LETTERS TO THE EDITOR

Microcystic adnexal (sclerosing sweat duct) carcinoma of intraoral minor salivary gland origin: an extracutaneous adnexal neoplasm?

To the Editor:

It was with utmost interest that we read the report by Basile and Lin, describing a salivary gland tumor of the lip mimicking a microcystic adnexal carcinoma (MAC) of the skin. Because intraoral minor salivary and cutaneous sweat glands share a similar architecture, it is not surprising that tumors arising in both appendages show a striking morphologic overlap. Pleomorphic adenoma versus chondroid syringoma, membranous basal cell adenoma versus cylindroma, and salivadenoma papilliferum versus syringadenoma papilliferum are authentic entities. Similarly, myoepithelioma and myoepithelial, adenoid cystic, mucopidermoid and mucinous carcinomas are found at both sites. A recent article regarding pilair differentiation in palatine pleomorphic adenomas is also appealing. Despite these facts, cutaneous-type adnexal lesions or their mimics occurring in the extracutaneous intraoral region are one of the most challenging areas of oral pathology. Excluding common sebaceous pathoses, basal cell carcinoma, trichoepithelioma, tubulopapillary hydroadenoma, keratocyst, steatocystoma simplex, and epidermal choristoma are prime examples.

Primary minor salivary gland tumors with microscopic features ascribed to MAC, also referred to as sclerosing sweat duct or syringomatous carcinomas, are rarely reported. Clinically, they were characterized as follows: 1) 8 patients ranged in age from 43 to 77 years (mean 63); 2) 5 were women; 3) 3 involved the lip, 3 the tongue, and the remaining 2 were located in the buccal mucosa; 4) the lesion was a slow-growing ill-defined mass and present from 6 months to 19 years; 5) the tumor size varied from 1 to 3 cm; and 6) all patients were free of recurrence and metastasis from 1.6 to 3 years after excision. Microscopically, the tumor was composed primarily of deceptively benign-looking ducts and cords with invasion of small nerves, often in a targetoid arrangement. Unlike cutaneous MAC, there was no apparent zonal phenomenon with solid nests superficially with small tubules deep. Four cases had foci of squamous differentiation. The stroma was characteristically sclerotic. Tumor cells were negative for vimentin, smooth muscle actin, and/or S-100 protein.

We would like to add our experience of a palatal tumor histologically similar to MAC. The lesion clinically appeared as a 1.6 × 1.0 cm ulcerated nodule of 6 months’ duration in a 52-year-old woman (Fig. 1, A). The patient had developed neither recurrence nor metastasis at the time of writing (4 years after surgery). The partial maxillectomy specimen consisted exclusively of haphazardly scattered small nests and strands of tubulotrabecular carcinoma embedded in a markedly sclerotic stroma (Fig. 1, B and C). Smaller solid clusters and single tumor cells invaded the underlying bone (Fig. 1, D) and exhibited extensive perineural growth (Fig. 1, C and E). Tubular structures most frequently lined by a single layer of cytologically uniform cuboidal cells often contained intraluminal mucin, some of which had tail-like extensions (Fig. 1, B, C, and F). Squamous metaplasia was occasionally seen in the superficial nests (Fig. 1, F). The staining of vimentin, smooth muscle actin and S-100 protein was essentially negative (Fig. 1, E). The Ki-67 labeling index was <3%. This tumor failed to meet established criteria for any existing entity of the salivary glands, resulting in considerable diagnostic difficulty. Reluctantly, a MAC-like tumor was considered at that time. After reading the paper of Basile and Lin, we reevaluated our case by extensive sampling of the entire specimen. Semiserial sections were notable for the presence of small numbers of substantial cribriform areas that are otherwise typical of polymorphous low-grade adenocarcinoma (PLGA) (Fig. 1, G). We reconsider that these architectural and cytologic appearances form part of the spectrum of PLGA, regardless of both the absence of other characteristic polymorphous growth patterns (e.g., solid, papillary, cystic and fascicular) and the contradictory immunoprofile (PLGA is strongly and diffusely positive for vimentin and S-100 protein).

