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REVIEW ARTICLE

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Clinicopathological characteristics of desmoplastic ameloblastoma: A systematic review

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Abstract

The aim of the present review was to systematically present the clinicopathological data of desmoplastic ameloblastoma (DA) from articles published in the literature. A comprehensive search of the databases (PubMed, Medline, SCOPUS, Web of Science, and Google Scholar) for published articles on DA was conducted. A total of 238 cases were identified and analyzed from 76 published papers. DA showed a slight male predilection (male: female=1.07:1) with a predominance in the fourth and fifth decades of life. Mandibular involvement (52.55%) was most commonly seen with a marked tendency for the anterior region (mandible: 40.9%, maxilla: 48.07%). The size of the lesion ranged from .5 cm to 20.4 cm, with the majority of cases measuring more than 3 cm in size (53.84%). Radiologically, most of the lesions presented mixed radiolucency and radiopacity (62%), and root resorption was observed in only seven cases. The majority of the lesions showed ill-defined margins upon radiographic examination (65.78%). Most of the cases were treated with resection (78.57%), and five of the 10 recurrent cases were treated by enucleation/curettage. DA is characterized by the unique presentation of clinicopathological parameters. It is not possible to comment on its aggressive/recurrent nature and best treatment modality due to inadequate follow-up data.

KEYWORDS

ameloblastoma, desmoplastic ameloblastoma, jaw tumor, odontogenic tumors, systematic review

1 | INTRODUCTION

Desmoplastic ameloblastoma (DA) is a unique variant of ameloblastoma with peculiar clinical, imaging, and histologic features (International Classification of Diseases for Oncology code: 9310/0). It was first reported by Eversole et al. in 1984.[1] Later, in 1987, Waldron et al. considered it as a separate clinicopathological entity.[2] However, the 2017 World Health Organization classification included it as a follicular pattern variant and not as a separate clinicopathological entity.[3] It is also known as ameloblastoma with pronounced desmoplasia, and accounts for approximately 4%-13% of ameloblastomas, which is consistent with the global incidence of 0.9%-12.1% of DA relative to ameloblastomas in general.[4-8] Approximately 238 cases have been reported in the literature to date. This tumor is more commonly seen in the anterior region of the jaw as a mixed radiopaque-radiolucent lesion resembling benign fibro-osseous lesions because of hyalinization seen in the stroma. Histologically, DA is characterized by small nests and strands of "compressed" odontogenic epithelia supported by pronounced collagenized stroma with a tendency of penetration into the surrounding bone. Investigators have observed that recurrence in cases with DA is almost as high as with the conventional ameloblastomas.[9,10] Despite the fact that so many cases of DA have been reported in the scientific literature, the true biologic profile of DA is still not well understood. The aim of the present review was to comprehensively appraise the clinical,

radiological, histopathological, histogenetic, and therapeutic aspects of DA, and thus attempt to further speculate on the possible biologic profile of the tumor to enhance knowledge of this unusual variant.

2 | MATERIALS AND METHODS

2.1 | Search strategy and selection criteria

The present systematic review was conducted according to the guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement. We performed a comprehensive search of the databases (PubMed, Medline, SCOPUS, Web of Science, and Google Scholar), along with cross-references to published articles on DA for eligible studies/case reports published since 1987 up until now. Keywords included a combination of "ameloblastoma", "desmoplastic ameloblastoma", and/or "odontogenic tumors". Additional citations identified through the reference lists of selected references and bibliographical linkages were included in the review. Journals related to oral pathology, oral surgery, and oral medicine were also searched for these keywords. The inclusion criterion was human case reports of DA; cases of peripheral, malignant, metastatic, recurrent, and hybrid cases were excluded from the study. For the analysis, the following clinicohistopathological data were pooled: age, sex, size, site of lesion, duration, histopathologic characteristics, radiographic presentation, immunohistochemical observations, treatment rendered, and recurrence status.

3 | RESULTS

In the present review, a total 238 cases were identified and analyzed from 76 papers[1,2,5-12,16-86] published in the English medical literature (Table 1). The clinicopathological data of 238 cases are summarized in Table 2.

