

A Case of Tubular Apocrine Adenoma

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Tubular apocrine adenoma(TAA) is a rare benign neoplasm usually found on the scalp. It has been designated as an apocrine histogenesis on the basis of its ultrastructural characteristics, enzyme, and immunohistochemical phenotype.

Histopathologically the neoplasm consists of tubules or cysts, which show signs of apocrine secretion and sometimes needs to be differentiated from papillary eccrine adenoma.

We report a typical case of TAA on nostril which shows differentiation toward apocrine in nature. (Ann Dermatol 14(2) 102-105, 2002).

Key Words : Tubular apocrine adenoma, Nostril

TAA is a relatively rare skin appendage tumor which was first described by Landry and Winkelmann in 1972¹. It is a distinctive benign neoplasm with apocrine differentiation in which epithelial elements mostly assume the shape of tubules and cysts. The neoplasm is difficult to differentiate histologically from papillary eccrine adenoma (PEA) in some cases. Falck and Jordaan² proposed to include TAA and PEA under a single heading, tubulopapillary hidradenoma because there are enough similarities between these two entities such as tubular structures with papillary projections into lumina.

Herein, we report a case of TAA that occurred on nostril, which is a very unusual site and showed characteristic signs of apocrine secretion.

CASE REPORT

The patient was a 58-year old Korean woman who presented with a slow-growing nodule on the left nostril which had been present for 6 years. It was

asymptomatic and sometimes bled after irritation.

Examination revealed skin colored to erythematous, firm, 0.7x0.5 cm sized, lobulated nodule on the left nostril (Fig. 1). Family history was denied.

The lesion was completely excised. Histologic examination revealed a well-defined, unencapsulated dermal tumor consisting of numerous irregularly dilated tubules and cyst of varying sizes. The tubules were comprised of two rows of epithelial cells. Outer layer consisted of cuboidal or flattened cells, with the luminal columnar cells showing decapitation secretion. Amorphous eosinophilic material was seen in the lumina (Fig. 2). The luminal border and the intraluminal substances reacted positively with the D-PAS (Fig. 3). The epithelial cells were strongly positive with anti-cytokeratin antibody (Fig. 4). Luminal cells of the tubules were stained with the anti-epithelial membrane antigen (EMA) antibody (Fig. 5). S-100 protein was detected in the peripheral layer of the tubular structures and stroma (Fig. 6). In following up for 12 months, there is no evidence of recurrence.

DISCUSSION

TAA is a benign sweat gland neoplasm comprising tubules and cysts showing signs of apocrine secretion and papillation into the lumina is commonly observed. Clinically it presents as a single, well circumscribed nodule that displays a verrucous, sometimes eroded, surface usually found on the scalp³. However, involvement of perianal area,

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Fig. 1. Asymptomatic 0.7×0.5 sized, skin colored to erythematous lobulated nodule on left nostril.

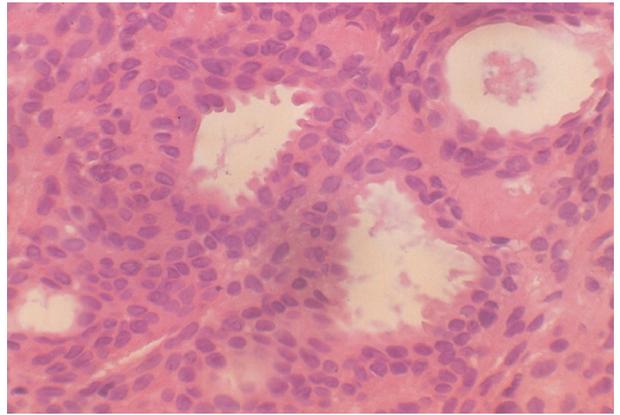


Fig. 2. The outer layer consists of cuboidal or flattened cells and the inner layer is composed of columnar cells and shows apparent decapitation secretion in the lumen (H&E $\times 400$).

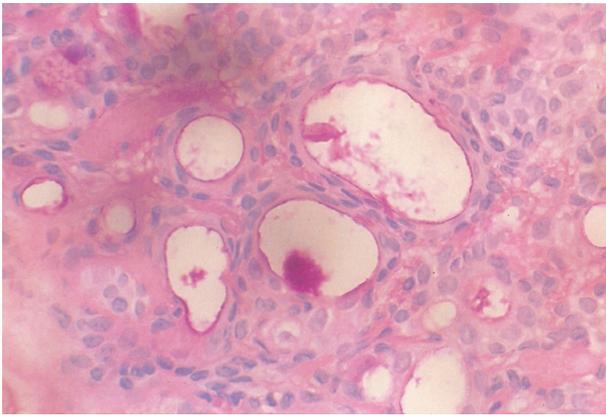


Fig. 3. The luminal border and the intraluminal substances are positively stained with the D-PAS (D-PAS $\times 400$).

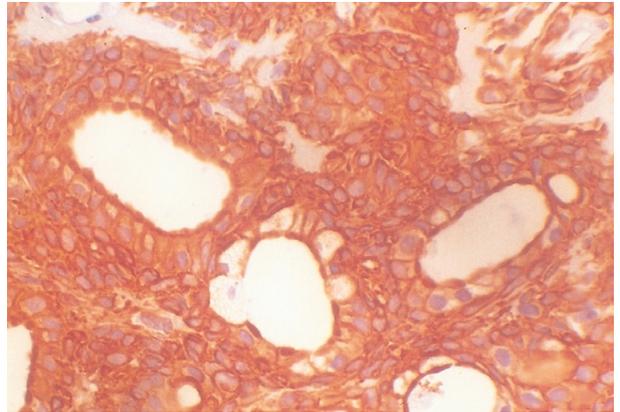


Fig. 4. The epithelial cells were strongly positive for the anti-cytokeratin antibody (cytokeratin $\times 400$).

