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Case Report

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Sebaceous Induction Associated With Syringocystadenoma Papilliferum and Apocrine Hidrocystoma

Gülşen Akoğlu,[1] MD, Sibel Orhun,[2] MD, Çağla Çiçek,[3] MD

Address: [1]Dermatovenereology Reconstructive and Aesthetic Surgery, Ataturk Training and Research Hospital, Ankara, [2]Pathology Reconstructive and Aesthetic Surgery, Ataturk Training and Research Hospital, Ankara,

[3]Plastic Reconstructive and Aesthetic Surgery, Ataturk Training and Research Hospital, Ankara

E-mail: gusemd@yahoo.com

**Corresponding Author:* Dr. Gulsen Akoglu, Dermatovenereology, Ankara Ataturk Training and Research Hospital, Bilkent, Ankara

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Abstract

Observation: Syringocystadenoma papilliferum and apocrine hidrocystoma are benign cutaneous tumours which usually occur separately. Sebaceous induction is usually defined in dermatofibromas and melanocytic nevi. However, its association with SCAP and apocrine hidrocystoma has not been reported before. Herein, we describe a 19-year-old female patient presented with a painless reddish nodule over a yellowish alopecic plaque on her scalp. Histopathological examination showed papillomatosis with a marked inflammatory infiltrate with numerous plasma cells in the stroma and many multifocal and multi-locular cystic apocrine hidrocystomas. Below the epidermis, immature sebaceous glands, positive for epidermal growth factor receptor and without an association of a normal hair follicle, were detected. We may suggest that sebaceous induction may accompany syringocystadenoma papilliferum and apocrine hidrocystoma in a single cutaneous lesion.

Introduction

Syringocystadenoma papilliferum (SCAP) and apocrine hidrocystoma are benign cutaneous tumours which usually occur separately [**1, 2**]. Sebaceous induction is usually defined in dermatofibromas and melanocytic nevi [**3**]. However, its association with SCAP and apocrine hidrocystoma in a single cutaneous lesion has not been reported before.

Case Report

A 19-year-old female patient was admitted to our dermatology outpatient clinic with the complaint of painless nodule on her scalp. The lesion has emerged about one year ago over a hairless area which was present since birth. The patient did not express any trauma or bleeding over the lesion. She did not have any systemic disease. Dermatological examination revealed a reddish nodule with 1 cm diameter on a rough yellowish thin alopecic plaque with irregular shape measuring about 3x2 cm in size (**Figure 1**). Lymphadenopathy or organomegaly were not detected on physical examination.