4 | DISCUSSION

4.1 | Clinical features

4.1.1 | Incidence

As it is rare, the incidence of this entity is quite low, and is difficult to determine from the currently-available literature. However, it is possible that the incidence of DA could be escalating because of increased awareness and accurate diagnoses being made. The reported incidence for DA was 8.8% in a case series by Keszler et al.,[11] which is lower than the 12.7% reported by Waldren and el-Mofty.[2] Of the 89 cases of ameloblastomas studied by Takata et al., 7.9% were diagnosed as DA, and only 1.1% were diagnosed as hybrid lesions.[12] In Japan, DA account for 5.3% of all cases of intraosseous ameloblastomas diagnosed before the age of 27 years.[13,14] In an Indian study, ameloblastomas were found to account for 1.18% of the total surgical specimens received over a period of 25 years, with DA accounting for only 2.25% of the various histological variants of ameloblastoma.[15]

4.1.2 | Age and sex distribution

The reported mean age for DA is 38.8 years, with a known male predilection, although Keszler et al. found a higher female prevalence in their case series.[11] The observation of a mean age of 41.11±13.46 years, with an age range of 13-83 years in this series, is slightly lower than the 42.9 years reported by Philipsen et al.,[9] but falls within the age range of the third and fifth decades, as recently reported by Sivapathasundharam et al.[10] A male (88)-to-female (82) ratio of 1.07:1 was observed in our review, which is in accordance with the findings by Waldron and el-Mofty.[2]

4.1.3 | Location and clinical presentation

DA presents itself as a tumor or swelling in a similar fashion to other ameloblastomas. According to our findings, a maximum number of cases reported with a chief complaint of painless swelling of the jawbone. Previously-reported cases of DA have shown this variant to have a strong predilection for the anterior premolar region of the jaw, occurring with equal frequency in both the maxilla and the mandible, in contrast to solid multicystic ameloblastomas.[1,5,10,11,17,22,45] In the present analysis, the site was stated in 137 cases, of which 65 (47.44%) occurred in the maxilla, and 72 (52.55%) in the mandible. Of the 52 cases of maxillary involvement, the anterior region (25/52, 48.07%) showed the most common involvement, followed by the anterioposterior region (21/52, 40.38%) and posterior region (6/52, 11.53%). Similarly, in the case of the mandible, the anterior region (18/44; 40.9%) was the most commonly-affected area compared to the anterio-posterior region (10/44; 22.72%) and the posterior region (16/44; 36.36%). The most common clinical presentation was expansion of the cortical plates. The size of the tumor generally varied between 1.0 and 8.5 cm in diameter,[18] but in the present series, the size of the lesion ranged from 0.5 to 20.4 cm, with 53.84% (35/65) cases found to be more than 3 cm in size.

4.2 | Radiographic examination

Multilocular appearance (n=36, 50%) is the most common radiographic presentation as compared to the unilocular (18, 25%). Eighteen (25%) cases did not show any locularity. Of the 76 cases, the majority of the lesions exhibited ill-defined borders (n=50, 65.78%). According the present review, mixed radiographic appearance was the most common presentation (62/100, 62%). This appearance could be because of hyalinization seen in the stroma of DA. Root resorption was present in eight cases (22.22%), and was absent in 22 cases (61.11%). Twenty-seven cases revealed tooth displacement. The ill-defined borders of DA make high-resolution computed tomography images and magnetic resonance imaging helpful in treatment planning.[19]

4.3 | Histopathology

DA show some variation from the typical core characteristics demonstrated by other histological ameloblastoma subtypes. It has a greater tendency to grow in thin strands, cords, and islands of odontogenic

