	NA	NA	Res	NR32M
Fukushima et al. <sup>27</sup>	70	M	NA	>3.0	Max: PM, M, sinus	I	No Loc	Mixed	NA	NA	Res	NR18M
Thompson et al. <sup>28</sup>	31	F	Black	>3.0	Max: L2-6, sinus	I	No Loc	Mixed	Yes	Yes	Res+BG	NR3Y

Author	Age (Y)	Sex, <sup>1</sup>	Race	Tumor size (cm)	Tumor Location, <sup>2</sup>	Borders, <sup>3</sup>	Locularity, <sup>4</sup>	Radiology, <sup>5</sup>	Tooth resorption	Tooth displacement	Treatment, <sup>6</sup>	Follow-up, <sup>7</sup>
Keszler et al. <sup>29</sup> (14 cases)	19-62 (Mean 37.8) in 15, 1NA	11 F 2 M 1 NA	NA	NA	2Max; 10Mand; 2NA	4W	4 Uni 5 Multi	9 RL; 3 Mixed	NA	NA	5 Res 9 Cure	3R; 11NR
Ashman et al. <sup>30</sup>	53	M	Black	>3.0	Mand: A, PM	W	No Loc	Mixed	NA	Yes	Res	NR9M
Ng and Siar <sup>31</sup>	21	F	Chinese	<3.0	Mand.L1-4	I	NA	RL	NA	NA	Res	NA
	25	M	Chinese	NA	Max, right	NA	NA	Mixed	NA	NA	Res	NA
	25	M	Malay	NA	Mand.R2-L4	NA	Multi	RL	NA	NA	Res	NA
	27	F	Chinese	NA	Mand.R2-L4	NA	NA	Mixed	NA	NA	NA	NA
	29	F	Kadazan	NA	Mand.R5-L5	NA	Multi	Mixed	NA	NA	Res	NA
	32	F	Indian	NA	Max.L2-3	NA	NA	NA	NA	NA	NA	NA
	33	M	Malay	3.0	Max.R1-L3	NA	NA	Mixed	NA	NA	NA	NA
	33	F	Malay	NA	Max.R2-L5	I	NA	NA	NA	NA	Res	NR1Y
	38	M	Malay	<3.0	Max.L2-3	NA	NA	NA	NA	NA	Res	NA
	40	F	Chinese	NA	Mand:anterior	NA	Multi	RL	NA	NA	NA	NA
	41	F	Malay	NA	Max, R3	NA	NA	NA	NA	NA	Res	NA
	42	F	Chinese	NA	L, Horiz ramus	NA	NA	NA	NA	NA	NA	NA
	43	F	Malay	NA	Mand.R3-L3	NA	NA	NA	NA	NA	NA	NA
	43	F	Chinese	NA	Mand.R4-L2	NA	Multi	RL	NA	NA	NA	NA
	44	F	Indian	<3.0	Mand.L3	NA	NA	NA	NA	NA	Res	R4Y
	46	F	Sikh	NA	Max.Left	NA	NA	Mixed	NA	NA	Res	NR1Y
	60	M	Chinese	NA	Mand.L2-3	NA	Multi	RL	NA	NA	NA	NA
Kaffe et al. <sup>32</sup>	41	M	NA	>3.0	Max: L2-R7, sinus	I	No Loc	Mixed	NA	NA	Res	NR3Y
Philipsen et al. <sup>33</sup>	21	M	Chinese	>3.0	Max: L1-R6	I	Multi	RL	Yes	Yes	Res+BG	NA
<i>a</i>	53	M	Chinese	>3.0	Max: L1-8	I	Multi	RL	Yes	Yes	Res	NA
<i>a</i>	55	M	Chinese	>3.0	Mand: L3-7	I	Multi	Mixed	Yes	NA	Res	NA
Tanimoto et al. <sup>34</sup>	24	F	Japanese	>3.0	Mand: L2-6	I	NA	Lucent	No	Yes	Res	NR1Y
Higuchi et al. <sup>35</sup>	46	M	NA	>3.0	Max:A	I	Multi	Mixed	No	Yes	NA	NA
	38	F	NA	>3.0	Mand:A	W	Multi	Mixed	Yes	Yes	NA	NA
	53	M	NA	<3.0	Max:PM,M	W	Uni	Mixed	No	Yes	NA	NA
<i>a</i>	58	M	Japanese	>3.0	Mand:A, PM	NA	Multi	Mixed	NA	NA	Cure	NR
<i>a</i>	70	M	Japanese	>3.0	Mand:PM,M	NA	Multi	Mixed	NA	NA	Res	NR
Yoshimura and Saito <sup>36</sup>	36	F	Japanese	>3.0	Max: A, PM, M, sinus	I	Multi	Mixed	NA	NA	Res	NR9Y
Waldron and el-Mofly <sup>1</sup> (14 cases)	21 to 68 (mean, 45.5)	7M,7F	NA	13 < 3.0 1 > 3.0	2 Mand.A; 3 Mand. PM; 2 Mand.M; 6 Max.A; 1 Max.PM	5I;1W	1 Uni 5 No loc	4 Mixed 2 RL	NA	NA	NA	2R1NR 11NA
5 cases <sup>a</sup>	25-82	2M,3F	NA	NA	4 Mand: PM, M, 1 Mand	NA	NA	NA	NA	NA	NA	NA
Eversole et al. <sup>4</sup>	48	F	Malaysian	>3.0	Max: PM, M	I	No Loc	Mixed	NA	NA	Res	NR2Y
	50	F	Caucasian	<3.0	Mand: A, PM, M	I	Uni	Lucent	NA	NA	NA	NA
	47	F	Black	NA	Mand: A	I	No Loc	Mixed	NA	Yes	Res+BG	NA

