Myoepithelioma arising from the buccal gland: Histopathological and immunohistochemical studies

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Abstract: A rare case of myoepithelioma of the buccal gland in a 54-year-old Japanese woman is reported. As the swelling exhibited a normal mucosal color and was relatively well defined, showing no ulcers, a benign salivary gland tumor was suspected upon clinical inspection. Microscopically, the parenchyma of the present case mainly consisted of plasmacytoid cells with round nuclei and eosinophilic cytoplasm, and partial spindle cells with eccentric nuclei. The stroma was composed of fibro-hyalinized or myxoid connective tissue that separated from the parenchyma. Immunohistochemically, the cytoplasm of the plasmacytoid and spindle cells was moderately positive for vimentin and GFAP, whereas the buccal gland adjacent to the tumor was negative for these antibodies. S-100 protein reactivity is strong for both types tumor cells. Actin reactivity was negative for both types of tumor cells, notwithstanding the fact that myoepithelial cells of the buccal gland were positively stained. Anti-cytokeratin reactivity was weak for both types of tumor cells in portions of the plexiform and solid areas; nevertheless, the buccal glands were moderately positive. These results suggest that neoplastic myoepithelial cells exhibit abnormal differentiation and modification. There have been only two published reports of myoepithelioma arising from the buccal gland in the literature to date. (J. Oral Sci. 42, 39-42, 2000)

Key words: myoepithelioma; buccal gland;
immunohistochemical study.

Introduction

Myoepithelioma, which is the presence of a benign salivary gland, mainly affects the parotid gland of the major salivary gland (1). Table 1 presents a compilation of reported myoepitheliomas from the Japanese literature. The occurrence of tumors in the minor salivary glands is relatively rare, and the minor salivary gland most frequently affected is the palatal gland (2). Thus, a tumor of the buccal gland is uncommon, and until now only a few histopathological and immunohistochemical studies involving the minor salivary glands have been performed extensively and conclusively. We observed a rare case of myoepithelioma arising from the buccal gland and studied it histopathologically and immunohistochemically.

Case Report

A 54-year-old Japanese woman came to the Nihon University Dental Hospital at Matsudo with the chief complaint of a painless swelling in the left buccal mucosa. The patient had noticed the swelling for five years. As the swelling exhibited the normal color of the mucosa and was relatively well defined without ulcer (Fig. 1), a benign salivary gland tumor was clinically suspected. The tumor was resected under local anesthesia. Macroscopically, the resected, oval, and relatively well encapsulated tumor specimen measured 1.0 cm × 0.7 cm × 0.6 cm. The cut surface was grayish-white in color and had a solid or granular appearance. There has been no evidence of recurrence eight months after the operation.

Results

Microscopically, the tumor of the present case was encapsulated by fibrovascular connective tissue but was located adjacent to the normal buccal gland. The parenchyma was homogenous, with a round to ovoid

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outline. The growth pattern exhibited solid, plexiform, and scattered types. The parenchyma mainly consisted of plasmacytoid cells with round nuclei and eosinophilic cytoplasm, and partial spindle cells with eccentric nuclei (Fig. 2). The tumor cells appeared to show cellular and nuclear pleomorphism, but mitotic figures were absent. The stroma was composed of fibro-hyalinized or myxoid connective tissue that separated from the parenchyma. The myxoid tissue was stained with alcian blue, whereas the fibro-hyalinized tissue was positive for periodic acid schiff

reactions. The spindle cells were mainly scattered in the myxoid tissue.

An immunohistochemical study was performed on formalin-fixed, paraffin-embedded sections using the labeled streptavidin-biotin methods. The primary antibodies used in the present case were anti-cytokeratin (anti-CK), anti-actin, anti-glial fibrillar acidic protein (GFAP), anti-S-100 protein, and anti-vimentin. The slides to be stained for CK and actin were digested with pronase E solution (Sigma Chemical's Trypsin 0.4 g/400 ml Tris

Fig. 1 The intraoral view at the first medical examination reveals a tumor in left buccal mucosa.

Fig. 2 Photomicrograph showing parenchyma with solid and plexiform growing pattern (H&E, $\times 200$).

Fig. 3 The tumor was composed of mainly plasmacytoid and partly of spindle cells which are strongly positive for vimentin ($\times 400$).

Fig. 4 Though the myoepithelial cells of the buccal gland were highly positively stained for actin, the tumor cells were negative ($\times 400$).

Table 1 Myoepithelioma of the minor salivary gland from Japanese literatures

Authors	Reported year	Age(years)	Sex	Location
Tajima <i>et al.</i> ⁽⁵⁾	1984	56	F	Cheek
Hamada <i>et al.</i>	1986	60	M	Palate
Wakano <i>et al.</i>	1987	67	M	Palate
Ogawa <i>et al.</i> ⁽⁶⁾	1988	85	M	Palate
		79	M	Palate
		68	M	Palate
		53	M	Palate
		63	F	Palate
		38	F	Palate
		26	F	Palate
Kawabe <i>et al.</i> ⁽⁷⁾	1991	53	M	Palate
Hara <i>et al.</i>	1993	53	M	Palate
Kiyomihara <i>et al.</i>	1993	31	M	Palate
		31	F	Palate
Hiruta <i>et al.</i>	1994	35	F	Palate
Sakata <i>et al.</i>	1997	47	M	Palate
Nakajima <i>et al.</i>	1997	20	M	Palate
Kamata <i>et al.</i>	1997	60	M	Lingual
Watanabe <i>et al.</i>	1998	69	F	Palate
Jinde <i>et al.</i> ⁽⁸⁾	1998	15	M	Palate
Sugiura <i>et al.</i>	2000	54	F	Cheek

M: male, F: female

buffer) at 37°C for 10 minutes.