TABLE 1 Distribution of clinicopathological data for desmoplastic ameloblastomas

Year	Author	No. of cases	Age (years)	Sex	Race	Location	Size	Locularity	Borders	Radiodensity	Tooth resorption	Tooth displacement	Treatment	Follow up
1984	Eversole et al.[1]	3	48	Female	Malaysian	Maxilla P	>3	No locules	I	Mixed	NA	NA	Res	NR2Y
			50	Female	Caucasian	Mandible A+P	<3	Unilocular	I	RL	NA	NA	NA	NA
			47	Female	Black	Mandible A	NA	No locules	I	Mixed	NA	Yes	Res+BG	NA
1987	Waldron & el-Mofty.[2]	14	21-68, 45.5	7 male, 7 female	NA	2 mandible A, 5 mandible P, 6 maxilla A, 1 maxilla P	13:<3, 1:>3	1 unilocular, 5 no locules	5 I, 1 W	4 mixed, 2 RL	NA	NA	NA	2R, 1NR, 11 NA
1990	Yoshimura & Saito[37]	1	36	Female	Japanese	Maxilla A+P	>3	Multilocular	I	Mixed	NA	NA	Res	NR/9Y
1991	Huguchi et al.[6]	3	46	Male	NA	Maxilla A	>3	Multilocular	I	Mixed	No	Yes	NA	NA
			38	Female	NA	Mandible A	>3	Multilocular	W	Mixed	Yes	Yes	NA	NA
			53	Male	NA	Maxilla P	<3	Unilocular	W	Mixed	No	Yes	NA	NA
1991	Tanimoto et al.[39]	1	24	Female	Japanese	Mandible A+P	>3	NA	I	RL	No	Yes	Res	NR1Y
1992	Philispen et al.[86]	2	21	Male	Chinese	Maxilla A+P	>3	Multilocular	I	RL	Yes	Yes	Res+BG	NA
			53	Male	Chinese	Maxilla A+P	>3	Multilocular	I	RL	Yes	Yes	Res	NA
1993	Kaffe et al.[41]	1	41	Male	NA	Maxilla A+P	>3	No locules	I	Mixed	NA	NA	Res	NR3Y
1993	Ng et al.[8]	17	21	Female	Chinese	Mandible A	<3	NA	I	RL	NA	NA	Res	NA
			25	Male	Chinese	Maxilla	NA	NA	NA	Mixed	NA	NA	Res	NA
			25	Male	Malaysian	Mandible A	NA	Multilocular	NA	RL	NA	NA	Res	NA
			27	Female	Chinese	Mandible A	NA	NA	NA	Mixed	NA	NA	NA	NA
			29	Female	Kadazan	Mandible A+P	NA	Multilocular	NA	Mixed	NA	NA	Res	NA
			32	Female	Indian	Maxilla A	NA	NA	NA	NA	NA	NA	NA	NA
			33	Male	Malaysian	Maxilla A	3	NA	NA	Mixed	NA	NA	NA	NA
			33	Female	Malaysian	Maxilla A+P	NA	NA	I	NA	NA	NA	Res	NR1Y
			38	Male	Malaysian	Maxilla A	<3	NA	NA	NA	NA	NA	Res	NA
			40	Female	Chinese	Mandible A	NA	Multilocular	NA	RL	NA	NA	NA	NA
			41	Female	Malaysian	Maxilla A	NA	NA	NA	NA	NA	NA	Res	NA

(Continues)

TABLE 1 (Continued)

Year	Author	No. of cases	Age (years)	Sex	Race	Location	Size	Locularity	Borders	Radiodensity	Tooth resorption	Tooth displacement	Treatment	Follow up
			42	Female	Chinese	Mandible P	NA	NA	NA	NA	NA	NA	NA	NA
			43	Female	Malay	Mandible A	NA	NA	NA	NA	NA	NA	NA	NA
			43	Female	Chin	Mandible A	NA	Multilocular	NA	RL	NA	NA	NA	NA
			44	Female	Indian	Mandible A	<3	NA	NA	NA	NA	NA	Res	R4Y
			46	Female	Indian	Maxilla	NA	NA	NA	Mixed	NA	NA	Res	NR1Y
			60	Male	Chinese	Mandible A	NA	Multilocular	NA	RL	NA	NA	NA	NA
1993	Ashman et al.[40]	1	53	Male	Black	Mandible A+P	>3	No locules	W	Mixed	NA	Yes	Res	NR9M
1995	Raubenheimer et al.[7]	1	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
1996	Keszler et al.[11]	14	19-62, 37.8 in 13, 1 NA	11 female, 2 male, 1 NA	NA	2 maxilla, 10 mandible, 1 NA	NA	4 unilocular, 5 multilocular	4 W	9/RL, 3/Mixed	NA	NA	5 res, 9 cur	3 R, 11 NR
1996	Thompon et al.[19]	1	31	Female	Black	Maxilla A+P	>3	No locules	I	Mixed	Yes	Yes	Res+BG	NR3Y
1996	Lo et al.[42]	1	NA	NA	NA	Mandible	NA	NA	NA	NA	NA	NA	NA	NA
1997	Fukushima et al.[43]	1	70	Male	NA	Maxilla P	>3	No locules	I	Mixed	NA	Yes	Res	NR18M
1998	Lam et al.[5]	5	64	Male	Chinese	Maxilla A+P	>3	No locules	I	RL	NA	NA	Res	R2Y
			18	Male	Chinese	Maxilla A	>3	Multilocular	I	RL	NA	NA	Res	NR53M
			68	Female	Chinese	Maxilla P	<3	Multilocular	I	RL	NA	NA	Cur	R74M
			37	Female	Chinese	Mandible A	<3	Multilocular	I	RL	NA	NA	Res	NR77M
			37	Male	Chinese	Mandible A	<3	No locules	I	RL	NA	NA	Res	NR32M
1998	Lee et al.[44]	1	83	Female	Asian	Mandible A	<3	Multilocular	W	Mixed	NA	Yes	Res	NR
1998	Sakashita et al.[70]	1	60	Female	Japanese	Maxilla A	>3	Multilocular	I	RL	NA	NA	Res	NR
1998	Ludvikoya et al.[71]	2	50	Male	NA	Mandible P	>3	No locules	I	RL	NA	NA	Res	NR
			42	Female	NA	Mandible P	<3	No locules	I	RL	NA	NA	Res	NR3Y
1999	Kawai et al.[21]	1	56	Male	Japanese	Mandible P	NA	Unilocular	W	RL	NA	NA	Res	NA
1999	Takata et al.[45]	6	53	Male	Japanese	Maxilla A	>3	No locules	W	RL	Yes	NA	Res	NR
			33	Female	Japanese	Maxilla A	3	Multilocular	I	Mixed	No	NA	Cur	NR
			51	Male	Japanese	Maxilla A+P	<3	Unilocular	W	RL	No	NA	Res	NR
			52	Male	Japanese	Maxilla A+P	>3	Multilocular	I	Mixed	No	NA	Res	NR