The existence of oral cavity tumors with eccrine differentiation is not accepted in either standard pathology textbooks or in other authentic publications to our knowledge. The previously reported cases of intraoral MAC have significant overlap with PLGA (e.g., minor salivary gland lesion of middle-aged woman, indolent clinical behavior, favorable prognosis, and characteristic microscopic features); therefore, this unique tumor is probably not a sweat gland type of cutaneous carcinoma, but may better be sorted into a tubular subtype of PLGA accompanied by a pronounced stromal desmo-
plasia. Before considering a rare intraoral occurrence of MAC, the possibility of PLGA has to be seriously ruled out in such circumstances. Because the utility of immunohistochemistry as an adjunct is questionable, the meticulous scrutiny of multiple sections at different levels will increase the diagnostic specificity, as in the present case.

**Fig. 1.** A, Ulcerated tumor on the hard palate. B, Proliferating small tubules and cord-like trabeculae within the submucosa (hematoxylin and eosin [HE], original magnification ×100). C, Desmoplastic stroma surrounds infiltrating tubular structures. Arrows indicate perineural growth (HE, original magnification ×100). D, Invasion of adjacent bone (HE, original magnification ×200). E, Neurotropism. Perineural tumor cells are negative for S-100 protein (streptavidin biotin method, original magnification ×400). F, Intraluminal mucin and squamous metaplasia (periodic acid−Shiff, original magnification ×400). G, Additional section shows large cribriform structures with pseudomicrocystic spaces (HE, original magnification ×200).

**References**

Myoepithelial cells are functionally deficient in sialadenosis: Still an assumption

To the Editor:

We read with great interest the article published by Ihrler et al. on “Pathogenesis of sialadenosis: possible role of functionally deficient myoepithelial cells.” In that article, the authors investigated the possible role of functionally deficient myoepithelial cells in the development of sialadenosis. Morphometric analysis of gland was performed after using immunohistochemistry for CK-14, α-actin, and Ki-67 on normal and sialadenosis parotid gland. In sialadenosis, acini were much larger; there was a minor decrease in the density of the distribution of myoepithelial cells stained for CK-14 and a major decrease in the density of the distribution and thickness of the myofilament component of myoepithelial cells stained for α-actin; and the proliferation of acinar and myoepithelial cells was reduced. It was concluded that there is functional myoepithelial insufficiency leading to loss of mechanical support for the acini causing acinar enlargement. Although the authors performed comprehensive and extensive research, we would like to discuss certain aspects of the study.

First, histologically and electron microscopically, it is known that in sialadenosis there is enlargement of acini due to accumulation of secretory granules. We think that this acinar enlargement causes compression of myoepithelial cells between the basal plasma membrane of acinar cells and basement membrane, leading to decreased thickness of myofilament component of myoepithelial cells (pressure effect). The possibility of this phenomenon cannot be ignored in sialadenosis. Similarly, the possibility of pressure atrophy of myoepithelial cells secondary to compression rather than functional atrophy cannot be overlooked.

Second, as in case of fat cells, where sometimes owing to pressure from the accumulated lipids nuclei may not be visible, nuclei of compressed myoepithelial cells may not be visible in severely enlarged acini in sialadenosis. In such situations, expression of Ki-67 may be less, because it is a nuclear marker.

The theoretic possibilities mentioned in the first and second points could be appreciated/explored by correlating the area of acini with Ki-67 and α-actin expressions.

Third, the markers used in this study (CK-14, α-actin, and Ki-67), alone and together, do not express the functional potential of the myoepithelial cells. Therefore, we think that it is inappropriate to some extent to conclude that there is functional insufficiency of myoepithelial cells. But to the best of our knowledge, no methodology is available that can analyze the functional potential of myoepithelial cells. However, the authors have performed the best possible method to study the myoepithelial cells.

Finally, we conclude that functional insufficiency of myoepithelial cells in sialadenosis should be considered as an assumption, needing further studies to come to a definite conclusion.

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REFERENCE


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