Case Report

Locally Invasive Dermal Squamomelanocytic Tumor: An Uncommon Case of a Biphenotypic Neoplasm that Usually Occurs on Sun-Damaged Skin of Elderly Individuals

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We report a locally invasive dermal squamomelanocytic tumor arising on the right jawline of an 85 year old woman with extensive sun damage treated with complete excision. Histological examination revealed a well circumscribed but unencapsulated dermal tumor comprised of epithelioid squamous cells and heavily pigmented melanocytes. AE1/AE3 cytokeratin stain, HMB-45, and Melan-A stain highlight the different component populations. The population of epithelioid squamous cells comprised the predominant population as encountered in other reported cases. The diagnostic features, reported cases, and management of this uncommon tumor were reviewed.
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INTRODUCTION

The incidence of melanoma continues to increase and squamous cell carcinoma is also increasing at epidemic proportions.[1,2] With the increase in tumors, unusual variants are also becoming more prevalent. The cutaneous squamomelanocytic tumor is an unusual tumor that has both epithelial and melanocytic components.[3] This tumor usually occurs on markedly sun damaged skin of elderly individuals.[4] Rather than a "collision" tumor in which two contiguous lesions coexist,[5] the biphasic nature of the squamomelanocytic tumor suggests that it is best considered combined and showing dual differentiation.[3,6] The intermingling of both squamous and melanocytic components represents more than just the simple colonization of a squamous process, but is evidence of divergent differentiation in a single tumor.[6]

CASE REPORT

An 84 year old woman was noted to have a rapidly growing, indurated, hard black nodule along the right angle of the jaw. The clinical impression was that of a malignant melanoma. The area was excised and the excisional specimen revealed a dome-shaped tumor which was well-circumscribed and involved the superficial and deep dermis (**Figure 1**). The tumor was composed of atypical epithelioid cells with enlarged nuclei and prominent nucleoli (**Figure 2**). Numerous dermal mitoses were evident, and atypical mitoses were encountered (**Figure 2**). S-100, Melan-A, and HMB-45 stains decorated atypical neoplastic cells and an AE1/AE3 cytokeratin stain decorated other atypical neoplastic cells that comprised the tumor (**Figure 3-5**). Our patient has done well with simple excision, with no evidence of local recurrence or metastasis at a recent 2 year follow up appointment.

DISCUSSION

Non-melanoma skin cancer is common, yet tumors that exhibit combined features are very uncommon. Understanding the pathogenesis of such tumors may help clarify early steps in carcinogenesis. Tumors arising from the follicular-sebaceous-apocrine unit often exhibit a varying composition with different amounts of the constituent components.[7,8] Tumors with both an epithelial and melanocytic component have been recognized for a long time. Melanoacanthomas are a type of Seborrheic keratosis colonized by melanocytes. Matricomas are a follicular tumor that can be associated with a significant population of melanocytes.[9-15] Both carcinosarcomas and squamomelanocytic tumors exhibit different components that are intimately associated with one another. In carcinosarcomas, neoplastic elements are closely opposed to one another[16] whereas in a squamomelanocytic tumor, they are intermixed with one another within discrete tumor lobules. Carcinosarcomas with epithelial and mesenchymal elements

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are considered biologically aggressive,[16] although carcinosarcomas of the skin have a better prognosis than carcinosarcomas noted in other areas. Carcinosarcomas are often found in the head area. Similarly, dermal squamomelanocytic tumors are often found on exposed areas of the head, but may involve the arms and other sites.[17,23]