(Continues)

TABLE 1 (Continued)

Year	Author	No. of cases	Age (years)	Sex	Race	Location	Size	Locularity	Borders	Radiodensity	Tooth resorption	Tooth displacement	Treatment	Follow up
			54	Male	Japanese	Mandible A+P	>3	No locules	W	Mixed	No	NA	Res	NR
			17	Male	Japanese	Mandible A+P	<3	Multilocular	I	Mixed	No	NA	Cur	R4
2000	Louis et al.[72]	1	33	Male	Black	Maxilla A+P	<3	NA	I	Mixed	No	Yes	Res+BG	NR18M
2000	Takata et al.[12]	6	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2000	Saran et al.[46]	1	25	Female	Indian	Maxilla A+P	<3	Unilocular	W	NA	NA	NA	Res	NR10M
2001	Kishino et al.[22]	9	41	Male	Japanese	Maxilla A+P	>3	Multilocular	W	RL	No	NA	Res	NR17Y
			28	Male	Japanese	Maxilla A	>3	NA	I	Mixed	No	NA	Res	NR72M
			57	Male	Japanese	Maxilla A	<3	NA	W	RL	No	NA	Res	NR8Y
			58	Female	Japanese	Maxilla A	3	Unilocular	W	RL	No	NA	Cur	NR16Y
			17	Male	Japanese	Mandible P	<3	Unilocular	W	RL	No	NA	Cur	NR5Y
			50	Male	Japanese	Mandible P	>3	NA	I	Mixed	No	NA	Res	NR6Y
			51	Male	Japanese	Mandible P	>3	NA	I	Mixed	No	NA	Res+BG	NR23Y
			47	Male	Japanese	Mandible A+P	>3	NA	I	Mixed	No	NA	Res+BG	NR20Y
			42	Male	Japanese	Mandible P	>3	NA	I	Mixed	No	NA	NA	NA
2001	Philipsen et al.[9]	1	42	Female	Caucasian	Mandible A	NA	Unilocular	I	Mixed	NA	Yes	Res+BG	NR36M
2001	Kumamoto et al.[24]	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2002	Mintz & Velez[47]	2	52	Male	NA	Maxilla A+P	NA	Multilocular	I	Mixed	NA	Yes	Res	NR
			51	Female	NA	Mandible P	NA	Multilocular	I	Mixed	NA	Yes	Res+BG	NR1Y
2002	Manuel et al.[73]	1	20	Female	NA	Maxilla A	<3	Unilocular	W	Mixed	No	Yes	Res	NR22M
2002	Iida et al.[74]	1	52	Male	Japanese	Maxilla A+P	NA	Multilocular	I	Mixed	NA	Yes	Res	NR
2002	Fukumashi et al.[33]	3	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2002	Beckely et al.[75]	1	31	Male	Hispanic	Mandible A+P	<3	No locules	I	Mixed	No	Yes	Res+BG	NR19M
2003	Durmus et al.[49]	1	68	Male	Turkish	Mandible A	<3	Unilocular	W	RL	Yes	NA	Cur	NR32M
2003	Maresi et al.[48]	1	62	Male	NA	Mandible	NA	NA	NA	Mixed	NA	NA	NA	NA
2004	Piilai et al.[76]	1	24	Female	Indian	Maxilla A+P	<3	Multilocular	I	RL	No	Yes	Cur+BG	R2M
2004	Kumamoto & Ooya[25]	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2005	Kumamoto et al.[26]	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA

(Continues)

TABLE 1 (Continued)

