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Pleomorphic Adenoma Consisting of Multiple Cysts with Squamous Epithelial Lining: Findings on MRI, FNAC, and Histopathological Examination

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Abstract

The incidence of pleomorphic adenoma (PA) of the minor salivary glands is reported to be 10%, and while the histological findings in PA can be diverse, keratin-filled cysts lined by squamous epithelium are rarely reported. The condition can, however, present with cyst formation in some cases. We review a rare case of pleomorphic adenoma in the buccal mucosa that involved the formation of multiple squamous epithelium-lined cysts in a 69-year-old woman. Magnetic resonance imaging (MRI), fine needle aspiration cytology, and histopathological examination were performed. Physical examination revealed a painless, mobile, elastic hard mass in the right buccal mucosa, measuring 2.5 × 1.0 cm. The MRI revealed a well-defined lesion with different signal intensities in the medial and distal regions of the right cheek. The medial side of the lesion showed a low signal intensity on T1-weighted imaging (T1WI) and T2-weighted imaging (T2WI), while the distal side showed a low signal intensity on T1WI, and a high signal intensity on T2WI and short tau inversion recovery (STIR) imaging. Fine needle aspiration of the lesion was performed under local anesthesia and a cytological diagnosis of an epidermoid or dermoid cyst was made. The tumor was completely resected under local anesthesia combined with intravenous sedation. The histopathological examination demonstrated the proliferation of atypical tumor cells with poor atypia and the formation of glandular, alveolar, large, and small cysts. The cysts were lined by keratinized squamous epithelial cells, their cavities were filled with keratinous material, and foreign body reaction was observed

after rupture. Histopathological evaluation led to the diagnosis of pleomorphic adenoma. The patient had no evidence of recurrence, 2 years and 3 months after the surgery. In conclusion, the presence of multiple cysts lined by squamous epithelium can pose a significant diagnostic challenge in patients with PA. It is important to make the correct diagnosis in order to avoid unnecessarily aggressive therapy.

Keywords

Salivary Gland Tumor, Pleomorphic Adenoma, Cystic Formation

1. Introduction

Pleomorphic adenoma (PA) is the most common benign tumor of the major and minor salivary glands. The parotid gland is the most common site of PA. The incidence of PA of the minor salivary glands is reported to be 10% [1]. The minor salivary glands located in the hard palate (42.8% - 68.8%) are the most commonly affected, followed by those in the upper lip (10.1%) and then the ones in the buccal mucosa (5.5%) [2]. Clinically, it presents as a slow growing, painless, sessile and firm mass which may or may not have an ulcerated surface.

Histologically, PA is characterized by a wide spectrum of morphological patterns, including squamous cells, mucous cells, oncocytes, sebaceous cells, bone tissue, adipose tissue, and crystalline material [3]. It is composed of a mixture of glandular epithelium and myoepithelial cells within a mesenchyme-like tissue, and the proportion of each component varies widely among individual tumors. The histomorphological variations are so extensive that in an incisional biopsy specimen, diagnosis can be challenging. Extensive keratin-filled cysts lined by squamous epithelium are a rarely reported phenomenon in PA. We present an unusual case of PA of the buccal mucosa with the formation of multiple squamous epithelium-lined cysts, accompanied by magnetic resonance imaging (MRI), fine needle aspiration cytology, and histopathological investigations. Informed consent was obtained from all individuals included in the study. All procedures performed in studies involving human participants were in accordance with the ethical standards of the Committee on Studies Involving Human Beings of Nihon University School of Dentistry at Matsudo (EC-20-19-023-1) and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

2. Case Report

A 69-year-old woman presented with the complaint of a mass that had grown slowly in size over the previous 8 months in the right buccal mucosa. She had no past history of surgery, trauma, or infection of the buccal mucosa. There was a past medical history of cerebral infarction, hypertension, diabetes, and nephrolithiasis. Physical examination revealed hypoesthesia of the right lower lip and

no facial asymmetry. On intraoral examination, a mass measuring approximately 2.5 cm × 1.0 cm was observed in the right buccal mucosa. The lesion was well-defined, peanut-like in form, movable, painless, and elastic hard. There were no associated palpable lymph nodes (Figure 1). Fine needle aspiration of the lesion was performed under local anesthesia; an epidermoid or dermoid cyst was suspected after cytological findings. A provisional diagnosis of epidermoid cyst was made because the differential diagnoses included a benign salivary gland tumor. The lesion was completely resected under local anesthesia combined with intravenous sedation. There was no evidence of recurrence in the patient over a postoperative follow-up period of 2 years and 3 months.

