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Cytology of Plasmacytoid Type Myoepithelioma
– Report of Two Cases –

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Myoepithelioma is a rare benign tumor of salivary gland myoepithelial cells, most commonly as a spindle subtype. Here, we present two cases of fine needle aspiration cytology of plasmacytoid myoepithelioma arising from a parotid gland and a hard palate. Aspirates showed plasmacytoid cells with pink-staining, homogeneous, abundant eosinophilic cytoplasm eccentrically displacing the nucleus in cohesive and dissociated forms. Rarely, nuclear grooves and intranuclear cytoplasmic inclusions were evident. These unfamiliar cytologic findings of uncommon myoepithelioma often cause diagnostic difficulties in preoperative aspiration cytology. Recognition of those rare findings provides a reliable diagnostic clue.

Key Words : Myoepithelioma; Biopsy, fine needle; Cytology; Salivary gland neoplasms

Myoepithelioma is a rare (1.5%) benign neoplasm of the salivary glands, which generally occurs in the parotid gland and less often in the minor accessory salivary gland of the oral cavity.[1-3] Histologically, myoepithelioma appears as a spindle, plasmacytoid or mixed subtypes. Of these, the spindle cell type is most common subtype, and the plasmacytoid variant is rare, which complicates preoperative aspiration cytology-based diagnosis.[4,5] Little information concerning myoepithelioma cytology exists, especially plasmacytoid variants.[4,6,7] Here, we describe cytological findings of plasmacytoid myoepithelioma from two patients that displayed disparate cytologies.

CASE REPORTS

Case 1

A previously healthy 33-year-old woman presented for evaluation of a firm palpable mass on the left infra-auricular area, which had developed 8 years previously. The size of the mass had increased. Neck CT revealed a 2.0 cm, oval-shaped mass of low density at the superficial lobe of the left parotid gland. Fine needle aspiration and subsequent superficial parotidectomy were performed. The mass was easily excised from the underlying subcutaneous fat, with no connection with adjacent structures. The 11 year postoperative follow-up was unremarkable.

Case 2

A healthy 52-year-old woman presented for evaluation of a movable mass on the left hard palate, which had been detected at another hospital 1 year before. At that time, the patient had refused mass excision. The patient had radiation history for cervical squamous cell carcinoma (stage IIb) 12 years before. Neck CT showed a well-demarcated 1.5 × 1.5 cm oval mass at the posterior side of the left hard palate with central low density. Fine needle aspiration and excision were performed. The mass was easily excised from the adjacent structures. The 10 year postoperative follow-up was unremarkable.

Cytological findings

Fine needle aspirations were performed using a 22-gauge needle. Smear slides were prepared from the aspirated material and

stained with Papanicolau and hematoxylin-eosin stains. Excised specimens were fixed in 10% formalin and stained with hematoxylin and eosin. A paucicellular smear was taken from case 1 and a highly cellular smear was taken from case 2. The cytologic smears from both lesions yielded cohesive mononuclear cells with occasional scattered cells (Fig. 1A). The round-to-oval shaped mononuclear cells showed plasmacytoid nuclear appearances (Fig. 1B); the cells displayed plump eosinophilic cytoplasm, vesicular nuclei containing dispersed chromatin and inconspicuous nucleoli with occasional nuclear grooves (Fig. 1C). Intranu-