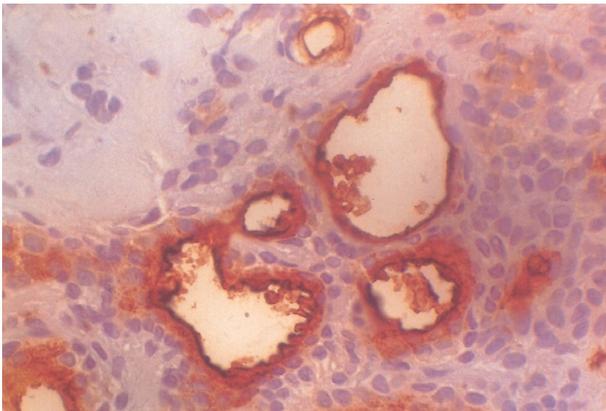


Fig. 5. Luminal cells of the tubules are positive for the anti-epithelial membrane antigen (EMA) antibody (EMA $\times 400$).

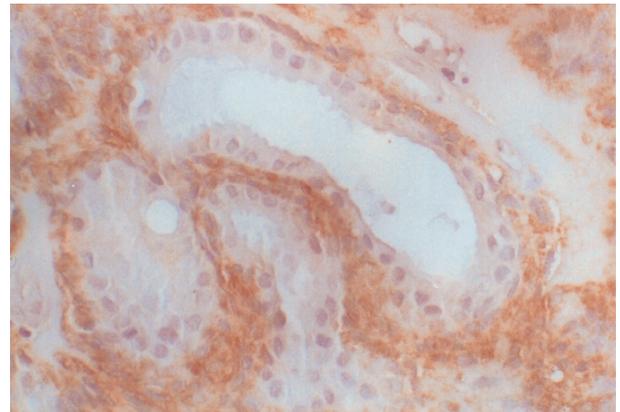


Fig. 6. S-100 protein was detected only in the peripheral layer of the tubular structures and stroma (S-100 $\times 400$).

trunk, and axilla has also been reported^{4,5}.

In the Korean literature, Kang *et al*⁶ reported two cases of TAA. One patient was a male who had had a finger-tip sized cystic mass on the scalp, and the other patient was a female who had had a dark red hard nodule on the left forearm. And You *et al*⁷ reported a case of multiple TAA, which appeared as three separated nodules on the right deltoid area. In our patient, the lesions occurred on the nostril, which is a very uncommon site.

The occasional association of TAA with nevus sebaceous and syringocystadenoma papilliferum (SCAP) suggests the hamartomatous character^{1,4,8}.

Fisher⁹ criticized that this neoplasm simply represented a variant of SCAP because of location on the scalp, concurrence with what was interpreted as basal cell carcinoma, two layers of epithelial cells lining tubules, arrangement in tubules, plasma cell in the stroma and connection to the skin surface. However, according to Umbert and Winkelmann¹⁰, TAA was different histopathologically from SCAP in that neither invaginations nor papillary projections into lumina was seen in TAA, and they suggested that TAA is an independent clinical entity consisting of a benign appendage tumor of apocrine origin that is often associated with an organoid nevus.

PEA, first reported in 1977 by Rulon and Helwig¹¹, has similar histological features with TAA, and some authors^{2,12} propose that a single term, tubulopapillary hidradenoma be employed for both lesions.

The histopathological feature is the presence of numerous irregularly shaped tubular structures that are lined by two layers of epithelial cells. The peripheral layer consists of cuboidal or flattened cells, and the luminal layer is composed of columnar cells with characteristic decapitation secretion¹³.

Several immunohistochemical studies have been done which supported either apocrine or eccrine differentiation of the neoplasm^{2-5,8-18}. The findings in favor of apocrine differentiation include connection of some tubular structures of the neoplasm to pre-existing inflammation, presence of apocrine secretion in cells along the luminal border that lined tubules, and immunohistochemical positivity of human milk fat globule and GCDFP-15, positivity for acid phosphatase, lysozyme, α 1- antichymotrypsin and negativity for phosphorylase^{5,18}.

The findings in favor of eccrine differentiation seem to be positivity of some of the neoplastic cells for S-100 protein and ferritin¹⁷. In our case, definite decapitation secretion was noted on the luminal cells of tubules, and immunohistochemically, cells comprising tubules were positive for cytokeratin and EMA. S-100 was negative in luminal cells but weakly positive in cells of outer layer of the tubules, which might be exhibiting myoepithelial cells of the secretory portion of the apocrine gland.

TAA can be determined to be benign on the basis of symmetry, sharp circumscription, smooth borders of tubules. Complete surgical excision is always curative and no metastasis has ever been reported. Burket and Zelickson¹⁹ reported a case of TAA which showed tubules formed by neoplastic cells in perineural space and between thick bundles of collagen, and they proposed that TAA might not be benign but behave in a quietly aggressive fashion, so wide excision should be recommended.

Our case should be considered apocrine in nature on the basis of the presence of apocrine secretion in cells that line lumina and negativity for S-100 protein of tumor cells except some cells of peripheral layer. It is also interesting that the lesion in our patient occurred on nostril which is a very uncommon site for this tumor.

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