Year	Author	No. of cases	Age (years)	Sex	Race	Location	Size	Locularity	Borders	Radiodensity	Tooth resorption	Tooth displacement	Treatment	Follow up
2005	Hirota et al.[77]	1	17	Female	Japanese	Maxilla P	NA	Multilocular	W	Mixed	NA	Yes	Res	NR7Y
2006	Desai et al.[50]	1	32	Male	Indian	Mandible P	>3	Unilocular	W	RL	NA	NA	Cur	NR2Y
2006	Adebeyi et al.[51]	4	36.5±4.4	4 male	NA	4 mandible	NA	NA	NA	NA	NA	NA	NA	NA
2007	Shashikanth et al.[78]	1	32	Female	Indian	Maxilla A	<3	Multilocular	I	Mixed	Yes	Yes	Res	NR1Y
2007	Kumamoto & Ooya[29]	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2007	Sivapathasundaram et al.[10]	4	25	Male	Indian	Maxilla A+P	NA	Unilocular	W	Mixed	No	Yes	Res	NA
			40	Male	Indian	Maxilla A+P	NA	Unilocular	W	Mixed	No	No	Res	NA
			30	Female	Indian	Maxilla A+P	NA	NA	I	Mixed	No	No	Res	NA
			32	Female	Indian	Maxilla A+P	3	Multilocular	I	Mixed	Yes	Yes	Res	NR1Y
2008	Curran & Byerly[79]	1	56	Male	Black	Mandible P	<3	NA	W	NA	No	NA	Res	NR51M
2008	Smullin et al.[80]	1	44	Female	NA	Maxilla A+P	<3	NA	W	NA	NA	NA	Res	NR1Y
2008	Punnya et al.[52]	1	32	Male	Indian	Maxilla A	>3	NA	I	Mixed	NA	NA	Cur	NA
2008	Kummamoto & Ooya[30]	3	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2008	Ide et al.[53]	3	52	Male	NA	Maxilla A	NA	NA	NA	Mixed	NA	NA	NA	NA
			31	Male	NA	Maxilla A	NA	NA	NA	Mixed	NA	NA	NA	NA
			36	Male	NA	Maxilla A	NA	NA	NA	Mixed	NA	NA	NA	NA
2008	Sathi et al.[54]	4	50	Male	NA	Mandible	NA	NA	NA	NA	NA	NA	NA	NA
			38	Male	NA	Mandible	NA	NA	NA	NA	NA	NA	NA	NA
			33	Female	NA	Maxilla	NA	NA	NA	NA	NA	NA	NA	NA
			59	Male	NA	Maxilla	NA	NA	NA	NA	NA	NA	NA	NA
2009	Bologna-Molina et al.[27]	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2010	Fulco et al.[55]	3	34.3	NA	NA	3 mandible	3.5	NA	NA	NA	NA	NA	NA	NA
2010	Amaral et al.[56]	1	25	Male	NA	Mandible	>3	NA	NA	Mixed	NA	NA	Res	NR12M
2010	De Medeiros et al.[23]	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2011	Sheikh et al.[57]	1	45	Female	NA	Maxilla A	>3	Na	I	Mixed	NA	Yes	Res	NA
2011	Effiom & Odukoya[58]	17	NA	10 male, 7 female	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA

(Continues)

TABLE 1 (Continued)

Year	Author	No. of cases	Age (years)	Sex	Race	Location	Size	Locularity	Borders	Radiodensity	Tooth resorption	Tooth displacement	Treatment	Follow up
2011	Kato et al.[59]	1	29	Female	NA	Maxilla	NA	NA	NA	Mixed	NA	NA	NA	NA
2011	Katsura et al.[60]	1	45	Female	NA	Maxilla A+P	NA	Multilocular	NA	Mixed	NA	NA	Res	NA
2012	Medeiros et al.[61]	1	NA	NA	NA	Mandible	NA	NA	NA	NA	NA	NA	NA	NA
2012	Siar et al.[32]	8	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2012	Siar et al.[85]	22	44.8±13.7	13 male, 9 female	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2012	Cervelli et al.[62]	1	13	Male	Indian	Mandible	>3	NA	NA	Mixed	NA	NA	NA	NA
2013	Bologna-Molina et al.[28]	5	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2013	Nair et al.[16]	1	60	Male	NA	Mandible	NA	Multilocular	NA	Mixed	NA	NA	Res	NA
2013	Belgaumi et al.[38]	1	47	Female	NA	Maxilla	NA	Multilocular	I	Mixed	NA	NA	Res	NA
2013	Savithri et al.[20]	1	26	Female	NA	Maxilla	NA	NA	NA	Mixed	NA	NA	NA	NA
2014	Ramesh et al.[63]	1	45	Female	NA	Maxilla	>3	NA	NA	Mixed	NA	NA	NA	NA
2014	Majumdar et al.[64]	1	55	Male	NA	Mandible	NA	NA	NA	Mixed	NA	NA	NA	NA
2014	Figueiredo et al.[65]	1	49	Female	NA	Maxilla	NA	Multilocular	NA	NA	NA	NA	NA	NA
2014	Filizzola et al.[66]	2	43	1 male, 1 female	NA	1 maxilla, 1 mandible	NA	NA	NA	NA	NA	NA	NA	NA
2014	Selvamani et al.[67]	1	34	Female	NA	Mandible	NA	NA	NA	NA	NA	NA	NA	NA
2015	Bologna-Molina et al.[31]	2	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2015	Diniz et al.[68]	2	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2015	Khalil et al.[69]	1	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2016	Milman et al.[81]	6	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
2016	Lamichhane et al.[82]	1	43	Female	Chinese	Mandible A	>3	Multilocular	I	Mixed	No	Yes	Res	NR10M
2016	Wankhede & Dive[83]	1	17	Female	Indian	Mandible A+P	>3	NA	I	NA	No	Yes	NA	NA
2016	Imran et al.[84]	1	44	Male	Indian	Mandible A+P	>3	NA	I	Mixed	No	Yes	Res	NA