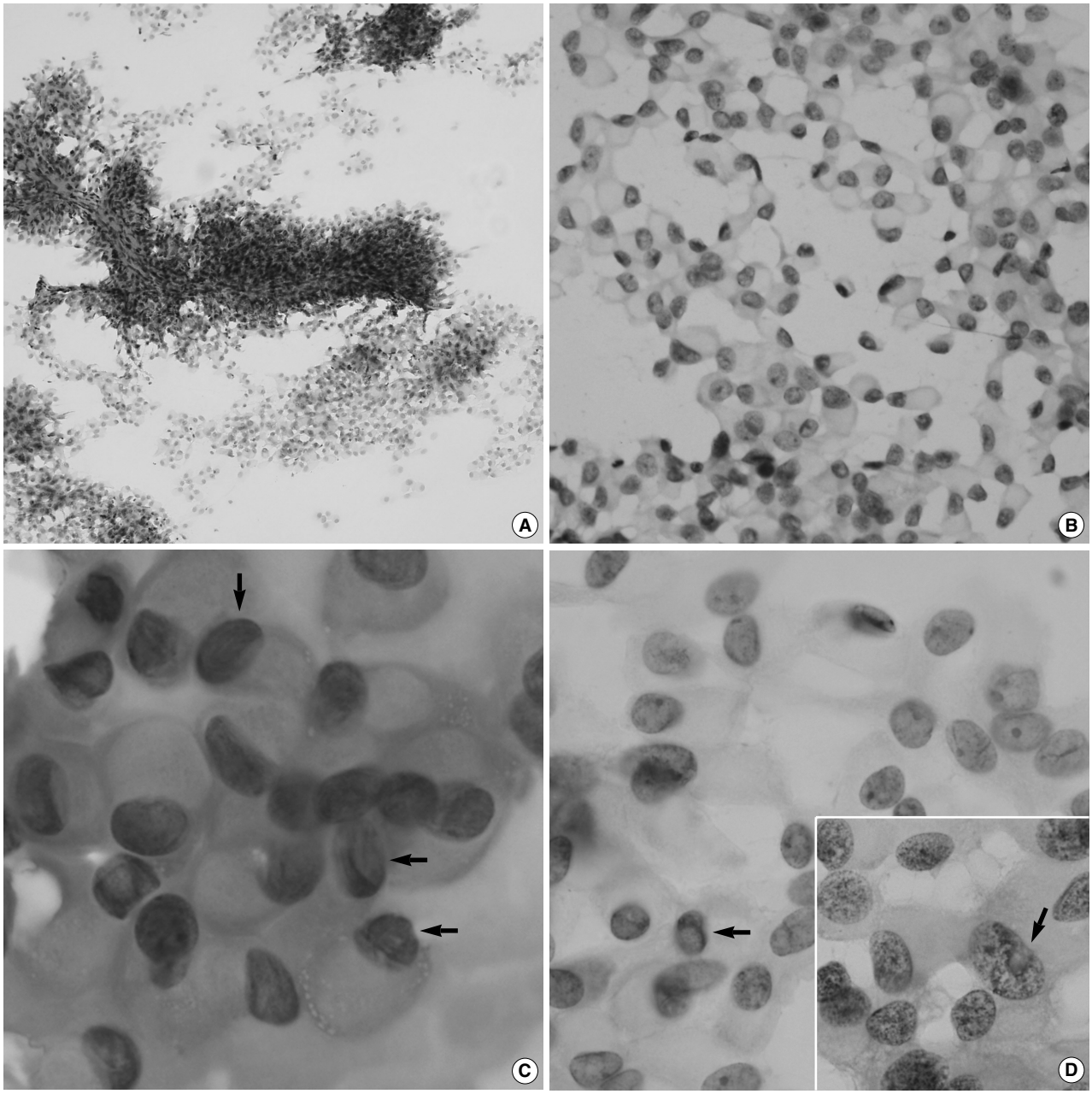

Fig. 1. Microscopic features of cells from both cases. (A) Case 2. Low magnification view reveals cellular smears of cohesive round-to-oval cells with occasional sheet formation and singly scattered forms (Papanicolaou stain). (B) Case 2. Round-to-oval cells with eccentrically located round nuclei, granular chromatin, and nucleoli and abundant dense cytoplasm (Papanicolaou stain). (C) Case 2. Ovoid shaped tumor cells show ovoid-shaped nuclei and abundant cytoplasm and well-defined borders, mimicking plasma cells (Papanicolaou stain). Note the nuclear grooves (arrows). (D) Case 1. Some tumor cells display nuclear clearing appearing nuclear inclusion-like lesion (Papanicolaou stain). Inset indicates intranuclear inclusions (arrow, Papanicolaou stain).

clear inclusions were rarely detected (Fig. 1D). Myxoid background was not evident. Mitosis and cellular pleomorphism were absent and no components of ductal epithelial cells were found. Based on these cytological findings, both cases were diagnosed as myoepithelial cell-rich salivary gland tumors highly suggestive of plasmacytoid type myoepithelioma.

