

Journal section: *Oral Medicine and Pathology*
 Publication Types: *Case Report*

doi:10.4317/medoral.15.e846

Peripheral desmoplastic ameloblastoma: Histopathological and immunohistochemical profile of a case

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Bologna-Molina R, Mosqueda-Taylor A, de Almeida-Oslei P, Toral-Rizo V, Martínez-Mata G. Peripheral desmoplastic ameloblastoma: Histopathological and immunohistochemical profile of a case. *Med Oral Patol Oral Cir Bucal*. 2010 Nov 1;15 (6):e846-9.
<http://www.medicinaoral.com/medoralfree01/v15i6/medoralv15i6p846.pdf>

Received: 21/10/2009

Accepted: 21/02/2010

Article Number: 3247 <http://www.medicinaoral.com/>
 © Medicina Oral S. L. C.I.F. B 96689336 - pISSN 1698-4447 - eISSN: 1698-6946
 eMail: medicina@medicinaoral.com

Indexed in:

- SCI EXPANDED
- JOURNAL CITATION REPORTS
- Index Medicus / MEDLINE / PubMed
- EMBASE, Excerpta Medica
- SCOPUS
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Abstract

In this study we present a rare case of peripheral desmoplastic ameloblastoma and discuss its clinical features, histopathology, and immunohistochemical profile.

This article reports a new case of this unusual neoplasm in a 66 year-old woman in which the main complaint was an asymptomatic swelling located in the right body of mandible. Histopathological findings were similar to the two previously reported cases of this tumor. Positive immunohistochemical stain for laminin V and type IV collagen suggests an inductive effect of the epithelium over the stroma while the low index of p53 protein and Ki-67 expression in epithelium and stromal cells, as well as CD138 uniform positive-stain in epithelial cells, support the benign biological behavior of this lesion. Including this new case, currently there are only three reports of this rare neoplasm. Reports of new cases of peripheral desmoplastic ameloblastoma are necessary for a better understanding of the origin and behavior of this particular subtype of ameloblastoma.

Key words: *Ameloblastoma, peripheral, desmoplastic, immunohistochemistry.*

Introduction

Peripheral odontogenic tumors (POT) are uncommon lesions, accounting from 1.24% to 3.4% of all odontogenic tumors (1,2). According to previous reports, the most frequent POT are peripheral odontogenic fibroma (POF) and peripheral ameloblastoma (PA) with 40%

and 36% respectively (1). Contrary to POF its intraosseous counterpart (central odontogenic fibroma) has a higher potential of aggressiveness. On the other hand, it is well recognized the high rate of recurrence observed in central solid types of ameloblastomas, which far exceeds what has been reported for PA. Desmoplastic

ameloblastoma (DA) was initially defined as a central lesion but recently, two cases of peripheral desmoplastic ameloblastoma (PDA) were reported (3,4). In this study we present an additional case of this unusual neoplasm, and discuss its clinical features, histopathology, and immunohistochemical profile.

Case Report

A 66-year-old woman presented for diagnosis and management of an asymptomatic swelling located in the right body of mandible. Clinical examination revealed an exophytic mass of 2 cm of diameter, firm to palpation. An excisional biopsy was performed and the specimen was sent to histopathological study. No evidence of bone resorption in the area was observed during surgery. Five μm sections of paraffin-embedded tissue were

stained with H&E stain and immunohistochemistry was performed using avidin-biotin complex technique. (Table 1) shows antibodies, dilutions and sources of antibodies used in this case.

Microscopical examination disclosed a lesion constituted by irregular islands of odontogenic epithelium embedded in a collagenized stroma. Epithelial islands were arranged in nest, narrow cords and more solid sheets of odontogenic epithelium (Fig. 1a). Columnar cells with reverse polarization were not a predominant feature. Some islands were hypercellular with central squamous metaplasia and fusiform cells (Fig. 1b). Multiple microcystic degeneration was observed (Fig. 1a). Stroma was densely collagenized but myxoid changes were found adjacent to epithelial islands.

Table 1. Antibodies, sources, clones and dilutions applied in present case.

Antibody	Source/clone	Dilution
Cytokeratin cocktail	Dako™, AE1/AE3	1:500
CK1	Novocastra™,34 β B4	1:200
CK5	Novocastra™,XM26	1:400
CK6	Novocastra™,LHK6B	1:400
CK8	Dako™,OV-TL12/30	1:400
CK10	Dako™,DE-K-10	1:200
CK13	Novocastra™,KS-1A3	1:400
CK14	Novocastra™,NCL-L-LL002	1:400
Ck16	Novocastra™,LL025	1:400
CK18	Dako™,DC10	1:400
CK19	Dako™,RCK108	1:400
CK20	Dako™,K _s 20.8	1:200
Vimentin	Dako™,Vim 3B4	1:200
α SMA	Dako™,1A4	1:400
p53	Dako™,DO-7	1:200
Ki-67	Dako™,MIB-1	1:100
S-100	Dako™	1:12,000
CD138	Dako™,MI15	1:100
Collagen IV	Dako™,CIV 22	1:200
Laminin	Laminin	1:200