The cytoplasm of the plasmacytoid and spindle cells was moderately positive for S-100 protein, vimentin (Fig. 3), and GFAP, whereas the buccal gland adjacent to the tumor was negative for these antibodies. Nuclear and cytoplasm S-100 protein reactivity is usually strong for both types of tumor cells. Actin reactivity was negative for both types of tumor cells, notwithstanding the fact that myoepithelial cells of the buccal gland were highly positively stained (Fig.

Table 2 Results of immunohistochemistry

Primary antibody	Dilutions	Manufacturers	Plasmacytoid cell	Spindle cell	Salivary tissue
Cytokeratin	1 : 25	Dako A/S	—~±	—~±	++
Actin	1 : 25	Dako A/S	—	—~±	++
GFAP	1 : 50	Dako A/S	++~—	++~±	—
S-100	1 : 200	Dako A/S	++~+	++~±	—
Vimentin	1 : 100	Dako A/S	++	++~+	—

GFAP, glial fibrillar acidic protein; S-100, S-100 protein. ++, strongly positive;

+, moderately positive; ±, weakly positive.

4). The CK reactivity was weak for both types of tumor cells in parts of plexiform and solid areas; nevertheless, duct epithelial cells of the buccal gland were moderately positive (Table 2).

Discussion

Myoepithelioma is a relatively rare benign salivary gland tumor (2). Sheldon first reported myoepithelioma and classified it as a monomorphic adenoma composed of homogenous tissue (3). After repeated revision of salivary gland tumor classifications, myoepithelioma was gradually established and characterized under the criteria of the proliferation of solely neoplastic myoepithelial cells (1). In the archives of the AFIP (2), the frequency of myoepithelioma is less than 1% of all the salivary gland tumors. According to Barns *et al.* (4), among 42 myoepithelioma cases, 21 were located in the parotid gland, 11 in the palate, and 5 in the submandibular gland. In addition, two cases were located on the lip, and one case arose in the cheek, gingiva, and the retromolar area. Thus, the most frequent location was the palate (2) among the minor salivary glands, and tumors arising from the buccal gland were extremely rare. Up to now, there have been only two published reports of myoepithelioma arising from the buccal gland, making the present case the third such reported case (Table 1)(5-8). The reason that the tumor is infrequently located in the buccal gland is unclear, but it may be due a difference in the promotion factors based on the anatomical location of the pathogenesis (9). For example, the palate mucosa is considered masticative though the buccal mucosa is related to licking.

The average age of the Japanese patients diagnosed with myoepithelioma in the minor salivary glands was 50.5 years old, which was higher than the average age of 44 years reported by AFIP (2) for the same diagnosis. The present case was also an older patient (54 years old). Sex distribution was equal regarding this tumor (2). The Japanese cases were similar to this sex distribution, with 14 men and 10 women affected (Table 1).

Myoepithelioma is characterized by the proliferation of homogeneous neoplastic myoepithelial cells (1,2,6,7,10) and is considered to be benign. The histology of the present case was almost the same as that reported elsewhere; where the parenchyma was composed of plasmacytoid and spindle cells with plexiform and scattered patterns. The stroma consisted of fibro-hyalinized or myxoid tissue that was stained with PAS or alcian blue. Batsakis *et al.* (11) and Sciubba *et al.* (12) histologically subclassified myoepithelioma into plasmacytoid, spindle, and mixed types. This classification is now commonly used for the tumor. Kawabe *et al.* (7) reported that myoepithelioma

arising from the parotid gland was mainly composed of the spindle cell type. As for the tumors arising from the minor salivary glands, the plasmacytoid and spindle cell types were equally affected. There is also a malignant counterpart to the tumor named "malignant myoepithelioma" or "myoepithelial carcinoma" (2). The malignant counterpart lesions show invasive development, distant metastasis, and atypia (1,8). The present case, showing a partial approach to the adjacent buccal gland, was not an observed malignancy, but rather a slight pleomorphism of the nucleus was observed. Moreover, it was encapsulated and there was no evidence of malignant features or mitotic figures. As mentioned above, the findings of the present case indicated a benign myoepithelioma containing a mix of plasmacytoid and spindle cells.

Immunohistochemically, the myoepithelial cell markers (8), vimentin, S-100 protein, GFAP, actin and CK antibodies have been used to investigate the tissue origins of the tumor cells. In the present case, the cytoplasm of the tumor cells was moderately positive for vimentin and GFAP, whereas the salivary tissues adjacent to the tumor were negative. Nuclear and cytoplasm S-100 protein reactivity is usually strong for both types of tumor cell. However, actin reactivity was negative for both types of tumor cells, notwithstanding the fact that the myoepithelial cells of the buccal gland were highly positively stained. CK reactivity was weakly positive for both types of tumor cells in part of the plexiform and solid areas; nevertheless duct epithelial cells of the buccal gland were moderately positive reactive for CK. These findings suggest that the tumor cells have a myoepithelial origin because of having mesenchymal and epithelial features. The reactivity for actin in the present case was different from that in some of the previous reports. Nikai et al. (13) described some controversy over staining for CK and actin, indicating that these may be inadequate markers for the tumor cells. The characteristics of neoplastic myoepithelial cells were not the same as those of normal myoepithelial cells in the present case. The cause of the discord is that there is a discrepancy between neoplastic myoepithelial cells and normal myoepithelial cells in the salivary gland. It has been suggested that neoplastic myoepithelial cells exhibit abnormal differentiation and modification.

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