**Letter to Editor**

**Apocrine hidrocystoma with mucinous metaplasia**

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Hidrocystoma is a rare cystic lesion arising from the sweat glands, and is classified into apocrine and eccrine variants, with the majority being of apocrine nature [1]. It is usually found in the head and neck region, and commonly affects the periorbital area [1, 2]. Apocrine hidrocystoma is characterized histopathologically by the presence of a unilocular or multilocular cyst situated in the dermis, and the cyst wall is covered by a double layer of epithelial cells [1, 2]. The inner layer is composed of columnar cells with rich eosinophilic cytoplasm which shows luminal decapitation secretion, and the outer layer is consisted of flat myoepithelial cells [1, 2]. Occasionally, papillary projection of the epithelium into the lumen is observed, which is referred to as papillary apocrine gland cyst [1].

Mucinous metaplasia is an extremely rare phenomenon in the skin. In non-neoplastic skin, the most common lesion is mucinous syringometaplasia, which is characterized by the presence of epidermal invagination lined by non-keratinizing squamous cells and mucin-laden goblet-like cells accompanied by mucinous changes in the eccrine ducts [3-7]. This lesion is commonly seen in the plantar surface of feet and fingers [3-7]. Moreover, albeit extremely rare, non-neoplastic squamous epithelium with mucinous metaplasia of the external genitals has also been reported [8]. In neoplastic skin lesions, only a few cases of clear cell hidradenoma, hidradenocarcinoma, and in situ and invasive squamous cell carcinomas with mucinous metaplasia have been documented [9-14]. Herein, we report the first documented case of apocrine hidrocystoma with mucinous metaplasia and review the literature.

A 29-year-old Japanese female presented with a nodular lesion in the right subaural region, which had been noticed approximately 7 years earlier. Physical examination revealed a well-circumscribed elastic soft subcutaneous nodule, measuring 2 × 1.5 cm in diameter. Total resection of the nodule was performed under a clinical diagnosis of hemangioma.

Histopathological study of the resected specimen revealed a well-circumscribed unilocular cyst in the dermis. The cyst wall was lined by a double layer of epithelial cells (**Figure 1A**). The inner layer was composed of columnar cells with rich eosinophilic cytoplasm which showed luminal decapitation secretion, and the outer layer was consisted of flat myoepithelial cells [1, 2]. Occasionally, papillary projection of the epithelium into the lumen is observed, which is referred to as papillary apocrine gland cyst [1].

Immunohistochemical studies were performed using an autostainer (Ventana) by the same method as previously reported [15-19]. Cytokeratin 7 was expressed in the columnar cells, but cytokeratin 20 was not. Gross cystic disease fluid protein (GCDFP)-15 was also diffusely positive in these columnar cells (**Figure 2**).
Accordingly, an ultimate diagnosis of apocrine hidrocystoma with mucinous metaplasia was made.

Although normal human conjunctival epithelium has mucinous cells, none of these cells are observed in normal human skin. The presence of mucinous cells in non-neoplastic skin tissue and some kinds of cutaneous neoplasms is regarded as a metaplastic phenomenon [12]. Mucinous syringometaplasia is thought to be the result of long-standing pressure or trauma [3], and mucinous metaplasia of the external genital non-neoplastic squamous epithelium is also considered to be associated with chronic inflammation [8]. Moreover, only a limited number of cases of cutaneous neoplasms, such as hidradenoma, hidradenocarcinoma, and in situ and invasive squamous cell carcinoma, with mucinous metaplasia have been documented [9-14]. Mucinous metaplasia occurring in the above-mentioned lesions has been phylogenetically interpreted as an atavism, because mucinous cells are frequently observed in the apocrine glands of lower vertebrate and certain mammals [3]. Although only one case of eccrine mucinous metaplasia adjacent to apocrine hidrocystoma occurring in a 13-year-old girl has been reported [20], this case is the first documented case of apocrine hidrocystoma with mucinous metaplasia.

The mucin of the present case was considered as acid mucopolysaccharides because the mucinous cells were positive for both Alcian blue and PAS stainings. This finding corresponded to the histochemical analyses of the mucinous material of the previously reported cases of mucinous syringometaplasia and cutaneous neoplasms with mucinous metaplasia [3-9, 14, 20].

In conclusion, this case demonstrates that mucinous metaplasia occurs in apocrine hidro-
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cystoma. Mucinous metaplasia is an extremely rare phenomenon in both non-neoplastic skin tissue and cutaneous neoplasms. Additional studies are needed to clarify the histogenesis and molecular mechanism of mucinous metaplasia of the skin.

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