

A Case of Superficial Epithelioma with Sebaceous Differentiation

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Superficial epithelioma with sebaceous differentiation (SESD) is a rare benign neoplasm with peculiar histopathologic characteristics, which occurs in aged skin. We report upon a case of SESD occurring on the left upper back, which histopathologically is a superficial, multilobular tumor with numerous basaloid cells mixed with sebaceous cells attached to the overlying epidermis. This case represents a solitary benign neoplasm without any associated malignancy.

Key Words: Superficial epithelioma with sebaceous differentiation

INTRODUCTION

Superficial epithelioma with sebaceous differentiation (SESD) is a rare histologically distinct benign neoplasm. This tumor occurs as multiple or solitary lesions on the face, neck and trunk in elderly persons.^{1,2} To date, only 9 cases have been reported in literature.¹⁻⁵ We report upon a case of SESD occurring in the upper back of a 59-year-old woman.

CASE REPORT

A 59-year-old woman presented with a pruritic erythematous lesion on her left upper back that had been present for 2 years. There was no family

history of similar skin lesions. Physical examination showed a 15 × 20 mm sized yellowish to slightly brownish, erythematous, flat-topped, well-demarcated plaque, and surrounding erythematous macules with telangiectasiae and petechiae on the upper back (Fig. 1). Past history and routine laboratory studies were unremarkable. Histopathological examination of the lesion revealed a superficial, multilobular, proliferation of tumor cells with numerous broad attachments to the overlying epidermis (Fig. 2). The overall configuration was of a plate-like proliferation of basaloid cells with pale basophilic nuclei, eosinophilic cytoplasm, and poorly defined borders without apparent intercellular bridges. No mitoses were observed. At the epidermal-tumor junction, tumor cells gradually merged with the overlying epidermal cells. Sebaceous cells, scattered as small clusters, were observed on the lower portion of these lobules. Within the tumor mass, there were a few keratin-filled cystic structures lined with squamous epithelium of epidermal or sebaceous duct (Fig. 3). The basaloid cells did not show peripheral palisading or retracted spaces between the lobules and dermal collagens. The basement membrane surrounded the tumor lobules was positive for periodic acid-Schiff stain although the tumor cells were unstained. Only some of the sebaceous cells stained with anti-epithelial membrane antigen (EMA) antibody, while all of the sebaceous cells in the normal sebaceous glands showed strong positive EMA staining (Fig. 4). Neither basaloid cells nor sebaceous cells were stained by anti-carcinoembryonic antigen (CEA) antibody. Two weeks after excision of the tumor, her pruritus and surrounding erythematous

Received August 12, 2002

Accepted October 24, 2002

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macules faded with topical steroid application.

DISCUSSION

In 1980, Rothko, et al.¹ first reported six histologically distinctive cutaneous tumors, which had developed in a 48-year-old man, and involved his face, right axilla, trunk, and right thigh over an eight-year period. Distinct tumor features included superficiality, numerous broad attach-

ments to the overlying epidermis, a plate-like configuration with well-defined borders, and sebaceous differentiation. Rothko and coworkers described the disease entity as "superficial epithelioma with sebaceous differentiation". Friedman, et al.² in 1987 reported 5 cases with identical histologic features. Several groups reported upon additional 3 cases later.³⁻⁵ The clinical features of the 10 published cases including the present case, are summarized in Table 1. Age of onset has ranged from 38 to 72, and mean age is 60. Six of 10 patients were female. The most common tumor location is the face, although it has also been found on the back, neck, axilla or thigh. Clinical appearance is variable, showing small papules, nodules, and plaques. The present case appeared

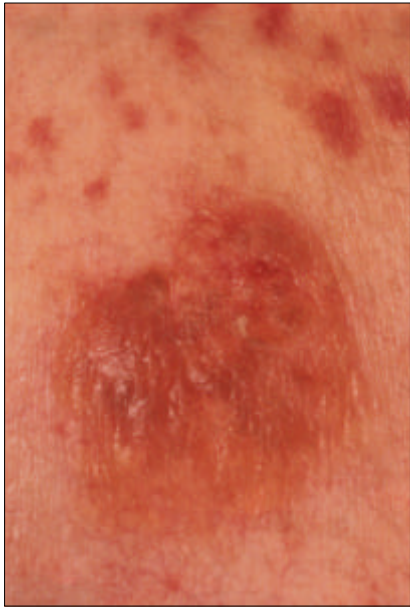


Fig. 1. A 15 × 20 mm, well-demarcated, yellowish to slightly brownish hard plaque with surrounding erythematous macules with telangiectasiae and petechiae on the upper back.

