

A Case of Papillary Eccrine Adenoma

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Papillary eccrine adenoma is a rare sweat gland tumor with a characteristic histopathological appearance and has a benign clinical course. Five cases were reported in Korea since a report by Song et al. in 1988.

We report a case of papillary eccrine adenoma in a 38-year-old male who had a firm, dark brown nodule on the medial surface of the left thigh. Microscopically, the tumor was composed of multiple dilated ducts lined by two or more layers of epithelial cells and the luminal cells showed papillary projections into the lumen in some tubules. Immunoperoxidase staining was positive for CEA, S-100, and EMA.

We excised the lesion completely and no evidence of recurrence was observed for 6 months. (*Ann Dermatol* 9:(2) 163~166, 1997).

Key Words : CEA, S-100, EMA, Papillary eccrine adenoma

Papillary eccrine adenoma is a rare benign sweat gland tumor first described by Rulon and Helwig in 1977¹. It is more common in females than males and presents principally as a dermal nodule or plaque. The extremities are the most frequent sites. It ranges in size from 0.5 to 2.0 cm. It has a benign clinical course and surgical excision is the choice of treatment.

Histologically, the tumor is characterized by the presence of multiple dilated ducts of varying sizes with branching and budding seen in longitudinal sections. The ducts are lined by two or more layers of epithelial cells and some tubules contain eosinophilic amorphous material.

We report a case of papillary eccrine adenoma that was studied by light microscopy and immunohistochemical methods, and the differential diagnosis with other cutaneous neoplasms that show similar histological features are discussed.

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REPORT OF A CASE

A 38-year-old male patient presented at our clinic with a nodule on the medial surface of the left thigh. The lesion had been present for 20 years and was asymptomatic. Examination revealed a firm, dark brown nodule with a smooth surface (Fig. 1). The tumor measured 1.0 × 0.8 cm and was slightly elevated. The clinical diagnosis was a hypertrophic scar or foreign body granuloma. The lesion was completely excised and no evidence of recurrence was observed for 6 months.

Microscopically, a well-circumscribed, unencapsulated dermal tumor was composed of multiple dilated ducts of varying sizes (Fig. 2). The ducts were lined by two or more layers of epithelial cells. The luminal cells were cuboidal, columnar or flattened and showed papillary projections protruding into the lumen in some tubules (Fig. 3). Some of the lumens contained eosinophilic granular materials positive for periodic acid-Schiff (PAS) and alcian blue. The luminal cells did not show decapitation secretion.

Immunoperoxidase staining was positive for S-100 protein in the cytoplasm of the epithelial cells of

Fig. 1. 1.0 × 0.8 cm sized firm, dark brown nodule with smooth surface on medial surface of the Lt. thigh.

Fig. 2. Multiple dilated ducts of varying sizes and some tubules filled with eosinophilic amorphous material(H & E stain, × 40).

Fig. 3. The tubules are lined by two or more layers of epithelial cells and showed papillary projections protruding into the lumen(H & E stain, × 100).

Fig. 4. Positive immunoperoxidase stain for CEA in the luminal cytoplasm and the intraluminal material(× 200).

the tubules(Fig. 4). Carcinoembryonic antigen (CEA) showed intense immunoreactivity with the cytoplasm of most epithelial cells and with the luminal materials(Fig. 5). The epithelial membrane antigen (EMA) also showed immunoreactivity with the cytoplasm of the epithelial cells lining the ducts and with the intraluminal secretions(Fig. 6).

DISCUSSION

In 1977, Rulon and Helwig reported 14 cases of a benign sweat gland tumor that they named papillary eccrine adenoma. They separated papillary eccrine adenoma from tubular apocrine adenoma described by Landry and Winkelmann². Rulon and Helwig had emphasized the benign clinical course and typical histological features. They considered

Fig. 5. Immunoperoxidase staining of neoplastic ducts with S-100 protein was positive ($\times 200$).

with assurance that the surgical excision and complete removal was the choice of treatment. Since then, about 30 cases have been mentioned in the literature. In Korea, Song et al. reported 3 cases of papillary eccrine adenoma in 1988³. They considered papillary eccrine adenoma to be of eccrine origin. In 1989, Rhee et al. reported a case of papillary eccrine adenoma in a young female who presented with a single intradermal tumor on the right postauricular area⁴. Thereafter, Kim et al. performed immunohistochemical studies with S-100 protein and CEA, and they also showed that papillary eccrine adenoma differentiated toward the eccrine secretory coil⁵.