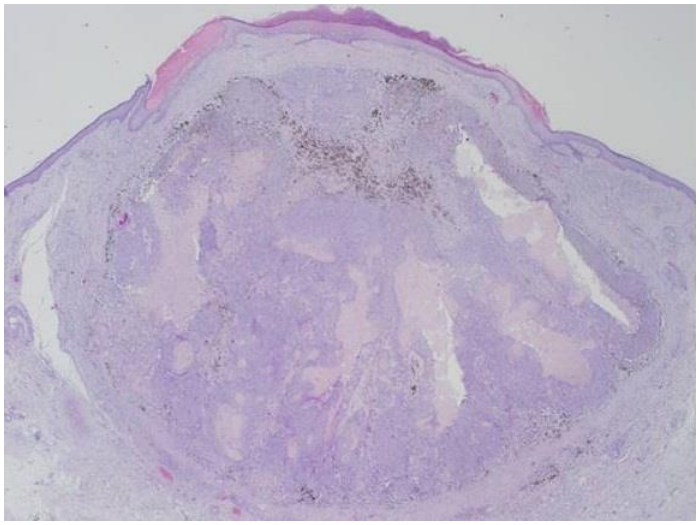

Figure 1. Low power examination at scanning magnification reveals a well circumscribed dermal nodule with areas of pigmentation and other areas that are not pigmented. Cystic areas are noted and the configuration is reminiscent of a matricoma. The dermal nodule does not show a connection with the overlying epidermis. Junctional nests and intraepidermal proliferation of melanocytes is not encountered. (Hematoxylin and eosin stained sections. Original magnification 20x).

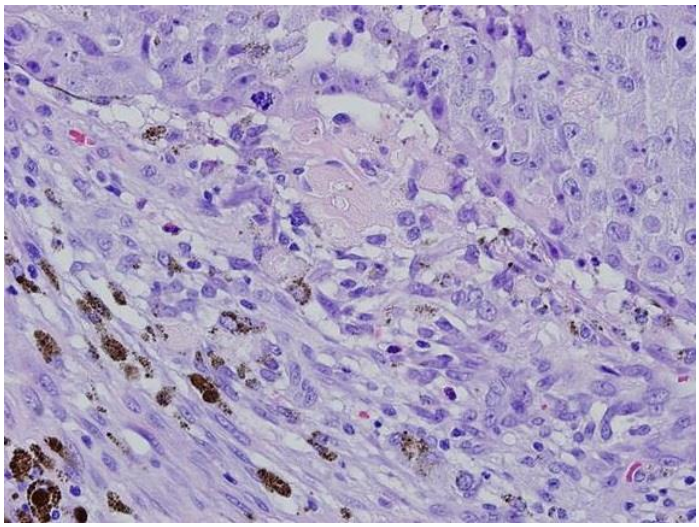

Figure 2. Some neoplastic cells are devoid of pigment whereas other atypical cells are replete with abundant melanin. Atypical mitotic figures are noted. Pigmented and non-pigmented cells are closely intermingled, making identification of lineage difficult without the use of special stains. (Hematoxylin and eosin stained sections. Original magnification 400x).

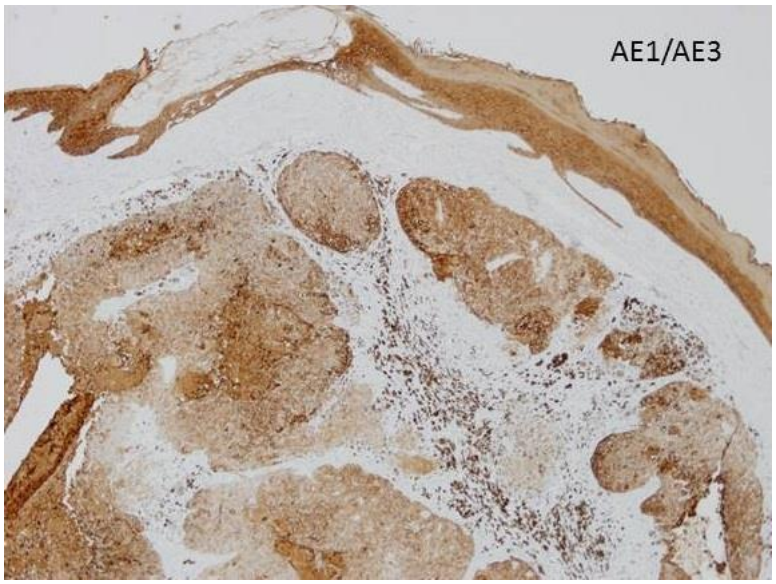

Figure 3. An AE1/AE3 cytokeratin stain decorates atypical keratinocytes within the dermal nodule. (Original magnification 100x).

