A Case of Papillary Tubular Adenoma (Tubulopapillary Hidradenoma)

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Tubular apocrine adenoma and papillary eccrine adenoma are rare sweat gland neoplasms that appear as a small solitary lesion on the scalp or extremities, respectively. Although these lesions are thought to be distinct entities, there are enough similarities between them to group them under the term tubulopapillary hidradenoma or papillary tubular adenoma.

We describe a case showing many tubular structures with papillary projection, syringocystadenoma-like structures, and eccrine hirocystoma-like structures in the axillary area. The term of papillary tubular adenoma or tubulopapillary hidradenoma may be prefered in this case.

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Key Words : Tubular apocrine adenoma, Papillary eccrine adenoma, Tubulopapillary hidradenoma, Papillary tubular adenoma

Tubular apocrine adenoma (TAA) and papillary eccrine adenoma (PEA) are rare sweat gland neoplasms that appear as a small solitary lesion on the scalp or extremities, respectively, and have usually been present for many years before being biopsied[1,2]. Fox and Cotton[3] reported a case of tubulopapillary hidradenoma developed in the axilla and demonstrated both eccrine and apocrine differentiation. They suggested that their case had arisen from the apoeccrine gland located in the axillary areas.

We report a case of papillary tubular adenoma (PTA) or tubulopapillary hidradenoma (TPH) that was studied by light microscopy and immunohistochemical methods and discuss the origin of this rare tumor.

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CASE REPORT

A 61-year-old male patient was referred from the department of cardiology to evaluate the skin lesion in the axillary area. The lesion had been present for 5 years and was aymptomatic. Examination revealed a firm, erythematous, walnut-sized nodule with a central punctum in the left axillary area (Fig. 1). He has suffered from coronary arterial disease for 10 years. The lesion was completely excised and no evidence of recurrence was observed for 5 months. Histopathological examinations revealed a relatively well-circumscribed, unencapsulated dermal tumor 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 almost all the tubules (Fig. 3). In one of the several sections, syringocystadenoma (SCAP)-like features were seen. One cystic invagination with numerous papillary projections extended downward from the epidermis (Fig. 4). The papillary projections and the lower portion of the invaginations were lined by glandular epithelium often consisting of two rows of cells. Occasionally,

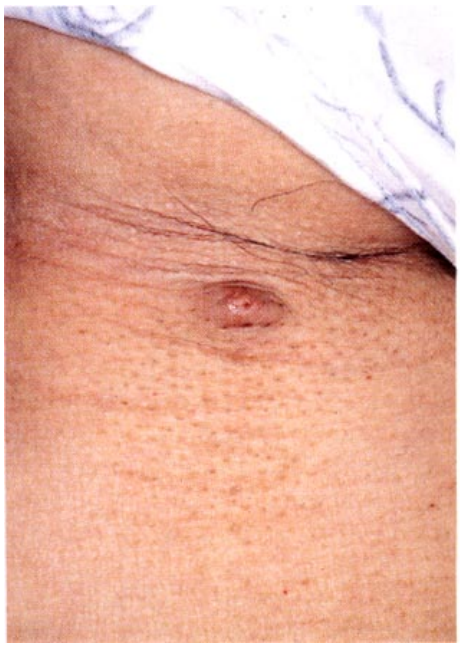

Fig. 1. A firm, erythematous, walnut-sized nodule with a central punctum in the left axillary area.

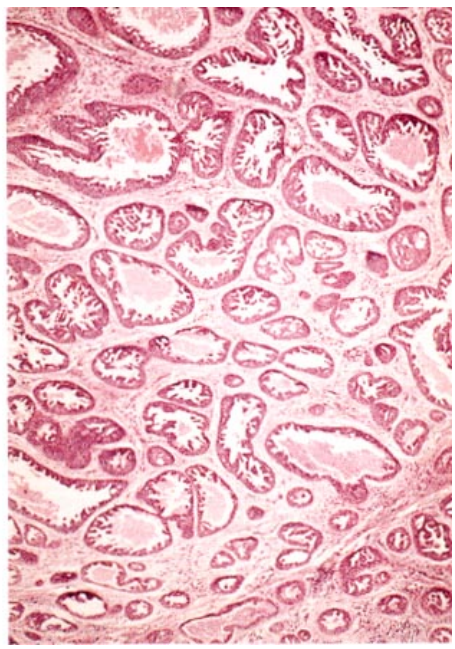

Fig. 2. Multiple dilated ducts of varying size in the dermis (H & E, ×40).