- MRI findings

She MRI scan revealed a well-defined lesion in the right cheek, with different signal intensities in its medial and distal regions. The medial side of the lesion showed a low signal intensity on T1-weighted imaging (T1WI), T2-weighted imaging (T2WI), and short tau inversion recovery (STIR) imaging, while the distal side of the lesion showed a low signal intensity on T1WI, and a high signal intensity on T2WI and STIR imaging (Figure 2).

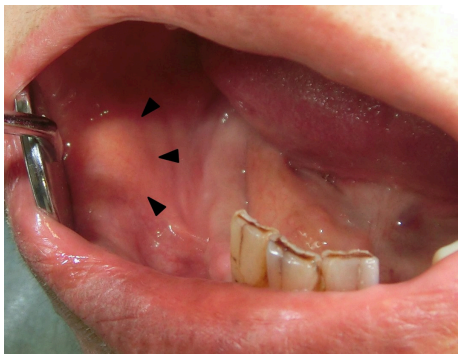

Figure 1. Photograph of the initial intraoral examination showing a well-defined and elastically hard mass (arrow head) in the right buccal mucousa.

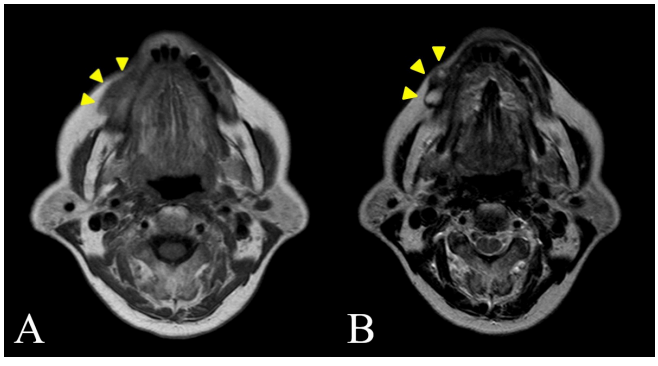

Figure 2. A: Magnetic resonance imaging showed the lesion (arrow head) showed a well-defined low signal intensity area on T1WI. B: There is a low signal intensity on mesial side of the lesion, and the distal side showed a high signal intensity on T2WI (arrow head).

- Cytological Findings

Alcohol-fixed direct smears were stained with the standard Papanicolaou stain. A cluster of keratin debris was seen (**Figure 3A**), and epithelial cells with keratohyalin granules and agglomerates of epithelial cells that were about to be enucleated were observed by higher magnification (**Figure 3B**). No atypical cells were found, and a cytological diagnosis of the epidermoid cyst was made.

- Gross and Histopathological findings

The resected specimen comprised of an encapsulated soft tissue mass measuring 2.3 cm × 1.2 cm × 1.0 cm. The mass was elastic and slightly hard, and its cut surface was firm and brown to yellowish white in color.

Histopathological examination of hematoxylin and eosin-stained sections revealed that the lesion was mostly covered with a fibrous capsule; one side of the nodule was cystic, while the other bore numerous cysts consisting of epithelial tumor cells (**Figure 4A**). In certain parts of the tumor, a foreign body reaction following rupture was observed. The tumor cells were structured in a tubular manner similar to that of glands, and an eosinophilic glass-like substance was present in the glandular cavity. The duct-like formations consisted of ductal luminal cells in the inner layer and myoepithelial-like cells in the outer layer (**Figure 4B**). The ductal structures occurred in variable sizes and shapes. Within some epithelial nests, foci of squamous metaplasia with keratohyalin granules were found. The cavities were filled with keratin debris (**Figure 4C**). The cystic cavities and tumor stroma contained calcified foci in some areas (**Figure 4D**). The tumor cells with a ductal structure and those with a cystic cavity were continuous with one another (**Figure 4E**). No intracapsular invasion or atypical cell was observed. The pathological diagnosis was pleomorphic adenoma with formation of multiple cysts with squamous epithelium lining.

3. Discussion

PA accounts for 54% - 65% of all salivary gland neoplasms and 80% of all benign salivary gland tumors [4]. PA frequently occurs frequently in the minor salivary glands located in the palate, and rarely in those located in the buccal mucosa. Various studies have reported different incidences of PA of the minor salivary glands of the cheek (**Table 1**) [5] [6] [7] [8]. In this case, the tumor was located on the side of the buccinator muscle facing the oral cavity in the right buccal mucosa, suggesting that the tumor was derived from the buccal glands.