Histological findings

The case 1 superficial parotidectomy specimen measured 4.5 × 3.0 × 2.0 cm, and a bulging firm mass was located in the subcapsular area. The cut surface of the gland showed an ill-defined, pale yellow, 2.0 × 1.5 × 1.0 cm homogeneous lobulated mass. Microscopically, the tumor was composed of round-to-polygonal shaped cells intermixed with a stroma of dense fibrous connective tissue (Fig. 2A). The tumor exhibited a solid architecture with a focal reticular pattern. The tumor cells had eccentric nuclei and an abundant, dense, hyaline, eosinophilic cytoplasm (Fig. 2B). Nuclear atypia or mitoses were absent. In case 2, the resected oval mass measured 1.5 × 1.5 × 0.7 cm. Histologically, it comprised sheets of round cells having a round-to-ovoid silhouette with dense chromatin nuclei located eccentrically, and abundant eosinophilic cytoplasm intermixed in a stroma of dense fibrous tissue. No myxochondroid stroma, mitosis or atypical cytology was observed. Immunohistochemically, tumor cells of both cases showed focal positive reaction for S-100 protein using a 1:1,200 dilution of polyclonal antibody (Dako, Glostrup, Denmark), 1:50 dilution of desmin antibody (ZC18; Dako), 1:100 dilution of smooth muscle actin antibody (HHF; Dako), prediluted vimentin antibody (V9; Dako) and prediluted pancytokeratin antibody (AE1/AE3; Dako). Tumor cells were immunonegative for epithelial membrane antigen (EMA; Dako, 1:200 dilution) or glial fibrillary acidic protein (GFAP; Dako, 1:350 dilution). Cases 1 and 2 were diagnosed as plasmacytoid type myoepithelioma.

DISCUSSION

Myoepithelioma was initially described as a variant of pleomorphic adenoma, and it is now recognized as a distinct tumor of different biological behavior.[1] Like a wide histologic range of myoepithelioma, diverse cytologic features of myoepithelioma render a diagnosis that is finally confirmed by histologic and immunohistochemical examination, generally being positive for cytokeratin, smooth muscle actin and S-100 protein.[8]

There have been only a few reports regarding the fine needle aspiration diagnosis of myoepithelioma.[4-7] Cytological differential diagnoses of myoepithelioma include other salivary gland tumors having predominant proportion of myoepithelial cells such as malignant myoepithelioma (myoepithelial carcinoma), pleomorphic adenoma, and adenoid cystic carcinoma.[2,9,10] Myoep-

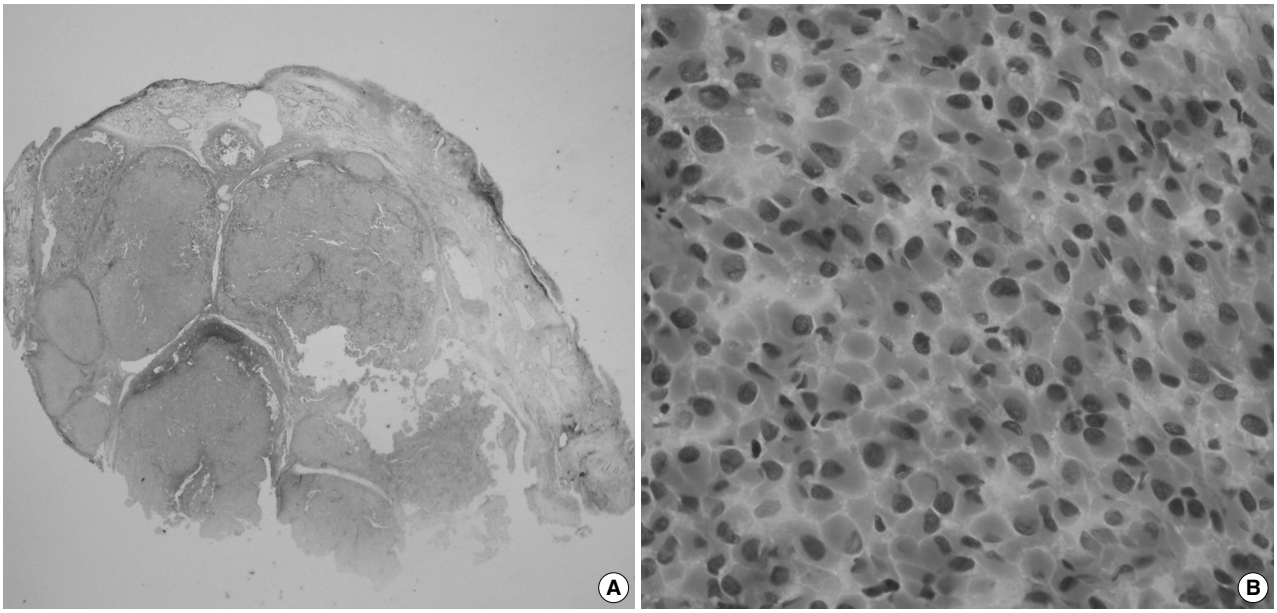

Fig. 2. Microscopic observation from case1 (A) Case 1. A well-defined ovoid mass is composed of sheets of ovoid shaped tumor cells with hyalinization. (B) High magnification reveals peripherally condensed and round nuclei, and eosinophilic cytoplasm.