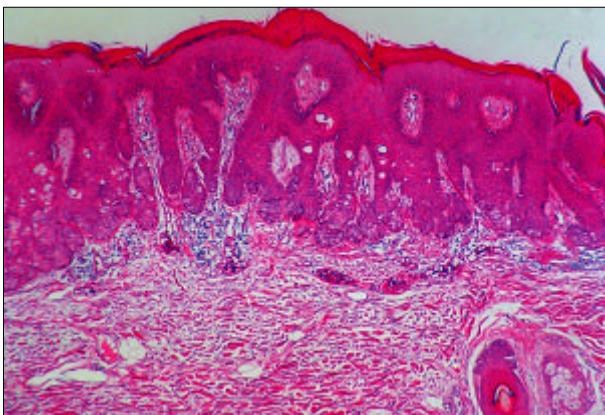


Fig. 2. A superficial plate-like tumor with numerous broad attachments to the overlying epidermis. Hematoxylin and eosin stain. Original magnification × 40.

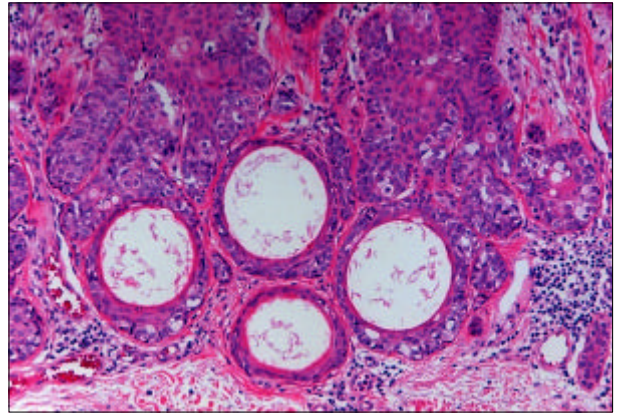


Fig. 3. Sebaceous cells and basaloid cells forming lobules. Multiple keratin-filled cystic structures are shown. Hematoxylin and eosin stain. Original magnification × 100.

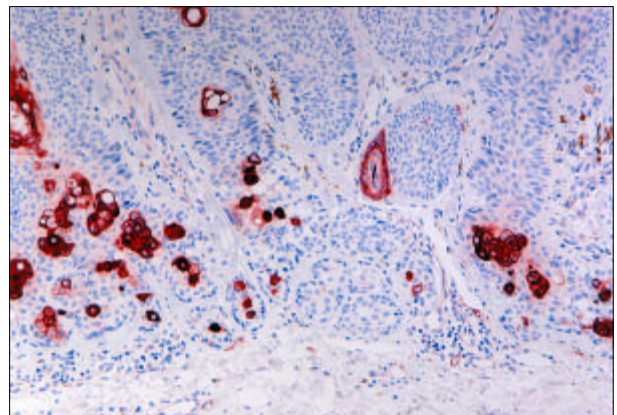


Fig. 4. Immunohistochemical staining with anti-epithelial membrane antibody showed positive reactivity for some sebaceous cells. Original magnification × 100.

Table 1. Summary of Superficial Epithelioma with Sebaceous Differentiation

Case No.	Authors	Age/Sex	Location	Lesion Size	Comments
1	Rothko, et al. ¹	48/M	Face, axilla, trunk, thigh	3 mm	
2	Friedman, et al. ²	66/F	Neck	10 mm	
3	Friedman, et al. ²	72/M	Cheek	7 mm	Internal malignancy
4	Friedman, et al. ²	72/F	Forehead	10 mm	
5	Friedman, et al. ²	57/M	Eyelid	8 mm	
6	Friedman, et al. ²	65/F	Cheek	4 mm	
7	Vaughan, et al. ³	55/F	Back	14 × 8 mm	Halo & telangiectasia
8	Kato, et al. ⁴	38/F	Eyelid	6 × 3 mm	
9	Akasaka, et al. ⁵	68/M	Cheek	4 mm	
10	Present case	59/F	Back	15 × 20 mm	Pruritus & telangiectasia

as a yellowish to erythematous plaque, which was similar to the lesions described by Vaughan, et al.³ and Kato, et al.⁴ The tumor size has ranged from 3 to 20 mm; the tumor in the present case measured 15 × 20 mm, and is the largest recorded to date.

