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Primary adenomyoepithelioma of the skin – a variant of apocrine mixed tumor?

Elisabeth Riedl, M.D.[1,2], Geoffrey J. Gottlieb, M.D.[3]

[1] Department of Dermatology, Medical University of Vienna, Vienna, Austria
[2] Department of Dermatology, Mount Sinai School of Medicine, New York, NY, USA
[3] Strata Pathology Services, Lexington, MA, USA

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Corresponding author: Elisabeth Riedl, M.D., Department of Dermatology, Mount Sinai School of Medicine, One Gustave L. Levy Place, Box 1047, New York, NY 10029. Email: elisabeth.riedl@meduniwien.ac.at.

ABSTRACT

Analogous to adenomyoepitheliomas of the breast, cutaneous adenomyoepithelioma is composed of two components, one being myoepithelial, the other ductal epithelial, and it lies in the spectrum of neoplasms having a "pure" myoepithelioma at one end and a mixed apocrine tumor at the other. We present the case of a 53-year-old woman with a 3 cm nodular lesion on her left forearm that had been present for many years. Histopathologic examination revealed a large lobulated neoplasm surrounded by a compressed fibrous pseudocapsule. Most of the cells that constituted the lesion displayed myoepithelial differentiation arranged in solid sheets, cords, and solitary units. Glandular and ductal structures with features of apocrine differentiation composed the second part of the neoplasm. The myoepithelial cellular component of the neoplasm stained for S100 protein and was negative for cytokeratin and carcinoembryonic antigen (CEA). Based on findings by conventional microscopy and immunohistochemistry, the neoplasm was classified as primary adenomyoepithelioma of the skin.

Introduction

Adenomyoepithelioma has been described as a rare benign neoplasm that occurs almost exclusively in the breast. In the breast it is defined as a neoplasm composed of two structures, namely, tubules limited by an inner epithelial layer of duct-like cells and an outer layer with mostly clear myoepithelial cells [1]. Apart from the breast, adenomyoepitheliomas have been described in the salivary glands and in the lung [2,3]. In the skin, these neoplasms seem to be exceedingly rare, with only few reports of adenomyoepitheliomas published to date [4-7]. We present the case of a 53-year-old woman with a clinically benign nodular cutaneous lesion that revealed histopathologic and immunohistochemical features of adenomyoepithelioma.

Case report

A 53-year-old woman presented to her dermatologist with a 3 cm asymptomatic nodule on the left forearm. The lesion had been present for several years. A trauma at the site years previously was reported. The biopsy specimen consisted of a 2.5 x 1.9 x 1.5 cm tan ellipse of skin and contained a hard yellow nodule measuring 1.5 cm in greatest dimension. Serial sections of tissue were prepared and stained with hematoxylin and eosin. Examination of the sections at scanning magnification revealed beneath an intact epidermis a zone of fibrosis within the upper part of the dermis (Figure 1 A, B). Beneath the scar, there was a large, lobulated neoplasm, which for the most part was surrounded by a compressed fibrous pseudocapsule. Each of the aggregates of neoplastic cells varied in sizes and shapes and some were large and nodular (Figure 1A–C). At higher magnification, most of the cells that constituted the lesion displayed myoepithelial differentiation with polygonal and "plasmacytoid" features (Figure 2). The cells presented sometimes as solid sheets, but also as cords and solitary units. In places glandular and ductal structures were evident; in some areas apocrine-type secretion within glandular structures was found (Figure 2 A–C). Focally, the myoepithelial cells contained a clear cytoplasm (Figure 2 D–F). In some areas cells were present within a myxoid stroma (Figure 2 G–H). Foci of cells with pleomorphic nuclei and mitotic figures were identified (Figure 2 K–L). The myoepithelial cellular component of the neoplasm stained for S100 protein and was negative for cytokeratin and carcinoembryonic antigen (CEA) expression. The positive cytokeratin stain highlighted the background epithelial/glandular components of the neoplasm. Stains for epithelial membrane antigen (EMA) and HMB-45 were negative.

Conclusions

We describe a cutaneous neoplasm composed of myoepithelial cells and a focal epithelial and glandular component. The S100 positivity indicates myoepithelial cells, while epithelial-glandular cells are negative for S100 protein expression. Myoepithelial cells did not stain with anti-cytokeratin bodies. Based on findings by conventional microscopy and immunohistochemistry, this neoplasm shows myoepithelial differentiation and focal epithelial lined tubules with features of apocrine secretion, findings that are consistent with the diagnosis of primary adenomyoepithelioma of the skin.