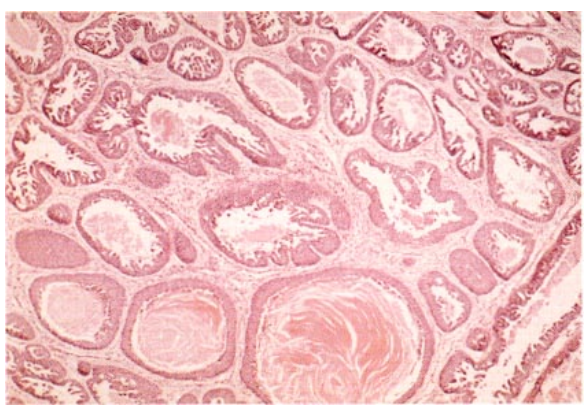

Fig. 3. The ducts are lined by two or more layers of epithelial cells with papillary projection (H & E, ×100).

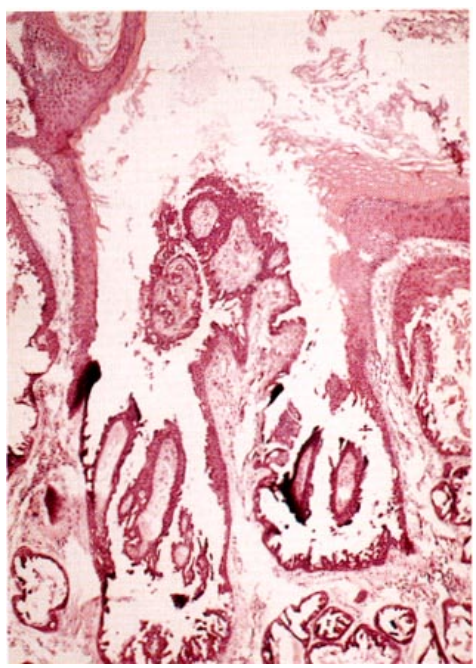

Fig. 4. Syringocystadenoma-like structure is seen (H & E, ×40).

the luminal row of cells showed decapitation-like secretion (Fig. 5). Beneath the cystic invagination, tubular structures with papillary projections were seen. In the deeper dermis, several dilated cystic structures reminiscent of eccrine hidrocystoma were also seen (Fig. 6). The epithelial cells were strongly positive with anti-cytokeratin antibody (Fig. 7a). The luminal side of the duct-like tubules, the cellular debris, and the intraluminal substances reacted positively with the anti-carcinoembryonic antigen (CEA) antibody (Fig. 7b). Luminal cells of the tubules were stained faintly with the anti-epithelial membrane antigen (EMA) antibody (Fig. 7c). S-100 protein was detected in the peripheral layer of the tubular structures

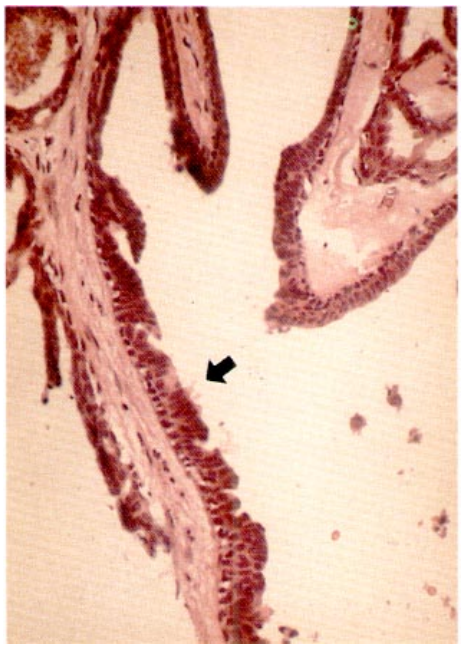

Fig. 5. The papillary projections and the lower portion of the invaginations are lined by glandular epithelium consisting of two rows of cells. The luminal row of cells occasionally show decapitation-like secretion (arrow) (H & E, ×200).

and stroma (Fig. 7d). Gross cystic disease fluid protein-15 (GCDFP-15) was negative even in the SCAP-like structures.