ithelial cells share epithelial and smooth muscle features, and often display different shape and size such as spindle, stellate, plasmacytoid, epithelioid and clear morphology. They are round-to-ovoid cells with a plasmacytoid appearance, or spindle shaped cells with occasional clear cytoplasm. In some cases, nuclear pleomorphism may be seen.[2,11] Myoepithelial neoplasm has three subtypes; spindle, plasmacytoid and mixed types.[12] Cytologic findings of intranuclear inclusion, prominent nucleoli, and binucleation in myoepithelial neoplasms are shared features of malignant melanoma.[2] Absence of melanin pigment is helpful for diagnosis of myoepithelioma. Intranuclear inclusion has also been reported in myoepithelial carcinoma, pleomorphic adenoma or neurofibroma.[13-15] Immunohistochemical staining for melanoma, epithelial and neural markers are distinguished by using aspiration slides. Myoepithelial carcinoma frequently shows increased mitotic indices in the aspiration cytology.[13] However, the fact that the malignant counterpart of myoepithelioma may show mild atypia or even a monomorphic population of small, fairly monotonous-appearing cells without obvious cytological features hinders the distinction between benign and malignant myoepithelioma in some cases. In pleomorphic adenoma, three cellular components are present in varying proportions: small and cuboidal shaped ductal epithelial cells, chondromyxoid stroma and myoepithelial cells arranged in flat sheets or trabeculae showing occasional squamous, oncocytic, or sebaceous metaplasia.[16] However, the cellular type of pleomorphic adenoma, i.e. pleomorphic adenoma with prominent myoepithelial overgrowth is a cytological pitfall, and its discrimination may be impossible unless histological confirmation is undertaken. Less than 5% of ductal/epithelial components are required for the diagnosis of myoepithelial neoplasm.[1,2] Red-to-purple, myxoid matrix in myoepithelioma is present as a scanty fibrillar substance and spherical globules (balls) surrounded by tumor cells may be vaguely reminiscent of adenoid cystic carcinoma.[10] Occasional presence of extensive hyalinized stroma and cribriform pattern containing mucin of myoepithelioma upon fine needle aspiration may be confused with adenoid cystic carcinoma.[6] Cytologic features such as necrosis, decreased cohesion, enlarged nuclei and nucleoli, coarse chromatin and hyperchromatic nuclei favor adenoid cystic carcinoma. In addition to the aforementioned differential diagnosis, the spindle type myoepithelioma may occasionally simulate low-grade spindle cell soft tissue sarcoma or schwannoma because of long tapering cytoplasmic- to-eosinophilic hair- like cytoplasmic processes, while myoepithelioma of the plasmacytoid variant does simulate plamacytoma.[4,6,17] Plasmacytoid type myoepithelioma shows odd-shaped cellular aggregates and singly scattered cells with round nuclei and finely granular cytoplasm resembling plasma cells, together with strands of metachromatic stroma. An reticular pattern composed of a mosaic arrangement of hyaline-like plump cytoplasm, and scant matrix and considerable fiber formation are characteristic features of the plasmacytoid type.[6] Monotonous medium-sized tumor cells show plasmacytoid appearances with no atypia or mitosis. Absence of cartwheel nuclear chromatin, Russell bodies, and paranuclear Golgi apparatus can help distinguish plasmacytoid type myoepithelioma from extramedullary plasma cell tumor.[4]

In the present two cases, cytologic findings were composed of isolated or clusters of plasmacytoid cells with a trace of myxoid stroma and rare acinar structures. Nuclear striations (zebra lines), first described by Kumar *et al*.,[7] were not presently common. Nuclear grooves are regarded as frequent in spindle type myoepithelioma and mixed type, but were rarely evident in the plasmacytoid type in the present cases. Although those nuclear inclusions and nuclear grooves are shared characteristics of malignant melanoma and pleomorphic adenoma, those features may also be a characteristic of myoepithelioma-like papillary carcinoma of the thyroid.[2]

The present diagnoses of plasmacytoid myoepithelioma were offered by fine needle aspiration, and were confirmed by histologic and immunohistochemical observations. To avoid the aforementioned diagnostic pitfalls, careful searching these cytologic features of plasmacytoid type myoepithelioma is valuable.