<sup>1</sup>F: Female, M: Male.

<sup>2</sup>Mand: Mandibular, Max: Maxillary, L: Left, R: Right, A: Anterior, P: Posterior.

<sup>3</sup>I: Ill-defined, W: Well defined.

<sup>4</sup>M: Multilocular, U: Unilocular.

<sup>5</sup>RL: Radiolucency, RO: Radiopaque, Mixed: Mixed radiolucent-radiopaque.

<sup>6</sup>Curr: Curettage; Res: Resection; BG: Bone graft.

<sup>7</sup>NR: No recurrence.

<sup>a</sup>Hybrid lesion of ameloblastoma.

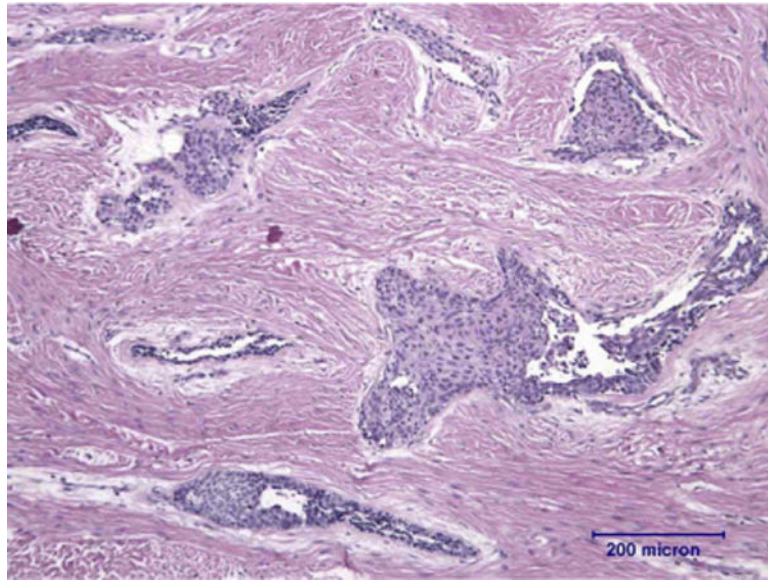
## References

1. Waldron CA, el-Mofly SK. A histopathologic study of 116 ameloblastomas with special reference to the desmoplastic variant. *Oral Surg Oral Med Oral Pathol.* 1987; 63(4):441–51. [PubMed: 3472145]