DISCUSSION

TAAs are usually found in the scalp and they have been designated an apocrine histogenesis on the basis of their ultrastructural characteristics, enzyme, and immunohistochemical phenotype[1,3,4]. Rulon and Helwig[2] reported on a series of tumors predominantly found on the distal limbs that, although similar to TAAs, were thought to have an eccrine derivation and were correspondingly termed PEAs. Although PEAs differ from TAAs in their lack of decapitation secretion, the resemblance to syringomas, and enzyme histochemistry together with some electron-microscopic appearances of eccrine derivation, many similarities seem to exist to group them under a single term, TPH or PTA[3,5-7]. Our case illustrates light microscopical and immunohistochemical evidence of both apocrine and eccrine differentiations. In most of the reported TAAs, the tumor was connected to the overlying epidermis and was necessary to differentiate the lesions from SCAP[8]. However, some authors thought that TAA and SCAP might occur together, especially when they are preceded by an organoid nevus, and they might represent a spectrum of disease[8-10]. In our case, SCAP-like structure was seen as a central punctum clinically. There is no unanimity about the direction of differentiation in SCAP[11]. Features of both apocrine and eccrine differentiation can be seen in particular examples. It is likely that, rather than arising from mature structures, SCAP arises from pluripotential cells with the potential to develop into primary epithelial germ structures of a variety of different types[11]. In immunohistochemical studies, a cytokeratin stain showed strong positivity in almost all the epithelial cells of tubular structures and CEA in the luminal cells and intraluminal substances. EMA was also positive. CEA has been found in eccrine and apocrine structures of normal skin, as well as benign and malignant sweat gland tumors. It cannot differentiate between eccrine and apocrine secretory epithelium but not in eccrine or apocrine ducts[12]. Its presence in our case supports its differentiation toward glandular epithelium. S-100 protein is found in eccrine secretory coils but not in eccrine ducts or acrosyringia[12,14]. S-100 was also present in myoepithelial cells of the secretory portion of apocrine glands and some apocrine neoplasms[15]. Although positivity for S-100 protein and EMA in our case indicates that it originates from glandular epithelium of sweat glands, differentiation toward the eccrine or apocrine route remains unclear. GCDFP-15, which was initially thought to be specific for the apocrine glands and tumors with

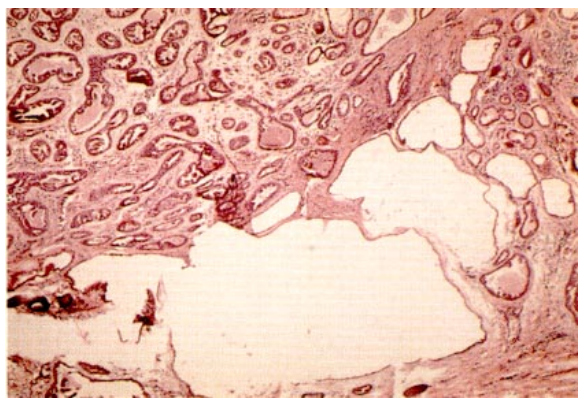

Fig. 6. Several eccrine hidrocystoma-like dilated cyctic structures are seen in the deeper dermis (H & E, ×40).