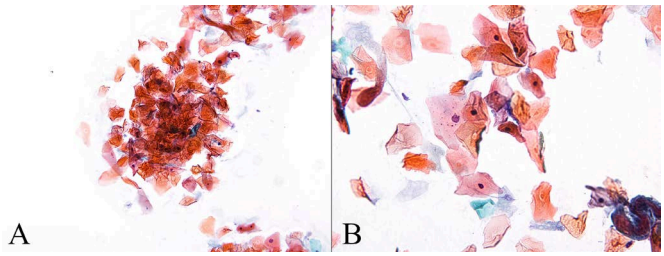

Figure 3. A: A cluster of squamous cells and horny substances. FNA (Pap. staining, ×20) B: Epithelial cells about to enucleate. FNA (Pap. staining, ×40).

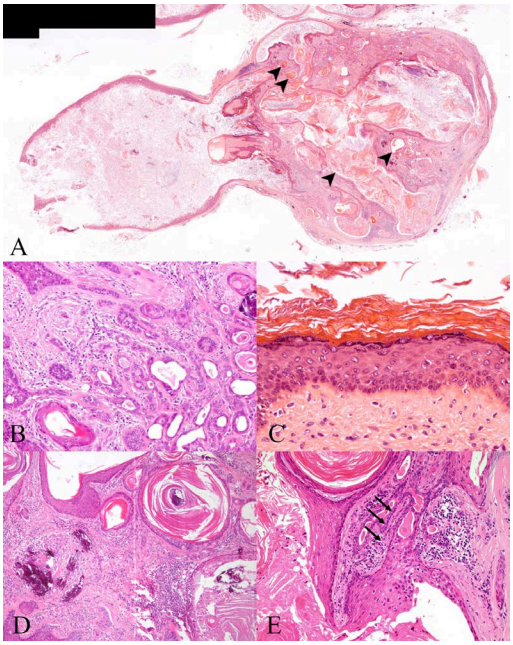

Figure 4. A: Large and small cyst formation. (arrow head) (H. E. staining, Gross) B: Higher power view of the tumor epithelial cells and duct-like structures, and myoepithelial-like cells in the outer layer. (H. E. staining, ×10) C: The cystic wall lined with a hyperkeratinized squamous epithelium with keratohyalin granules. (H. E. staining, ×40) D: Large and small cysts covered by hyperkeratinized squamous epithelium were keratin-filled. (H. E. staining, ×10) E: Tumor cells with ductal structure and cystic cavity are continuous. (arrow) (H. E. staining, ×10)

Table 1. Literature review of the frequency of pleomorphic adenoma of the minor salivary glands of the cheek.

Author	Year	Total number of pleomorphic adenoma cases	Number of cases in cheek	Percentage (%) of cases in cheek
Chaudhry, A.P., *et al.* [5]	1961	476	38	8
Isacsson, G. and Shear, M. [6]	1983	140	7	5
Cohen, M.A. [7]	1986	144	10	7
Buchner, A., *et al.* [8]	2007	149	19	13

Past reports of PA accompanied by the formation of multiple squamous epithelium-lined cysts are listed in **Table 2** [9] [10]-[24]. Several mechanisms of cyst formation in PA have also been suggested. Devgan, *et al.* [25] described that an etiology involving squamous metaplasia of the tumor cells and central necrosis followed by well-defined differentiation of the epithelium, was responsible for cyst formation. Hamdan, *et al.* [26] reported that ischemia could lead to the metaplastic changes in PA. Goulart, *et al.* [15] suggested that the exposure of minor salivary gland ductal epithelium to different irritants could play a significant role in the formation of keratin pearls. Lucas [27] described that cyst formation was

Table 2. Reported cases of pleomorphic adenoma with formation of multiple squamous epithelium-line cysts.