REFERENCES

1. Eveson JW, Auclair PL, Gneep DR, El-Naggar AK. Tumours of the salivary glands. In: Barnes L, Eveson JW, Reichart P, Sidransky D, editors. World Health Organization classification of tumours: Pathology and genetics of head and neck tumours. Lyon: IARC; 2005. p. 209-81.
2. Daneshbod Y, Daneshbod K, Khademi B. Diagnostic difficulties in the interpretation of fine needle aspirate samples in salivary lesions: diagnostic pitfalls revisited. Acta Cytol 2009; 53: 53-70.
3. Cuadra Zelaya F, Quezada Rivera D, Tapia Vazquez JL, Paez Valencia C, Gaitán Cepeda LA. Plasmacytoid myoepithelioma of the palate. Report of one case and review of the literature. Med Oral Patol Oral Cir Bucal 2007; 12: E552-5.
4. Lopez JI, Ugalde A, Arostegui J, Bilbao FJ. Plasmacytoid myoepithelioma of the soft palate. Report of a case with cytologic, immunohistochemical and electron microscopic studies. Acta Cytol 2000;

44: 647-52.
5. Hara H, Oyama T, Kimura M, Ishii E, Inoue T, Kasai H. Case of myoepithelioma originating from the palate: collagenogenesis in myoepithelial tumor cells. Diagn Cytopathol 1996; 15: 415-20.
6. Siddaraju N, Badhe BA, Goneppanavar M, Mishra MM. Preoperative fine needle aspiration cytological diagnosis of spindle cell myoepithelioma of the parotid gland: a case report. Acta Cytol 2008; 52: 495-9.
7. Kumar PV, Sobhani SA, Monabati A, Hashemi SB, Eghtadari F, Hamidi SA. Myoepithelioma of the salivary glands. Fine needle aspiration biopsy findings. Acta Cytol 2004; 48: 302-8.
8. Takai Y, Dardick I, Mackay A, Burford-Mason A, Mori M. Diagnostic criteria for neoplastic myoepithelial cells in pleomorphic adenomas and myoepitheliomas. Immunocytochemical detection of muscle- specific actin, cytokeratin 14, vimentin, and glial fibrillary acidic protein. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1995; 79: 330-41.
9. Oka K, Chikamatsu K, Eura M, Katsura F, Yumoto E, Tokunaga H. Clinical significance of fine-needle aspiration biopsy in major salivary gland tumors. Nippon Jibiinkoka Gakkai Kaiho 2002; 105: 1109-15.
10. Kim J, Kim KM, Kim YS, Lee A, Shim SI, Kim BK. Fine needle aspiration cytology of solid type adenoid cystic carcinoma of buccal mucosa; a case report. Korean J Cytopathol 2000; 11: 89-92.
11. Darvishian F, Lin O. Myoepithelial cell-rich neoplasms: cytologic features of benign and malignant lesions. Cancer 2004; 102: 355-61.
12. Katsuyama E, Kaneoka A, Higuchi K, Takasu K. Myoepithelioma of the soft palate. Acta Cytol 1997; 41: 1856-8.
13. Kuwabara H, Kohno K, Kishida F, *et al.* Imprint cytology of malignant plasmacytoid myoepithelioma of the palate. Acta Cytol 1998; 42: 440-3.
14. Chhieng DC, Paulino AF. Cytology of myoepithelial carcinoma of the salivary gland. Cancer 2002; 96: 32-6.
15. Lee JH, Park JK, Hur B. Fine Needle Aspiration cytology of malignant myoepithelioma of the salivary gland: a case report. Korean J Cytopathol 2002; 13: 28-32.
16. Pusztaszeri M, Braunschweig R, Mihaescu A. Pleomorphic adenoma with predominant plasmocytoid myoepithelial cells: a diagnostic pitfall in aspiration cytology. Case report and review of the literature. Diagn Cytopathol 2009; 37: 56-60.
17. Das DK, Haji BE, Ahmed MS, Hossain MN. Myoepithelioma of the parotid gland initially diagnosed by fine needle aspiration cytology and immunocytochemistry: a case report. Acta Cytol 2005; 49: 65-70.