While various adnexal neoplasms with a myoepithelial cellular component have been described in the skin, adenomyoepithelioma of the skin seems to be extremely rare. What are the criteria that distinguish adenomyoepithelioma from

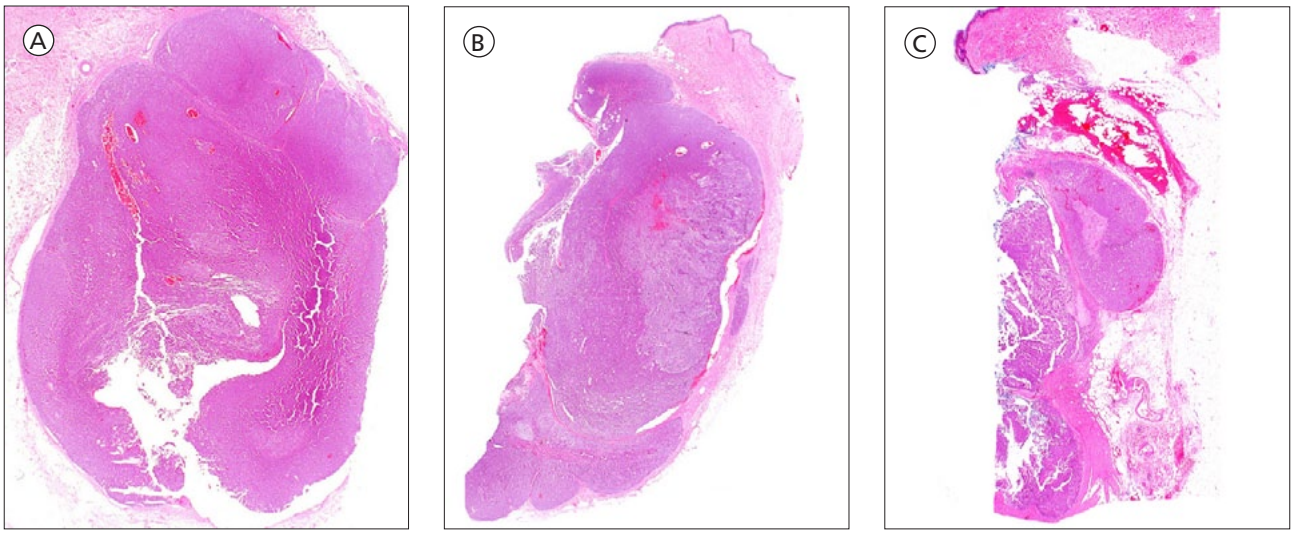

Figure 1. Histologic features of adenomyoepithelioma. The large lobulated neoplasm is located in the dermis and surrounded by a fibrous pseudocapsule (A). Beneath an intact epidermis a superficial zone of fibrosis (B) is present. The neoplasm is composed of tubular and ductal structures as well as solid-appearing tumor areas set in a fibromyxoid stroma (C). [Copyright: ©2012 Riedl et al.]