Author	Year	Location
Abiko, Y., *et al.* [9]	1993	palatal gland
Lam, K.Y., *et al.* [3]	1998	plate
Stewart, C.J.R., *et al.* [10]	2000	minor salivary gland
Aker, H., *et al.* [11]	2003	buccal mucosa
Brachtel, E.F., *et al.* [12]	2003	parotid gland
Batrani, M., *et al.* [13]	2008	buccal mucosa
Siddaraju, N., *et al.* [14]	2009	submandibular gland
Goulart, M.C., *et al.* [15]	2009	upper vestibule
Kaveri, H., *et al.* [16]	2014	palate
Reddy, V., *et al.* [17]	2015	retromolar trigone
Brisebois, S., *et al.* [18]	2015	palate
Jaishankar, H.P., *et al.* [19]	2016	palate
Sreelatha, S.V., *et al.* [20]	2017	buccal mucosa
Tandon, A., *et al.* [21]	2018	palate
Sharma, S., *et al.* [22]	2018	palate
Anjum, R., *et al.* [23]	2019	lip(Low)
Urs, A.B., *et al.* [23]	2019	buccal mucosa

associated with degeneration of the layer of tumor tissue containing hemorrhage. It has been reported that necrotic lesions may give rise to cysts in polymorphic adenomatous carcinoma [9]. Similar to Abiko, *et al.* [9], we assumed that the cysts in this case were formed due to the degeneration of squamous metaplasia of the tumor cells, owing to the following reasons: the lining epithelium was continuous with the tumor cells, the cyst wall consisted of hyperkeratinized squamous epithelium, and a large amount of keratinized material was stored within the cysts.

Squamous metaplasia and keratin pears are not unusual [28]. However, keratin-filled cysts lined by squamous epithelium are a rare finding. PAs with extensive squamous metaplasia can signify a potential pitfall in the histopathological diagnosis.

Fine needle aspiration cytology (FNAC) is a widely accepted investigation for differentiating between benign and malignant tumors and for the histological evaluation of salivary gland tumors. Its diagnostic accuracy has been reported as 80% - 95% in most series, provided that adequate and appropriately prepared materials are available [10] [29]. Despite its histologic diversity, PA can, in most cases, be easily diagnosed through cytological evaluation, due to its typical cytological appearance. The combination of bland epithelial cells and fragments of chondromyxoid stroma with spindle cells is very characteristic of this tumor

[30]. However, in some cases, major variations from this common cytological pattern may result in an erroneous cytological diagnosis, and subsequently, in a different treatment or surgical approach to the tumor [30]. Foci of squamous cells are integral features of PA; nonetheless, extensive squamous metaplasia is uncommon and can be easily misinterpreted as squamous cell carcinoma, especially on FNAC, due to its limited and selective sampling [22].

In addition, the diagnosis of PA becomes challenging in the absence of chondromyxoid stroma, making it imperative to understand this diagnostic pitfall [31]. In this case, only epithelial cells with keratohyalin granules as well as clumps of epithelial cells that were about to be enucleated were observed; therefore, a presumptive diagnosis of PA was not made. Such an observation could be explained by the relatively large cyst structure, which could have prevented the puncture needle from reaching the tumor parenchyma. In the presence of a salivary gland tumor with cystic change, the differential diagnoses of Warthin tumor, basal cell adenoma, cystadenoma, mucoepidermoid carcinoma, acinic cell carcinoma, and cystadenocarcinoma were considered. Histopathologically, PA of the minor salivary glands is usually unencapsulated [32], and approximately 50% of tumors of the minor salivary glands are considered to be malignant [1] [33]. Histopathologically, differential diagnoses include simultaneous occurrence of PA and dermoid cysts.

Dermoid cysts usually have a clear stratum granulosum all around. In this case, there is a clear granular layer, but it is limited. The cyst wall consisted of hyperkeratinized squamous epithelium, the lining epithelium was continuous with the tumor cells. It is rare for dermoid cysts to form a large number of large and small cyst structures locally. Based on the findings above, the simultaneous occurrence of PA and dermoid cysts was considered to be unlikely.

It is necessary to consider a frozen section diagnosis, owing to the high rate of false negatives for malignancy in cystic tumors. Surgical resection of the tumor is generally used in the treatment of patients with PA. PA of the minor salivary glands is widely excised including the periosteum or bone because the tumor cells infiltrate the periosteum to either form a nodule or cause detachment of the periosteum from the underlying bone. Enucleation is not the treatment of choice, as it can lead to high local recurrence rate. In this case, the suspicion of a dermoid cyst on FNAC led to minimal excision of the surrounding healthy tissue. No recurrence was observed 2 years and 3 months after the operation but owing to the high recurrence rate of PA, long-term follow-up is necessary.

4. Conclusion

In conclusion, PA consisting of multiple cysts lined by squamous epithelium can pose a significant diagnostic challenge. It is important to bear this presentation in mind in order to distinguish PA from malignant lesions and to avoid unnecessarily aggressive therapy. A frozen section analysis should be considered because of the high rate of false negatives for malignancy in cystic tumors.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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