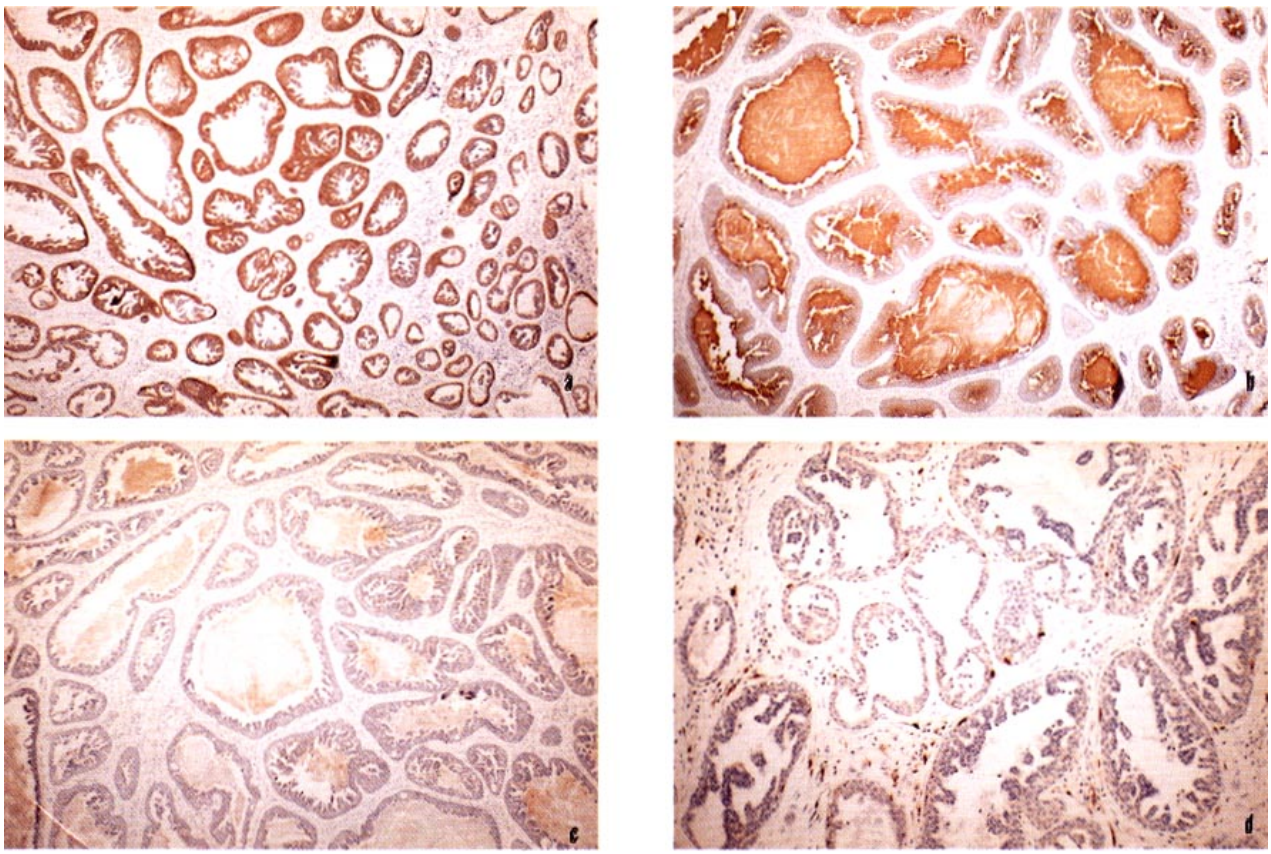

Fig. 7. Positive staining for cytokeratin (a, ×40), CEA (b, ×100), EMA (c, ×100), and S-100 (d, ×100).

apocrine differentiation16, was negative in almost all the tubular structures, even in the SCAP-like structures. Recently, GCDFP-15 is thought to be positive in the secretory components of the gland in apocrine and eccrine glands[17]. Negative result for GCDFP-15 in our case led to the speculation that GCDFP-15 is not a specific apocrine or eccrine marker.

The recently described apoeccrine sweat gland may provide an explanation for the appearance of both apocrine and eccrine differentiation in our case. This sweat gland, which develops after puberty and makes up 45% of the sweat gland number in the adult axilla, shows light and electronmicroscopic features of both apocrine and eccrine differentiation[18]. There is no literature on its distribution away from the axilla. It has been suggested that under appropriate tumorigenic stimuli, neoplasms that are originally eccrine may exhibit apocrine differentiation and vice versa[19]. In our case, the location was in the axillary area. The location and histopathological findings support that our case may have arisen from the apoeccrine gland located in the axilla. PEA appears to be more common in black people and is located mostly in the distal part of the extremities[12]. In Korea, 5 cases of PEA were described20-23. Those cases were considered to be of eccrine origin.

We feel that the terms of PTA or TPH may be prefered in our case, especially in the case developed from the axillary areas where apoeccrine glands are plentiful.

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