(Continues)

TABLE 1 (Continued)

Year	Author	No. of cases	Age (years)	Sex	Race	Location	Size	Locularity	Borders	Radiodensity	Tooth resorption	Tooth displacement	Treatment	Follow up
	Total	238	41.11±13.46	88 male, 82 female, 68 NA		13 maxilla, 25 maxilla A, 6 maxilla P, 21 maxilla A+P, 28 mandible, 18 mandible A, 16 mandible P, 10 mandible A+P, 101 NA	35 >3 30 ≤3 173 NA	18 unilocular, 36 multilocular, 18 no locules 166 NA	26 W, 50 I, 162 NA	62 mixed, 38 RL, 138 NA	8 yes, 28 no, 102 NA	27 yes, 2 no, 109 NA	66 res, 18 cur, 154 NA	10 rec

A, anterior; BG, bone graft; Cur, curettage; I, Ill-defined; NA, not applicable; NR, non-recurrent; P, posterior; Rec, recurrent; Res, resection; W, well-defined.

TABLE 2 Summary of clinicopathological data for desmoplastic ameloblastomas

Parameter	Values	%
Papers	76	
Total cases	238	
Age		
Mean	41.11±13.46	
Range	13-83 years	
Sex (n=170)		
Male	88	51.76
Female	82	48.23
Jaw (n=137)		
Mandible	72	52.55
Maxilla	65	47.44
Mandibular Region (n=44)		
Anterior	18	40.9
Anterior+posterior	10	22.72
Posterior	16	36.36
Maxillary region (n=52)		
Anterior	25	48.07
Anterior+posterior	21	40.38
Posterior	6	11.53
Size (n=65)	0.5-20.4 cm, 35 cases ≥3 cm	
Radiographic features		
Locularity (n=72)		
Multilocular	36	50.00
Unilocular	18	25.00
No locules	18	25.00
Borders (n=76)		
Ill defined	50	65.78
Well defined	26	34.21
Radiodensity (n=100)		
Mixed	62	62
Radiolucent	38	38
Root resorption (n=30)	8	22.22
Tooth displacement	27 cases	
Treatment (n=84)		
Resection	66	78.57
Curettage	18	21.42
Recurrence	10 (5 curettage)	

epithelia. The stromal component dominates, compressing the odontogenic epithelial components. The epithelial tumor islands are irregular or bizarre in shape, with a pointed or stellate appearance (kite shaped or animal configurations).[20] The epithelial cells at the periphery of the islands are cuboidal, with few hyperchromatic nuclei. Columnar cells with nuclear polarity are rarely evident. The islands have a swirled, hypercellular center, with spindle-shaped or squamous epithelial cells. Intrafollicular microcysts might occur centrally.[10] Connective tissue

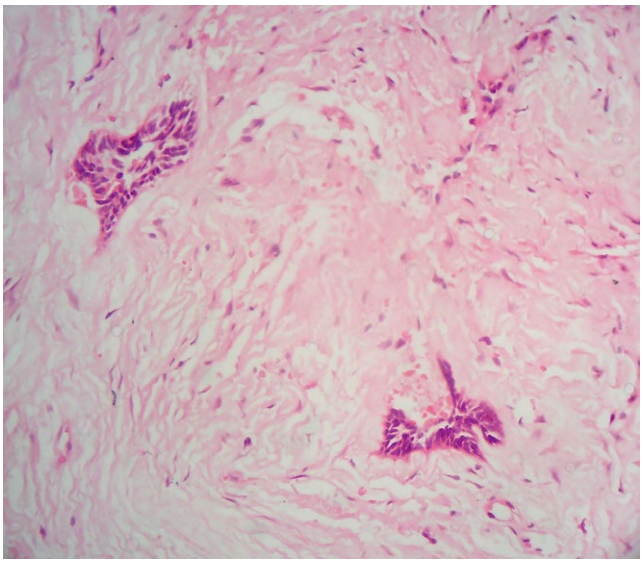

FIGURE 1 Photomicrograph of desmoplastic ameloblastoma showing highly-collagenous stroma and small compressed odontogenic follicles. (hematoxylin-eosin stain, total magnification ×400)