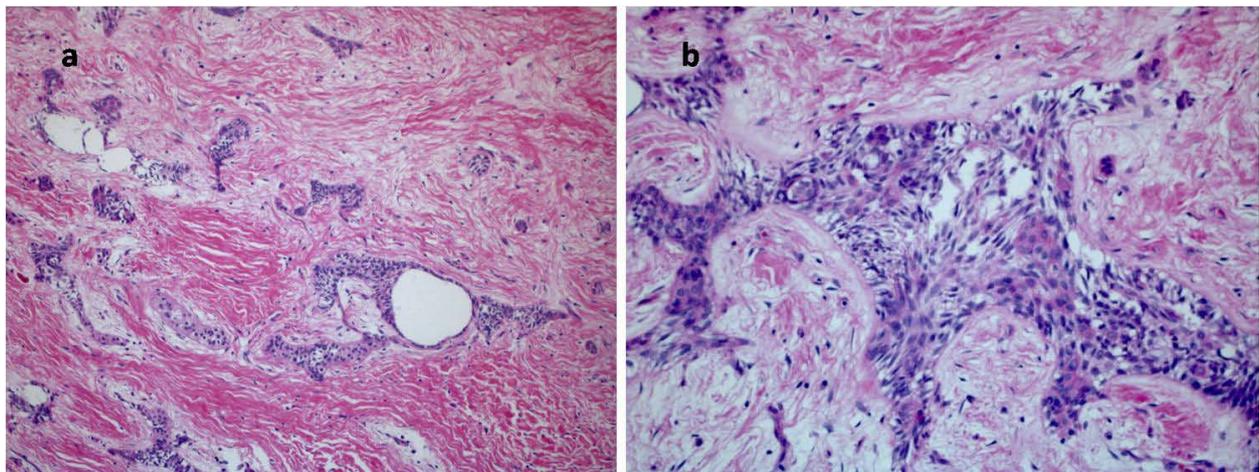


Fig. 1. Photomicrographs showing (a) neoplastic odontogenic epithelium associated to a densely collagenized stroma with multiple microcystic degeneration, (b) note the central squamous metaplasia (H&E stain, original magnification x200 and x400).

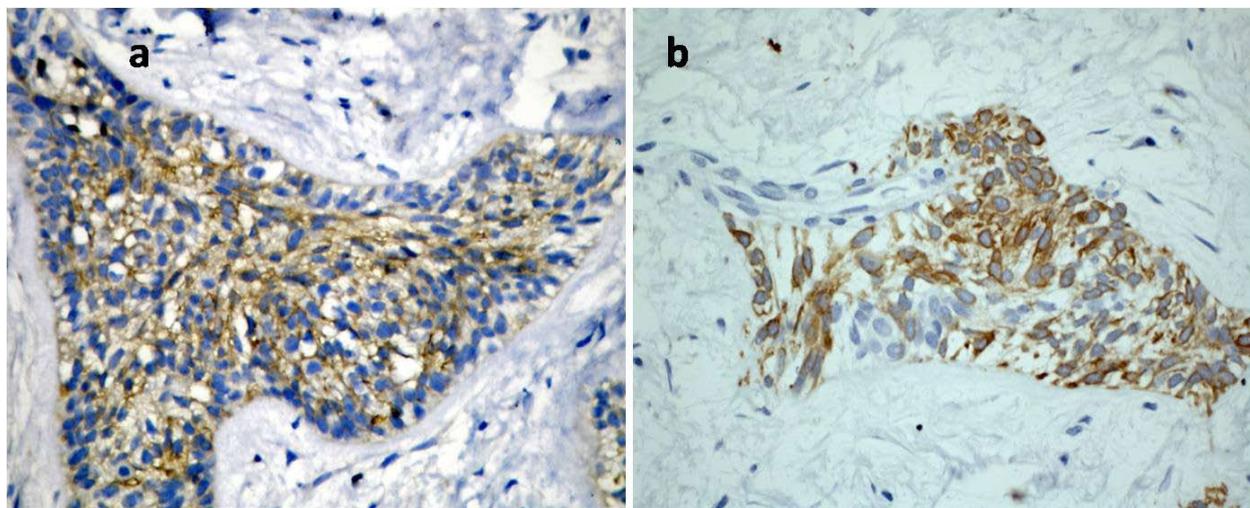


Fig. 2. (a) High expression of syndecan-1 (CD 138) in the ameloblastic epithelium, (b) Positive stain in suprabasal and central epithelial cells for CK14 in PDA (CD138 and CK14, original magnification x400).