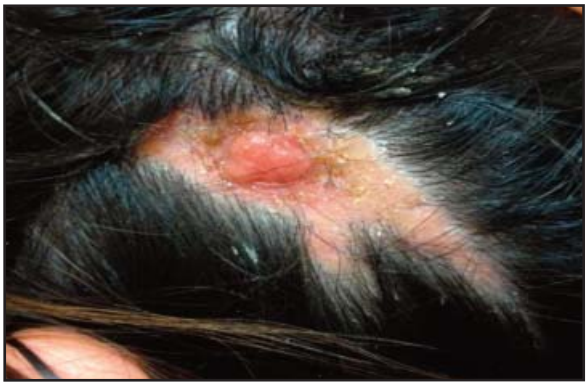


Figure 1. Whole cutaneous lesion on the scalp

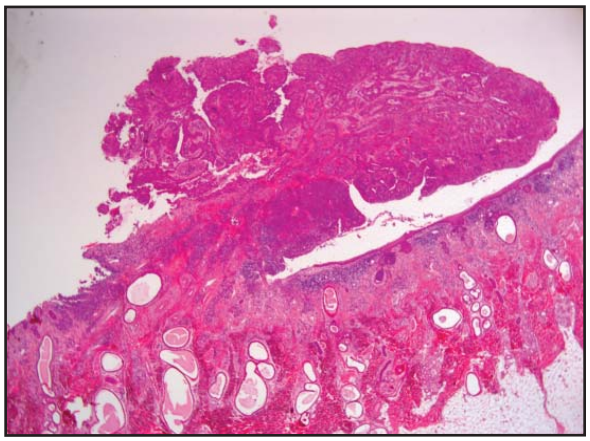
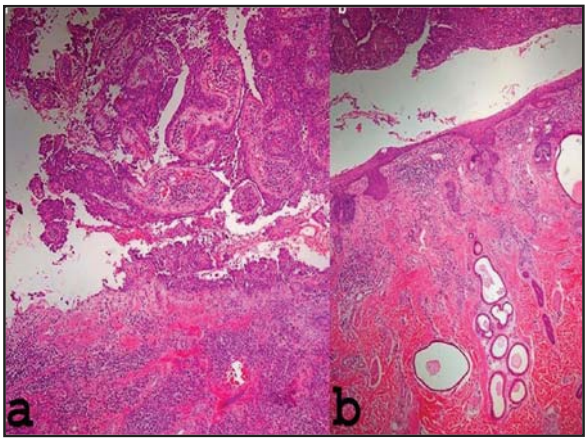


Figure 2. Panaromic picture of the lesion (H&E, x20)



Figures 3a and b. (**a**) Papillomatous SCAP lesion lined by columnar epithelium in the luminal side and cuboidal epithelium in the outer side with plasma rich stroma (H&E, x40); (**b**) Dermal numerous, close cystic apocrine hidrocystoma lesions beneath the papillary projections and immature sebaceous structures (H&E, x40)

The whole lesion was totally excised. Histopathological examination showed papillomatosis with a marked inflammatory infiltrate with numerous plasma cells in the stroma. Luminal layer of papillary projections was lined with columnar cells and the peripheral layer consisted of cuboidal and flattened cells. Focal squamous metaplasia was detected. Beneath the papillary formation of this SCAP lesion, many multifocal and multi-locular cystic apocrine hidrocystoma areas were observed. Below the epidermis, rudimentary sebaceous glands without an association of a normal hair follicle were detected (**Figures 2 and 3a and b**). The immature sebocytes were positive with anti epidermal growth factor receptor (EGFR) antibody (**Figures 4 and 5**). Depending on clinicopathological features, the patient was diagnosed as having SCAP associated with apocrine hidrocystoma and sebaceous induction.

Discussion

SCAP is a rare tumour which is usually located in scalp of children or adolescents. Mainly apocrine and less frequently eccrine histogenesis is considered to involve in pathogenesis. SCAP consists of invaginated duct-like structures lined by squamous epithelium with a transition to double-layered cuboidal and columnar epithelium having a stroma rich of plasma cells. The dilated ducts may form cystic spaces or villous projections [1]. SCAP was proposed to be a hamartoma which is abnormally arranged with follicular infundibular, apocrine glandular and ductal epithelium together with sebaceous structures [4]. About one third of cases are found to be associated with an organoid nevus [1]. SCAP may coexist with basal cell carcinoma, verrucous carcinoma, verruca or eccrine and apocrine tumours such as tubular apocrine adenoma,

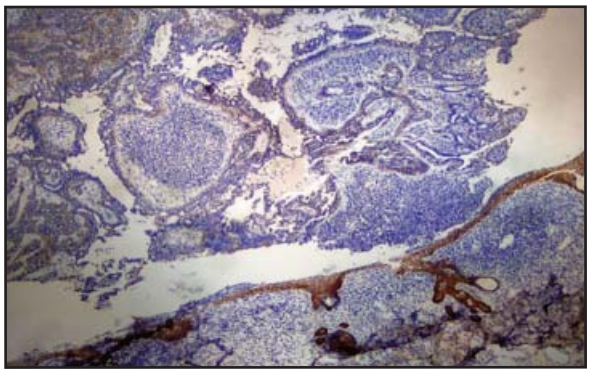


Figure 4. EGFR negative SCAP and apocrine hidrocystoma (x40)