stroma shows desmoplasia with hyalinization. (Figure 1). Myxoid changes of the juxta-epithelial stroma are often found.[38] Formation of metaplastic osteoid trabeculae (osteoplasia) could be present.[38] DA has ill-defined borders, suggesting its infiltrating process and aggressive biologic behavior.[20]

Kawai et al. reported a unique case of DA associated with a large cystic lesion.[21] The tumor epithelial cells, however, were not similar to the epithelial cells lining the lumen of the cyst. It was proposed that the cyst was formed by cystic degeneration of the tumor epithelial nests, because the epithelial cells lining the cyst wall consisted of "degenerated" epithelial cells.

4.4 | Immunohistochemistry

Kishino et al.[22] observed that some fibers of the DA tumor stroma stained brilliant purple with potassium monopersulfate-aldehyde fuchsin staining. These fibers continued from the oxytalan fibers of the periodontal membrane in the specimen involving a tooth. It was presumed that DA might have developed in the periodontal membrane of the related tooth.

A study carried out by Takata et al. showed transforming growth factor- β (TGF-β) positivity in five cases (2[+++] and 3[++]), and negativity in only one case of DA.[12] TGF-β is one of the most potent local factors for modulating extracellular matrix formation, and its presence in DA is responsible for increased matrix formation. Collagen IV was found to be positive in five cases (2[++] and 3[+]) and negative in one. The desmoplastic stroma of DA has been reported to show a strong positive reaction for collagen type VI (ruling out the scar tissue), the strong immunopositive reaction for fibronectin and type 1 collagen, and immunonegativity for tenascin. Moderate-to-intense immunolabeling for type I collagen has been observed in the stroma of all solid DA.[23] Fibronectin is a glycoprotein that possesses an important signaling function in cell adhesion and migration, whereas tenascin is a glycoprotein synthesized at specific time points, and its highest expression is observed during cell migration, in active areas of epithelial-mesenchymal interactions, and in neoplastic stroma. Type I collagen is found throughout the connective tissue, and is one of the most abundant components of the interstitial extracellular matrix, being highly resistant to proteases due to its unique supercoiled triple helix structure. Type I collagen varied expression in DA might suggest less invasive behavior.

Kumamoto found that all DA epithelial islands were diffusely positive for Fas and caspase 3 expression, while weakly reactive for Fas-L.[24] In another study, he discovered four cases positive for survivin (1[+], 1[++] and 2[+++]), and four for X-linked inhibitor of apoptosis protein expression (2[++] and 2[+++]).[25] DA express survivin reactivity, suggesting that these inhibitors of apoptosis proteins family proteins contribute to biologic properties of epithelial odontogenic tumors, as well as to cellular regulation during tooth development. These results suggest that apoptosis-related factors might be associated with the oncogenesis and cytodifferentiation of DA.

An immunohistochemistry study by Kumamoto et al. found four DA cases with positivity for p63, of which one had a moderately-positive expression in peripheral and central cells, while in three cases, it was strongly positive in most of the epithelial cells.[26] p73 was positive in four cases; one case exhibited mild expression in peripheral cells, while three cases showed strong positivity in peripheral and central cells. The expression of p63 and p73 suggests that these p53 homologs play a role in the differentiation and proliferation of DA odontogenic epithelial cells. Variations of predominantly-expressed isoforms suggest that p63 and p73 might function differentially in odontogenic tissues. According to Bologna-Molina et al. , DA has a lower proliferation index (Ki-67 and proliferating cell nuclear antigen), as well as the highest levels of syndecan-1.[27] There were no apparent differences in the distribution of syndecan-1 immunoexpression among basal and stellate reticulum-like cells. The high expression of syndecan-1 in DA could be associated with better cell-cell and cell-extracellular matrix adhesion in epithelial ameloblastic zones (islands), suggesting less aggressive and invasive behavior of this neoplasia, and could explain, to some degree, the unique clinicopathological features of this neoplasia, which also involves a very slow growing lesion that induces desmoplasia. In another study by Bologna-Molina et al.,[28] higher expression of Ki-67 and proliferating cell nuclear antigen was found compared to ameloblastic carcinoma. However, no differences were reported in the expression of these markers between DA and other variants of ameloblastomas. A study on platelet-derived growth factor showed expression for insulin growth factor (IGF) - 1 (+++), IGF-II (+++), IGF-I receptor (+++), platelet-derived growth factor (PDGF) - A-chain (+), PDGF B-chain (+), and PDGF α-receptor.[29] These results suggest that IGF, PDGF and their receptor contributes to cell proliferation and survival thus contributing to the intra-osseous progression of DA.