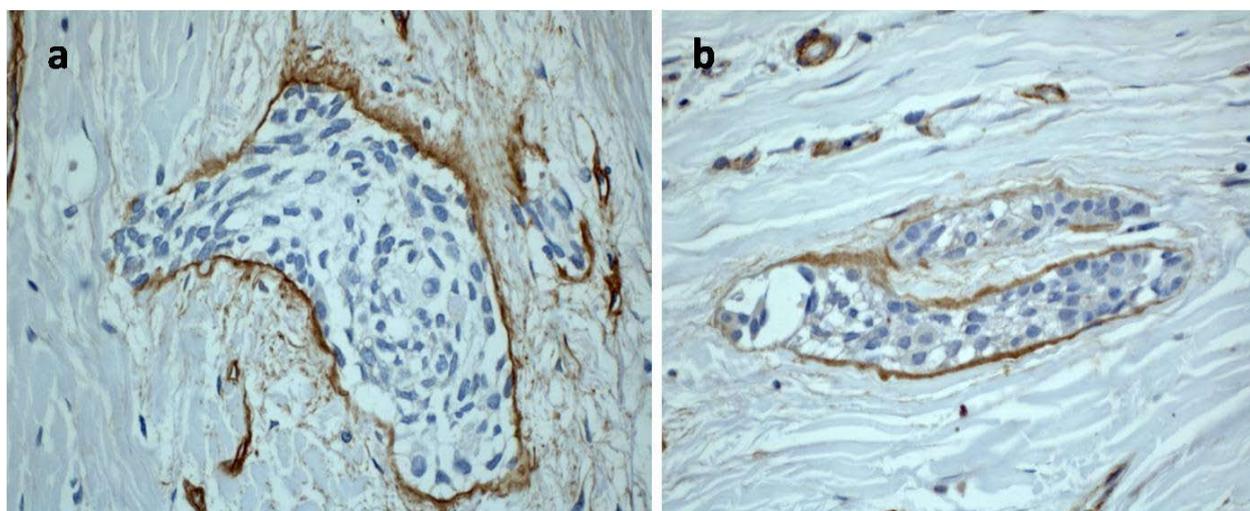


Fig. 3. Expression of Laminin V (a) and type IV Collagen (b) antibodies in basal lamina of PDA (original magnification, x400).

Odontogenic epithelial islands were positive for cytokeratin cocktail AE1/AE3, 34 β E12 and CD-138 (syndecan-1) (Fig. 2a). CK5 was positive in basal and suprabasal cells, while some suprabasal and central cells were positive to CK14 (Fig. 2b). Focal stain for CK13 and S-100 protein were observed in squamous central cells. Cytokeratins 1, 6, 8, 10, 16, 18, 19 and 20 were not expressed. Stromal cells were positive for vimentin; antibodies for muscle differentiation (HHF-35 and SMA), Bcl-2 and p21 were also negative. p53 and Ki-67 index were lower than 1% in epithelial islands and negative in stromal cells. On the other hand, basal membrane of epithelial tissue was intensely positive for laminin V and type IV collagen (Fig. 3).

After four year of follow-up there are no signs of recurrence.

Discussion

Since its original description by Eversole et al. (5) DA is considered a rare variant of Central Ameloblastoma (CA). CA represents a frequent odontogenic tumor accounting for 11-60% off all central odontogenic tumors (6,7) and DA accounts for 5.3% of all CA (8). To date, 145 cases of DA have been reported in English literature; (9) of these, only two can be considered as peripheral (3,4). It is well known that there are histological and biological behavioral differences among DA and the other types of CA. For example, DA poses a lower recurrence rate as compared to CA. The histological findings in our case are in accordance with previous reports of PDA and DA (3,10,11). Sivapathasundharam et al. (9) suggest that epithelial changes can be related to pres-

sure of stroma on epithelial tissue or may be associated to changes in maturation of a preexistent CA. However, Kishino et al. (8) and Kawai et al. (12) suggest that DA derives from periodontal ligament, and based their observation in the presence of oxytalan fibers in stroma of this neoplasm. In our case, the lack of continuity of the lesion with surface epithelium and the negative stain of PDA for CK19 in this case is in accordance with the observations of Philipsen et al. (11), who suggest that the origin of this neoplasm can be remnants of dental lamina located in gingival tissues. Immunohistochemical expression of CK5 in basal/suprabasal cells and CK14 in suprabasal/central cells in the present case offer evidence of maturation of the epithelial component; low index of p53 protein and Ki-67 expression in epithelium and stromal cells of PDA, as well as CD138 uniform positive-stain in epithelial cells, support the benign biological behavior of the lesion, as has been reported previously in their intraosseous counterpart. (13) High expression of syndecan-1 in DA could be associated with a better cell-cell and cell-extracellular matrix adhesion in epithelial ameloblastic zones (islands), suggesting a less aggressive and invasive behavior and could explain, to some degree, the unique clinicopathological features of this slow growing lesion, which in turn, may favor the presence of desmoplasia (14).

Another finding of interest in our case was the positive stain for laminin V and type IV collagen, suggesting an inductive effect of the epithelium over the fibrous stroma, which may result in a duplicated basal lamina; this findings support the observations of Lam et al. (15), who found inductive effects expressed in the form of deposits of an acellular eosinophilic matrix associated to epithelial islands in five of eight cases of DA. Periepithelial myxoid changes and the mechanism of desmoplasia are far from being understood (11).

Including this new case, currently there are only three reports of PDA. However, reports of new cases of PDA are necessary for a better understanding of the origin and biological behavior of this particular lesion.

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