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LETTER TO THE EDITOR

Microcystic Adenocarcinoma: An Initially Overlooked First Proposal of the Term

Fumio Ide[1,2] · Naoyuki Matsumoto[2] · Kentaro Kikuchi[1] · Kaoru Kusama[1]

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To the Editors,

Our attention has been drawn to the recent report by Zhang et al. [1], describing a single additional case of sclerosing microcystic adenocarcinoma, bringing to nine the number of examples documented in the English language literature [2–5]. Although the term "microcystic adenocarcinoma" was originally attributed to Mills et al. [4] in 2016 [1, 5], it is pertinent to make a few comments here on the first proposal of this entity in 2008 by Rosebush et al. [6] that escaped the attention of the vast majority of pathologists.

Following a report by Johnston and Toker [7] in 1982, a number of definite or possible cases of salivary microcystic adenocarcinoma with sclerosing features were documented under a variety of names [1–6, 8–10]. Without much supportive rationale, sclerosing microcystic adenocarcinoma is now only the designation that has survived this terminological conundrum [1, 5]. Readers interested in the previously discarded terms are referred to two review articles [5, 9]. It is clear that Rosebush et al. [6] initially introduced, in abstract form, the term "microcystic adenocarcinoma." Two cases of what they considered to be a distinct entity in the field of salivary gland tumor pathology were presented at the American Academy of Oral and Maxillofacial Pathology annual meeting in 2008. These cases, under the designation microcystic adenocarcinoma, appeared as a scar on the lower lip and buccal mucosa in two female patients aged 68 and 69 years, respectively. Microscopically, these tumors showed small infiltrating cords and single-file arrangements of cytologically bland epithelial cells exhibiting conspicuous ductal differentiation. They also displayed a biphasic or bilayered structure of flattened to cuboidal inner luminal ductal cells positive for cytokeratin, epithelial membrane antigen and carcinoembryonic antigen, as well as outer abluminal myoepithelial cells positive for S-100 protein and p63. The Ki-67 labeling index was estimated to be less than 5%, a result that has now been confirmed [3–5, 10]. Obviously, Rosebush et al. [6] were very aware of the significance of stromal sclerosis, and were undoubtedly among the first to appreciate the considerable morphological overlap between mucosal microcystic adenocarcinoma and cutaneous microcystic adnexal carcinoma [2]. Although the presence of perineural and/or intraneural epithelium is usually associated with microcystic adenocarcinoma [1, 3–5, 8–10], there was no mention of neurotropism. Nevertheless, their many original observations provided the conceptual framework for this entity.

At the American Academy of Oral and Maxillofacial Pathology 2012 annual meeting, Romañach et al. [10] inherited the term "microcystic adenocarcinoma" from the previous abstract of Rosebush et al. [6]. Four years later, Mills et al. [4] published a full-length paper on the topic, in which they suggested consideration of a very similar name "sclerosing microcystic adenocarcinoma" without citing the above two abstracts [6, 10]. We can sense from their reference list that their English literature review was perhaps not quite as extensive as it should have been. Although Mills et al. [4] were clearly not the first to propose the term "microcystic adenocarcinoma," their series of five cases brought knowledge about the entity to a new level, resulting in popularization of the term.

It is important to distinguish infiltrating but non-metastasizing sclerosing microcystic adenocarcinoma from highly malignant sclerosing (desmoplastic or microcystic adnexal carcinoma-like) squamous cell carcinoma in the extracutaneous head and neck [11–13]. The latter tumor is entirely confined to the deep tissue and shows a morphea-like infiltration

✉ Fumio Ide
idef@dent.meikai.ac.jp

[1] Division of Pathology, Department of Diagnostic and Therapeutic Sciences, Meikai University School of Dentistry, 1-1 Keyakidai, Sakado, Saitama 350-0283, Japan

[2] Department of Diagnostic Pathology, Tsurumi University School of Dental Medicine, 2-1-3 Tsurumi, Tsurumi-ku, Yokohama 230-8501, Japan

of single cells, narrow cords and thin strands surrounded by a sclerotic stroma. There is neither carcinoma nor dysplasia in the overlying mucosal epithelium [11, 12], nor are there any squamous (keratinizing) phenotypes. Furthermore, it can at times feature minute areas showing ductal differentiation [11], imparting a microcystic adenocarcinoma-like appearance to the lesion. It is essential for pathologists to be aware of this high-risk variant of oral squamous cell carcinoma when considering differential diagnoses [11, 12]. Other rare possibilities would include myoepithelioma [14], mucoepidermoid carcinoma [15] and odontogenic carcinoma [16] with a prominent desmoplastic stromal reaction, occupying most of the tumor volume. These intraoral tumors can be excluded primarily on the basis of careful microscopic assessment.

In summary, this brief review is the result of an overlooked abstract of Rosebush et al. [6] that came to our attention. As stated above, their study was noteworthy in that it established an entity that ultimately became known as sclerosing microcystic adenocarcinoma. In a strict historical sense, we credit Rosebush et al. [6] for not only coining the term "microcystic adenocarcinoma" but their original description of most of its now well-known pathological characteristics. With regard to terminology, the term "sclerosing microcystic adenocarcinoma" proposed by Mills et al. [4] in 2016 is virtually identical to that made by Rosebush et al. [6] eight years earlier.

Compliance with Ethical Standards

Conflict of interest The authors declare no conflict of interest.

Ethical Approval This article is a review of the literature and does not contain any studies with human participants or animals.

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