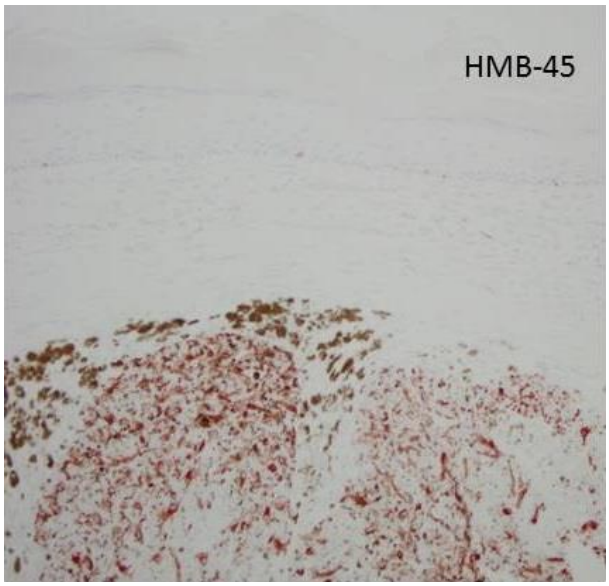

Figure 4. An HMB-45 stain decorates atypical melanocytes that are intimately associated with atypical keratinocytes (Original magnification 200x)

Squamomelanocytic tumors often occur in the elderly. Divergent differentiation of a stem cell is suspected to be causative. Rosen et al have demonstrated premelanosomes and keratin filaments in a single cell as well as coexpression of S-100 and keratin.[3] These features suggest that a pluripotential stem cell is essential for the formation of a dermal squamomelanocytic tumor. The squamomelanocytic tumor noted in our patient had two clearly malignant

populations. One population expressed AE1/AE3 cytokeratin, keratin formation on routine histological evaluation, atypical mitoses, and pleomorphic nuclei. The population of pigment laden cells that were intimately associated with the initial population also exhibited atypical mitoses and marked nuclear pleomorphism. Stem cell cancerization or some other interaction between neoplastic keratinocytes and melanocytes may explain this unusual phenomenon.[24] Miteva et al favor that squamomelanocytic neoplasms are a true malignant proliferation of two distinct cell phenotypes that exhibit "close paracrine interactions".[6] Stem cells are dynamic and plastic, and likely respond to stromal changes in their milieu.[24] These effects may be critical for the development of squamomelanocytic tumors, but the precise pathogenesis still needs to be elucidated. A recent molecular study revealed 11q13 amplifications in both the melanocytic and squamous components of a tumor, supporting the hypothesis that the tumor arose from a common progenitor cell.[25] We recommend performing a complete excision with margins that would be appropriate for a melanoma with a comparable depth of invasion, recognizing that additional studies need to be performed to identify the optimal treatment strategy. Although metastasis may occur, it is uncommon.[25]

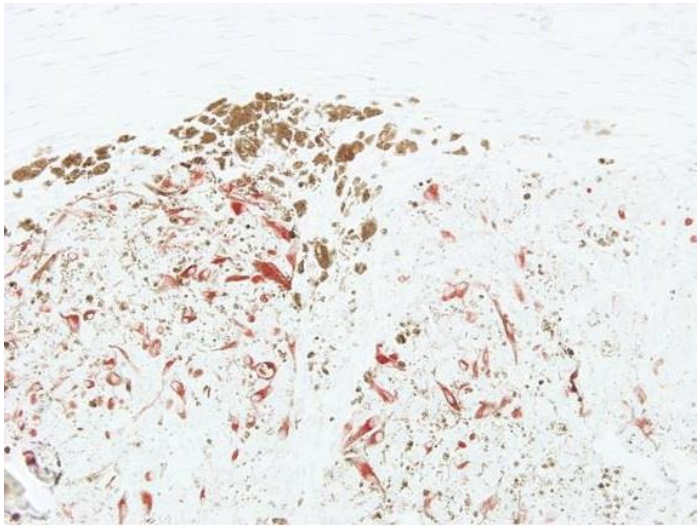

Figure 5. A Melan-A stain decorates atypical melanocytes that are intimately associated with the atypical keratinocytes. The melanophages fail to stain with the red chromogen and are identified by the conspicuous lack of staining. (Original magnification 400x)

CONCLUSION

Clinicians should be aware of the dermal squamomelanocytic tumor. This rare biphenotypic neoplasm arises on markedly sun damaged skin, shows combined features of melanocytic proliferation and a squamous proliferation. Both of the phases in our case showed evidence of malignancy. Complete excision is required. The prognosis and biologic potential is difficult to determine at this point, but further study should help clarify important steps in carcinogenesis.

CONFLICT OF INTEREST

The authors have no conflict of interest to disclose.

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