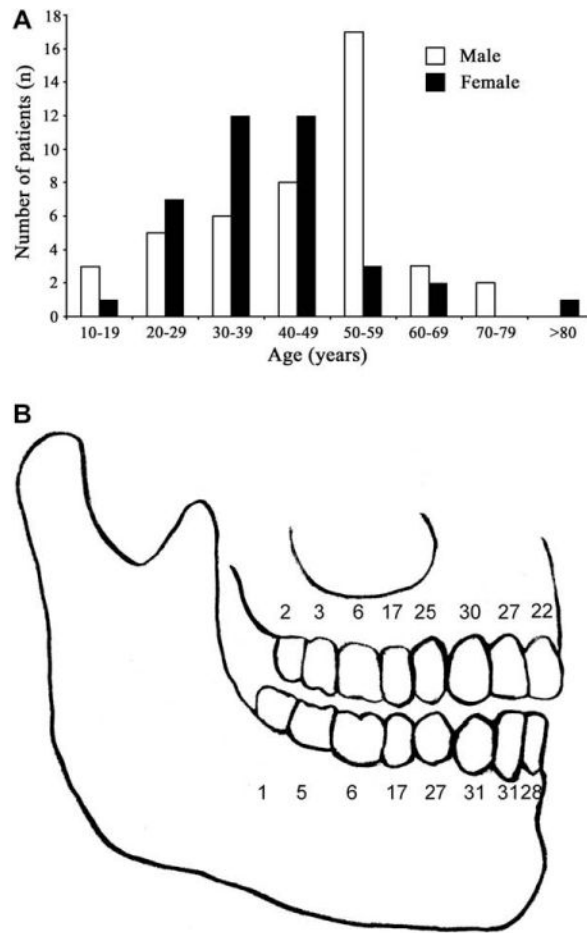
2. Smullin SE, Faquin W, Susarla SM, Kaban LB. Peripheral desmoplastic ameloblastoma: report of a case and literature review. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2008; 105(1):37–40. [PubMed: 17764981]
3. Philipsen HP, Reichart PA, Takata T. Desmoplastic ameloblastoma (including “hybrid” lesion of ameloblastoma). Biological profile based on 100 cases from the literature and own files. *Oral Oncol.* 2001; 37(5):455–60. [PubMed: 11377234]
4. Eversole LR, Leider AS, Strub D. Radiographic characteristics of cystogenic ameloblastoma. *Oral Surg Oral Med Oral Pathol.* 1984; 57(5):572–7. [PubMed: 6587306]
5. Gardner, DG., Heikinheimo, K., Shear, M., Philipsen, HP., Coleman, H. Ameloblastomas. In: Barnes, L.Eveson, EJ.Reichart, P., Sidransky, D., editors. World Health Organization classification of tumors: pathology and genetics of head and neck tumors. 3rd. Lyon: IARC Press; 2005. p. 296-300.
6. Kawai T, Kishino M, Hiranuma H, Sasai T, Ishida T. A unique case of desmoplastic ameloblastoma of the mandible: report of a case and brief review of the English language literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1999; 87(2):258–63. [PubMed: 10052385]
7. Curran AE, Byerly PD. Peripheral desmoplastic ameloblastoma: report of a rare case. *J Oral Maxillofac Surg.* 2008; 66(4):820–5. [PubMed: 18355614]
8. Sivapathasundharam B, Einstein A, Syed RI. Desmoplastic ameloblastoma in Indians: report of five cases and review of literature. *Indian J Dent Res.* 2007; 18(4):218–21. [PubMed: 17938502]
9. Shashikanth MC, Neetha MC, Ali IM, Shambulingappa P. Desmoplastic ameloblastoma in the maxilla: a case report and review of literature. *Indian J Dent Res.* 2007; 18(4):214–7. [PubMed: 17938501]
10. Desai H, Sood R, Shah R, Cawda J, Pandya H. Desmoplastic ameloblastoma: report of a unique case and review of literature. *Indian J Dent Res.* 2006; 17(1):45–9. [PubMed: 16900895]
11. Hirota M, Aoki S, Kawabe R, Fujita K. Desmoplastic ameloblastoma featuring basal cell ameloblastoma: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2005; 99(2): 160–4. [PubMed: 15660085]
12. Pillai RS, Ongole R, Ahsan A, Radhakrishnan RA, Pai KM. Recurrent desmoplastic ameloblastoma of the maxilla: a case report. *J Can Dent Assoc.* 2004; 70(2):100–4. [PubMed: 14756940]
13. Durmus E, Kalayci A, Ozturk A, Gunhan O. Desmoplastic ameloblastoma in the mandible. *J Craniofac Surg.* 2003; 14(6):873–5. [PubMed: 14600629]
14. Beckley ML, Farhood V, Helfend LK, Alijanian A. Desmoplastic ameloblastoma of the mandible: a case report and review of the literature. *J Oral Maxillofac Surg.* 2002; 60(2):194–8. [PubMed: 11815921]
15. Iida S, Kogo M, Kishino M, Matsuya T. Desmoplastic ameloblastoma with large cystic change in the maxillary sinus: report of a case. *J Oral Maxillofac Surg.* 2002; 60(10):1195–8. [PubMed: 12378499]
16. Manuel S, Simon D, Rajendran R, Naik BR. Desmoplastic ameloblastoma: a case report. *J Oral Maxillofac Surg.* 2002; 60(10):1186–8. [PubMed: 12378496]
17. Mintz S, Velez I. Desmoplastic variant of ameloblastoma: report of two cases and review of the literature. *J Am Dent Assoc.* 2002; 133(8):1072–5. [PubMed: 12198986]
18. Wakoh M, Harada T, Inoue T. Follicular/desmoplastic hybrid ameloblastoma with radiographic features of concomitant fibro-osseous and solitary cystic lesions. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2002; 94(6):774–80. [PubMed: 12464906]
19. Kishino M, Murakami S, Fukuda Y, Ishida T. Pathology of the desmoplastic ameloblastoma. *J Oral Pathol Med.* 2001; 30(1):35–40. [PubMed: 11140898]
20. Saran RK, Nijhawan R, Vasishtha RK, Rattan V. Desmoplastic ameloblastoma: a case report with fine-needle aspiration cytologic findings. *Diagn Cytopathol.* 2000; 23(2):114–7. [PubMed: 10888757]
21. Louis PJ, Fugler RC, August M. Mixed radiolucent/radiopaque lesion of the maxilla. *J Oral Maxillofac Surg.* 2000; 58(1):86–90. [PubMed: 10632170]