According to Kumamoto and Ooya. DA express immunoreactivity for the Bcl-2 homology domain 3 (BH3)-only proteins in most tumor cells.[30] BH3-only proteins have a role in apoptotic cell death of normal and neoplastic odontogenic epithelia. The distinctive expression patterns of these BH3-only proteins in ameloblastoma variants suggest

that the BH3-only proteins might be involved in the tumor cell differentiation of DA. Bologna-Molina et al. also observed that all of the DA exhibited the highest expression of glypican-1,[31] suggesting its possible role in the desmoplasia that characterizes DA. According to Siar et al., DA demonstrated strong expression for canonical Wnts-1, -8b, and -10a, and moderately for Wnts-3 and -8a.[32] Non-canonical Wnt proteins were not expressed in all the eight DA cases. These results suggest that the canonical Wnt pathway is most likely the main transduction pathway, and that Wnt-1 might be the key signaling molecule involved in DA tumorigenesis.

DA demonstrated diminished odontogenic characteristics, as basal cells are cytokeratin (CK) 19 negative (a marker for odontogenic epithelia). Immunostaining for CK in the basal layer revealed klotho (KL) 1, CK8, and protein kinase (PK) positivity. In the suprabasal layer, interleukin, KL1, CK19, CK8, CK13, and PK were positive in the cytoplasm.[33] DA illustrated fibronectin immunoexpression throughout the tumor stroma, but no expression of the same at the epithelial-mesenchymal interface, which might be related to the lack of similarities between pre-ameloblasts and the peripheral epithelial cells of DA.[23]

4.5 | Differential diagnoses

Histopathological features of DA involve two characteristics: extensive stromal desmoplasia and small tumor nests of odontogenic epithelia scattered in the stroma.[1] From these findings, other tumors that should be histologically differentiated from this type of ameloblastoma include ameloblastic fibroma, odontogenic fibroma, and squamous odontogenic tumor.[34,35]

Its unique radiographic appearance resembles a mixed radiolucent-radiopaque lesion, unlike the strictly radiolucent quality of other ameloblastomas. Therefore, it is usually a surprise diagnosis from a differential list composed mostly of fibro-osseous diseases and odontogenic cysts and tumors that are characteristically radiolucent-radiopaque. These include ossifying fibromas, fibrous dysplasia, osteoblastomas, osteosarcomas, calcifying epithelial odontogenic tumors, and calcifying odontogenic cysts.

4.6 | Treatment

Although various treatments have been proposed, complete resection is recommended for DA to avoid recurrence, because of the lack of distinct borders between the tumor and normal tissues in many cases. It has been proven that enucleation or curettage of DA results in recurrence, but small DA lesions can easily be enucleated in toto from the intraosseous bed. It was rightly said by Marx and Stein that "the best chemotherapy for odontogenic tumors is a jar of formalin", which is also applicable for this rare variant.[36] Present knowledge leads to the recommendation to apply the same treatment modality for this variant, as DA is otherwise identical to conventional ameloblastomas. The most common surgical intervention followed in various cases in this review was resection (78.57%, 66/84) followed by partial maxillectomy or mandibulectomy. Ten recurrent cases of DA have been reported to date. Five of these cases were treated by enucleation or curettage.[2,5,37] The duration of the recurrence ranged from 2 to 6 years. In the majority of the cases (81/238, 34.03%), the follow-up data were not available. Therefore, we are unable to comment on the recurrent nature and best treatment modality for this lesion. The prognosis cannot be established because of a lack of documentation, but complete resolution had been shown to occur with resection.[8]

5 | CONCLUSION

In conclusion, DA showed distinct male predilection with a predominance in the fourth and fifth decades of life, and a slight predilection for the mandible compared to the maxilla, with painless swelling as the most common presentation. The mixed radiolucent and radiopaque appearance of DA distinguishes it from conventional ameloblastomas. This feature makes fibro-osseous lesions the most common differential diagnosis for DA. Histologically, severe desmoplasia is the most prominent, feature with interspersed epithelial cells arranged in compressed islands or strands. Due to the unavailability of follow-up data in many cases, we are unable to comment on the recurrent nature and optimum treatment modality for this neoplasm.

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