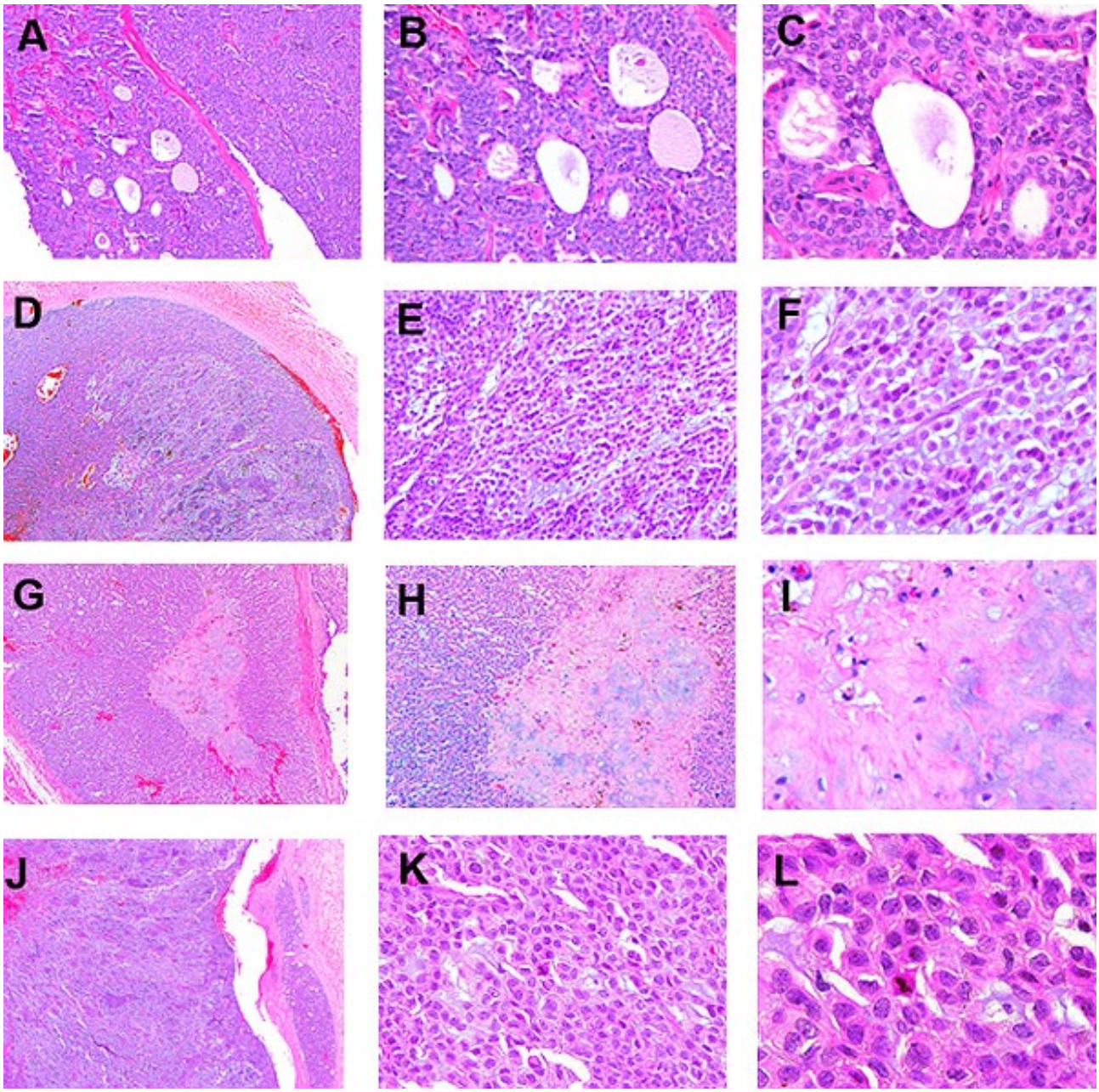

Figure 2. Cutaneous adenomyoepithelioma is made up of different cellular components. In foci glandular and ductal structures are present; occasionally apocrine-type secretion within glandular structures is found (A–C). In other areas myoepithelial cells with clear cytoplasm predominate (D–F). In various areas cells are present within a myxoid stroma (G–H). Within solid cell aggregation pleomorphic cells and mitotic figures are evident (K–L). [Copyright: ©2012 Riedl et al.]

other benign cutaneous neoplasms with myoepithelial differentiation? While myoepithelioma is defined as a benign neoplasm consisting exclusively of myoepithelial cells embedded in a myxoid stroma, adenomyoepithelioma shows in addition to myoepithelial cells a second component displaying various degrees of epithelial-ductal differentiation [6,8]. Mixed apocrine tumor, also termed "chondroid syringoma," at the other end of the spectrum is a benign adnexal neoplasm that, in addition to apocrine epithelium, manifests various degrees of follicular and/or sebaceous differentiation [9]. Clinically, myoepitheliomas occur usually in children and young adults and are located on the extremities, while mixed apocrine tumors affect older individuals and are usually found on the face [8,9]. The few cases of cutaneous adenomyoepitheliomas that have been described to date, including the present case, were described in older patients and were located on the extremities and on the trunk [6,7].

Pleomorphic adenoma in the breast is regarded as the analogue to mixed apocrine tumor in the skin, while adenomyoepithelioma of the breast is defined as a neoplasm with nodular aggregations of clear myoepithelial cells that surround epithelial lined tubules. Occasionally, the myoepithelial component predominates and loses the close association with epithelial structures [1].

In the skin, the predominance of myoepithelial cells in relation to ductal epithelial structures lacking features of follicular and/or sebaceous differentiation separates this neoplasm from mixed apocrine tumor. Therefore, myoepithelioma, adenomyoepithelioma and mixed apocrine tumor lie in the spectrum of neoplasms with pure myoepithelial differentiation at one end and apocrine-sebaceous-follicular differentiation at the other.

The most important question for the patient is the biologic potential of such a lesion. While the chronic course in our patient and the majority of the histopathologic features suggest a benign neoplasm, foci of cells having pleomorphic nuclei and mitotic figures were present. Both findings could indicate the potential for locally aggressive behavior and/or metastasis, as has been rarely described in adenomyoepitheliomas of the breast [2,10]. Therefore, adenomyoepitheliomas of the skin should be completely excised, as has been recommended in the reported case.

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