22. Takata T, Miyauchi M, Ogawa I, Zhao M, Kudo Y, Sato S, et al. So-called 'hybrid' lesion of desmoplastic and conventional ameloblastoma: report of a case and review of the literature. *Pathol Int.* 1999; 49(11):1014–8. [PubMed: 10594850]
23. Ludvikoya M, Michal M, Zamecnik M, Houba R, Sedlacek P, Csillagi E, et al. Desmoplastic ameloblastoma. *Cesk Patol.* 1998; 34(3):94–8. [PubMed: 9750421]
24. Sakashita H, Miyata M, Okabe K, Kurumaya H. Desmoplastic ameloblastoma in the maxilla: a case report. *J Oral Maxillofac Surg.* 1998; 56(6):783–6. [PubMed: 9632341]
25. Lee CY, Lee J, Hirata K, Tomich CE. Desmoplastic variant of ameloblastoma in an 83-year-old Asian female: report of a case with literature review. *Hawaii Dent J.* 1998; 29(3):12–4.
26. Lam KY, Chan AC, Wu PC, Chau KY, Tideman H, Wei W. Desmoplastic variant of ameloblastoma in Chinese patients. *Br J Oral Maxillofac Surg.* 1998; 36(2):129–34. [PubMed: 9643599]
27. Fukushima D, Kobayashi H, Takeda I, Yamane G, Tanaka Y. A case of desmoplastic ameloblastoma of the maxilla. *Bull Tokyo Dent Coll.* 1997; 38(3):223–7. [PubMed: 9566138]
28. Thompson IO, van Rensburg LJ, Phillips VM. Desmoplastic ameloblastoma: correlative histopathology, radiology and CT-MR imaging. *J Oral Pathol Med.* 1996; 25(7):405–10. [PubMed: 8890057]
29. Keszler A, Paparella ML, Dominguez FV. Desmoplastic and non-desmoplastic ameloblastoma: a comparative clinicopathological analysis. *Oral Dis.* 1996; 2(3):228–31. [PubMed: 9081764]
30. Ashman SG, Corio RL, Eisele DW, Murphy MT. Desmoplastic ameloblastoma. A case report and literature review. *Oral Surg Oral Med Oral Pathol.* 1993; 75(4):479–82. [PubMed: 8464613]
31. Ng KH, Siar CH. Desmoplastic variant of ameloblastoma in Malaysians. *Br J Oral Maxillofac Surg.* 1993; 31(5):299–303. [PubMed: 8218083]
32. Kaffe I, Buchner A, Taicher S. Radiologic features of desmoplastic variant of ameloblastoma. *Oral Surg Oral Med Oral Pathol.* 1993; 76(4):525–9. [PubMed: 8233435]
33. Philipsen HP, Ormiston IW, Reichart PA. The desmo- and osteoplastic ameloblastoma. Histologic variant or clinicopathologic entity? Case reports. *Int J Oral Maxillofac Surg.* 1992; 21(6):352–7. [PubMed: 1484205]
34. Tanimoto K, Takata T, Sueti Y, Wada T. A case of desmoplastic variant of a mandibular ameloblastoma. *J Oral Maxillofac Surg.* 1991; 49(1):94–7. [PubMed: 1985191]
35. Higuchi Y, Nakamura N, Ohishi M, Tashiro H. Unusual ameloblastoma with extensive stromal desmoplasia. *J Craniomaxillofac Surg.* 1991; 19(7):323–7. [PubMed: 1752974]
36. Yoshimura Y, Saito H. Desmoplastic variant of ameloblastoma: report of a case and review of the literature. *J Oral Maxillofac Surg.* 1990; 48(11):1231–5. [PubMed: 2213321]
37. Raubenheimer EJ, van Heerden WF, Noffke CE. Infrequent clinicopathological findings in 108 ameloblastomas. *J Oral Pathol Med.* 1995; 24(5):227–32. [PubMed: 7616462]
38. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: biological profile of 3677 cases. *Eur J Cancer B Oral Oncol.* 1995; 31B(2):86–99. [PubMed: 7633291]
39. Takata T, Miyauchi M, Ito H, Ogawa I, Kudo Y, Zhao M, et al. Clinical and histopathological analyses of desmoplastic ameloblastoma. *Pathol Res Pract.* 1999; 195(10):669–75. [PubMed: 10549030]
40. Kramer, IRH., Pingborg, JJ., Shear, M. World Health Organization histological classification of tumors. 2nd. Berlin: Springer; 1991.
41. Mehlich DR, Dahlin DC, Masson JK. Ameloblastoma: a clinicopathologic report. *J Oral Surg.* 1972; 30(1):9–22. [PubMed: 4500335]