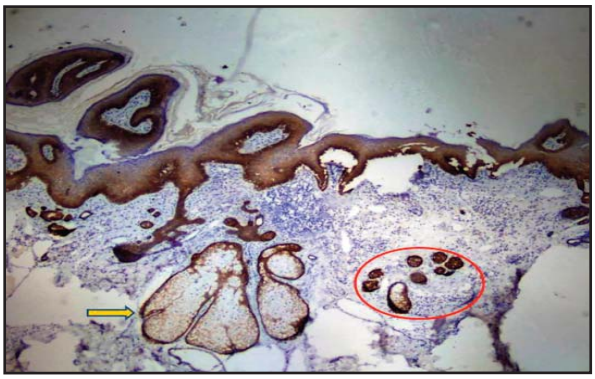


Figure 5. Sebaceous induction area staining positively with EGFR (circled); mature sebaceous structures stained positively with EGFR only at the periphery (arrow) (x40)

apocrine hidrocystoma, and papillary eccrine adenoma [**1, 5, 6**]

SCAP, apocrine hidrocystoma, and tubular papillary adenoma may form complex cutaneous tumours. Since histopathological features of 2 or 3 types may co-exist in a single lesion, the term "tubulocystic adenoma with apocrine differentiation" is proposed to define these cutaneous tumours [**2**]. Arias-Santiago et al. suggest that the association of SCAP with apocrine hidrocystoma arise from multipotent undifferentiated cell contents which then differentiate toward two lines of differentiation. On the other hand SCAP may be the only histopathological type which active primitive cells into the apocrine hidrocystoma [**5**].

Nevus sebaceous is the most important differential diagnosis in our case. Nevus sebaceous is considered to be a complex hamartoma involving sebaceous glands as well as other a dnexal structures. The structure of the sebaceous glands shows age-related alterations in nevus sebaceous. A decrease in number or complete absence of sebaceous is seen in 10-20% of patients, usually in infant patients. Sebaceous hyperplasia is the most common presentation of sebaceous glands, which is mostly seen in puberty. The connection of small sebaceous lobules or well-developed hyperplastic sebaceous glands to the surface epidermis or the infundibular region of the follicules is the typical feature of nevus sebaceous. Besides, sebaceous lobules may have holes and glands connect directly to the epidermal surface or to the infundibular area [**7**]. When compared, the histopathological structures of sebaceous glands of our case had distinct features. In multiple sections of the whole lesion, sebaceous hyperplasia was not detected. Moreover, they were immature and reduced in number which was not compatible with the age of the patient. In addition, these rudimentary sebaceous glands were not connected to the surface epidermis or any follicular infundibulum and they did not have any cystic dilatations or holes. We consider that we encountered a different structure of sebaceous glands associated with SCAP and apocrine hidrocystoma. We evaluated these rudimentary glands as sebaceous induction.

Sebaceous induction is defined as two or more rudimentary sebaceous glands overlying the very superficial level of cutaneous lesion without a normal hair follicle [**3**]. Sebaceous i nduction has been described in the histopathological features of dermatofibromas and m elanocytic nevi. Although the underlying aetiology is not clear, growth factors secreted by dermatofibromas, anatomic site specific microenvironmental features [**3**], ectopic hedgehog signalling [**8**] and low levels of beta catenin stimulation [**9**] are suggested as possible mechanisms involving sebaceous induction. Strong expression of EGFR in the undifferentiated sebocytes at the periphery of human sebaceous glands was reported and EGFR and its ligands were considered to involve in the sebocyte differentiation and lipogenesis [**10**]. Although signalling mechanisms could not be investigated in our patient, we detected that immature sebocytes were positive for EGFR, suggesting the role of EGFR signalling in the formation of sebaceous induction.

To the best of our knowledge, this is the first report of sebaceous induction which accompanies SCAP and apocrine hidrocystoma in a single cutaneous lesion. Although the rudimentary sebaceous glands may be speculated to be a rare feature of nevus sebaceous, the age of our case and lack of typical glandular features do not support this suggestion. We suggest that sebaceous induction associated with SCAP and apocrine hidrocystoma may be coexistence or some growth factors released from SCAP lesion or surrounding tissue or changes in the microenvironment may induce multipotent cells to form immature sebaceous structures.

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