Histopathologically, SESD is a superficial, well demarcated, plate-like proliferation of tumor cells with sebaceous differentiation, and should be distinguished from several epithelial tumors showing differentiation toward sebaceous cells, including nevus sebaceus, sebaceous hyperplasia, sebaceous adenoma, sebaceous epithelioma, sebaceous carcinoma, and basal cell epithelioma with sebaceous differentiation. Sebaceous hyperplasia is defined by the presence of four or more sebaceous lobules attached to the infundibulum of each pilosebaceous unit. Cells comprising the lobules are predominantly fully mature sebaceous cells without basaloid cell proliferation,⁶ which is not compatible with the findings of SESD. Sebaceous adenoma is a sharply circumscribed and multilobulated tumor composed of small generative cells at the periphery and mature sebaceous cells at the lobule centers. It is usually located in the mid or deep dermis and is surrounded by compressed collagen.⁷ Sebaceous epithelioma is a designation for all lesions having aggregations of basaloid, undifferentiated cells admixed with single or small clusters of mature sebaceous cells, or duct- or cyst-like formations.^{8,9} The plate-like architec-

tural pattern, superficial location and broad attachments to the epidermis, distinguish SESD from sebaceous adenoma and sebaceous epithelioma. Sebaceous carcinoma is easily differentiated from SESD, because it shows deeply invasive and poorly demarcated tumor cells with mitoses and cytological atypia.¹⁰ Immunohistochemically, most epithelial tumors react with antibodies against EMA. EMA stains both normal sweat and sebaceous glands, whereas CEA stains normal sweat glands not normal sebaceous glands.¹¹ The positive staining of tumor cells for EMA and negative staining for CEA in SESD is helpful to differentiate SESD from epitheliomas with eccrine or apocrine differentiation. SESD can also be differentiated from basal cell epithelioma with sebaceous differentiation, by referring to the missing characteristic features of basal cell epithelioma with sebaceous differentiation, such as peripheral palisading of basaloid cells and the retraction of collagen tissues around the tumor nests.¹⁰ The plate-like configuration of SESD is similar to that seen in tumors of the follicular infundibulum¹² and in basaloid follicular hamartoma,¹³ but sebaceous differentiation and keratin-filled cysts are not features of these lesions. Recently, Mahalingam M, et al.¹⁴ described a case, which appeared to be a hybrid lesion and which was interpreted as either a tumor of the follicular infundibulum with sebaceous differentiation or as a reticulate variant of SESD. They suggested a

new disease entity, "tumor of the follicular infundibulum with sebaceous differentiation". This is differentiated from SESD, as SESD is characterized by broad plate-like outgrowths from the epidermis, which contrast with the thin reticulate anastomosing strands observed in tumors of the follicular infundibulum with sebaceous differentiation. In addition, keratin-filled cysts, a characteristic feature of SESD are not present in tumors of the follicular infundibulum with sebaceous differentiation.

SESD seems to have a benign biological characteristic with limited growth, because none of the lesions reported in literature have recurred or spread after electrodesiccation, curettage, or simple excision. The patient of the present case was treated by simple excision, and is well without recurrence 6 months after the treatment, and the surrounding erythematous macules with telangiectasiae and petechiae seem to be due to non-specific irritation, as these faded away after excision of the tumor. She also did not have any associated disease or anomalies. Only one patient among the previously reported cases had esophageal and colon carcinoma and died from the metastasis. However, it is not clear whether the carcinoma was associated with SESD.² Other reported cases did not have any accompanying malignancies. Therefore, a possible association with the Muir-Torre syndrome is as yet unproven. SESD is a histologically distinctive benign neoplasm, which usually occurs as a solitary lesion on the face in the elderly.

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