Clinically, papillary eccrine adenoma appears to be more common in black people and in females¹. Papillary eccrine adenoma presents principally as a dermal nodule with a well demarcated margin on the distal extremities.

Histologically, papillary eccrine adenoma shows distinctive pathological features. The tumor is composed of multiple dilated ducts and shows in some tubules papillary projections protruding into the lumen. Some tubules are filled with eosinophilic granular material. In our case, PAS and alcian blue staining for eosinophilic material was positive. It is identical in previously reported cases of papillary eccrine adenoma in Korea^{3,4}.

Immunohistochemical stainings showed positivity for CEA^{6,7,8,9,10}, S-100 protein^{7,8} and EMA^{7,8,9,10}. CEA is a glycoprotein found in eccrine and apocrine structures of normal skin, as well as in benign and malignant sweat gland tumors^{11,12,13}. However, it

Fig. 6. Immunoperoxidase stain for EMA in the cytoplasm of the epithelial cells lining the ducts and intraluminal secretions ($\times 200$).

does not differentiate between eccrine and apocrine origin. S-100 protein is found in eccrine secretory coils, thus the neoplasms containing S-100 protein may be related to the eccrine coil⁴. EMA is present in normal eccrine and apocrine secretory epithelium¹⁵. Therefore, positivity for EMA may indicate that it originates from the glandular epithelium of the sweat gland.

The differential diagnosis of papillary eccrine adenoma includes aggressive digital papillary adenoma, apocrine hidrocystoma, hidradenoma papilliferum, syringoma and tubular apocrine adenoma. Aggressive digital papillary adenoma is located at the extremities, particularly on the fingers and toes of elderly patients. It shows more infiltrative characteristics with a more nodular and solid architectural configuration.

Apocrine hidrocystoma usually occurs as a solitary translucent nodule of cystic consistency, and may also show papillary projections into cystic spaces. The inner surface of the wall and the papillary projections are lined by a row of secretory cells showing decapitation secretion.

Hidradenoma papilliferum occurs only in women, usually on the labia majora or in the perineal or perianal region. The tumor represents an adenoma with apocrine differentiation. It is located in the dermis, is well circumscribed, is surrounded by a fibrous capsule. Within the tumor, there are cystic and tubular structures with papillary folds projecting into cystic spaces, but the epithelial cells of the luminal layer show active decapitation secretion as seen in the secretory cells of apocrine glands.

Syringoma exhibits cystic sweat ductal structures embedded in a fibrous stroma. The walls of which are lined by two rows of epithelial cells. Some of the ducts possess small, comma-like tails of epithelial cells, giving them the appearance of tad poles.

Finally, papillary eccrine adenoma must be differentiated from tubular apocrine adenoma. Papillary eccrine adenoma was initially thought to be the same as tubular apocrine adenoma² until Rulon and Helwig described the tumor as distinct clinical entity. At present, there is a controversy of the relationship between the papillary eccrine adenoma and the tubular apocrine adenoma. Some authors consider that they can be regarded as identical tumors^{16,17}, but others do not agree¹⁸. Papillary eccrine adenoma has sometimes both eccrine and apocrine differentiation in a tumor. The term tubulopapillary hidradenoma^{6,19} or tubular papillary adenoma has been proposed to encompass both papillary eccrine adenoma and tubular apocrine adenoma. In our case, however, we did not see the typical findings of tubular apocrine adenoma such as a decapitation secretion.

We experienced a case of papillary eccrine adenoma which has distinct histological features and a benign clinical course, and immunohistochemical stainings for CEA, S-100 protein and EMA were all positive. The results of the immunohistochemical stains are in agreement with previous reports^{6,7,8,9,10}, thus we also considered that papillary eccrine adenoma differentiates toward the eccrine secretory coils and is a separate entity from tubular apocrine adenoma.

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