**Figure 1.** Histological specimen of a desmoplastic ameloblastoma presenting with irregularly shaped epithelial island, surrounded by narrow zones of loose-structured connective tissue embedded in desmoplastic stroma. (HE Hematoxylin and eosin stain, original magnification  $\times 10$ .)



**Figure 2.** Age and gender distribution of 114 desmoplastic ameloblastomas (A). Distribution of tooth involvement among 61 desmoplastic ameloblastomas (B).



**Figure 3.** Clinical aspect of a desmoplastic ameloblastoma presenting with slow-growing mass with buccal/labial expansion.



**Figure 4.** Radiological features of desmoplastic ameloblastoma. The lesion presents with a “honeycomb appearance” with tooth displacement (A). Radiograph revealing a unilocular radiolucency (arrow) with tooth displacement in the mandible (B). Computed tomography present with an ill-defined mixed density (arrow) of buccal expansion (C).

**Table 1**

Summary of data on reported cases of desmoplastic ameloblastoma.

	No.	%
Patient age (yr)	114	
Mean, 41.9		
Range, 17–83		
<i>Gender (n=114)</i>		
Female	59	51.8
Male	55	48.2
<i>Tumor size (cm) (n = 72)</i>		
≥3.0	36	50.0
<3.0	36	50.0
<i>Tumor location (n = 113)</i>		
Maxilla	51	45.1
Mandible	62	54.9
<i>Tumor borders (n = 75)</i>		
Poorly defined	48	64.0
Well defined	27	36.0
<i>Tumor locularity (n = 73)</i>		
Multilocular	36	49.3
Unilocular	19	26.0
No loculation	18	24.7
<i>Tumor radiographic appearance (n = 90)</i>		
Mixed radiolucent/radiopaque	50	55.6
Radiolucent	40	44.4