Case Letter

Pedunculated Scrotal Nodule in an Elderly Male: A Rare Presentation of Trichoblastoma

To the Editor:

A 78-year-old man with no relevant dermatologic history presented for a routine skin examination. He had a large pedunculated nodule on the right scrotum that had been stable in size for more than 40 years. He reported the lesion had been diagnosed as a skin tag by his urologist 20 years prior and had remained asymptomatic in nature. On examination, a 2.5×1-cm, pedunculated, firm, rubbery nodule protruded from the anterior aspect of the right scrotal skin (Figure 1). Prominent telangiectases were noted on the surface of the lesion along with an irregular pigmented macule on the tip. There was no inguinal lymphadenopathy. Given the pigmentation at the tip of the lesion, he was advised to have it completely excised to rule out melanocytic atypia. The lesion subsequently was excised under local anesthesia and the resulting defect was primarily closed. A group of basaloid lobules within the dermis with scant stroma was noted (Figure 2). There was no epidermal connection and retraction artifact was absent. The epidermis appeared normal, except for basilar pigmentation corresponding to the pigmented macule at the tip of the lesion. Higher magnification of one of the basaloid islands demonstrated a papillary mesenchymal body (Figure 3). Immunohistochemistry with cytokeratin 20 showed positive staining of Merkel cells colonizing the basaloid islands (Figure 4). A diagnosis of trichoblastoma was rendered. Two nodular basal cell carcinomas (BCCs) were seen on initial presentation, but he remained free of recurrence of the scrotal tumor at 2-year follow-up.

Trichoblastoma is a rare benign tumor with differentiation toward the primitive hair follicle. The lesion typically presents as a slow-growing, solitary, well-circumscribed nodule that typically measures up to 3 cm and is predominantly located on the head and neck with a predilection for the scalp. Rarely the lesion will appear on the trunk, proximal extremities, or perianal and genital regions.[1] Trichoblastomas show no gender predilection and predominantly affect adults between the fifth and seventh decades of life. In children it is the most common neoplasm arising in preexisting nevus sebaceous (Jadassohn nevus).[2] Several clinical variants of trichoblastoma have been described, including giant trichoblastoma, pigmented trichoblastoma, and subcutaneous trichoblastoma, among others.[3]

Histologically, these tumors are characterized by well-circumscribed basaloid islands in the dermis with peripheral palisading closely resembling BCCs and trichoepitheliomas. Unlike BCCs, retraction artifact in trichoblastoma is a rare finding, while papillary mesenchymal bodies and an increased number of

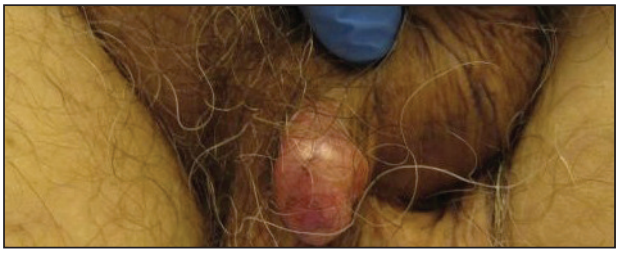

Figure 1. Firm, pedunculated, flesh-colored nodule on the scrotum.

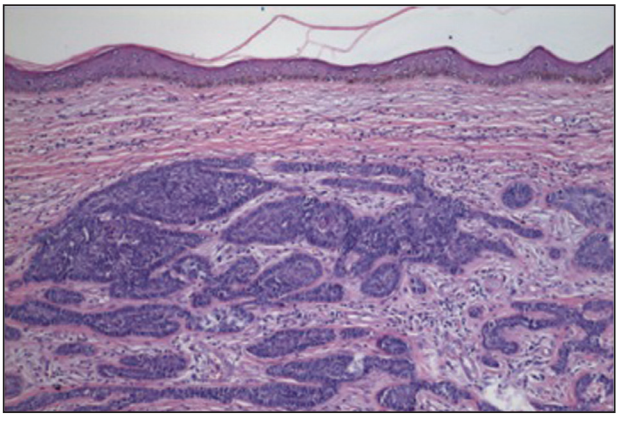

Figure 2. Basaloid tumor islands within the dermis with no epidermal connection (H&E, original magnification ×20).

Dr. Navi is from the Department of Dermatology, Stanford University, Redwood City, California. Drs. Egbert and Nord are from Palo Alto Veterans Affairs Healthcare System, California.

The authors report no conflict of interest.

Correspondence: Daniel Navi, MD, 450 Broadway St, Pavilion C, Redwood City, CA 94063 (dnavi@stanford.edu).

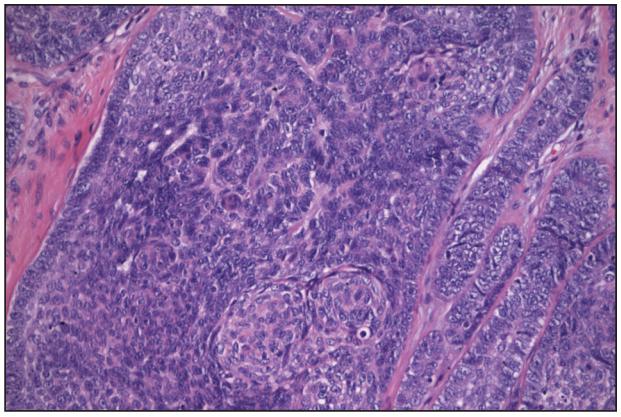

Figure 3. Papillary mesenchymal body adjacent to several dermal basaloid islands (H&E, original magnification ×40).

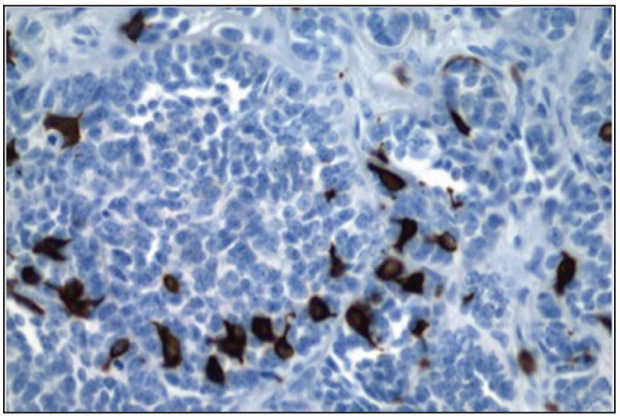

Figure 4. Cytokeratin 20 immunohistochemical staining indicated Merkel cells within the basaloid islands (original magnification ×40).

Merkel cells characterize both trichoblastomas and trichoepitheliomas, as in our patient.[4] Other histologic features include lack of epidermal connection and location in the deeper dermis and/or subcutis. There also are reported cases with sebaceous, pigmented, rippled patterns and clear cell differentiation on histology.[3,5]

The genetic basis for trichoblastoma remains unclear. Although these primitive hair follicle tumors share histologic features with BCC and trichoepithelioma, which have both demonstrated *PTCH* (patched) mutations, no classic mutations in the *PTCH* gene were seen in 11 cases of trichoblastoma.[6] In contrast, there have been reports of trichoblastoma occurring in Brooke-Spiegler syndrome and a case of a somatic *CYLD* (cylindromatosis) frameshift mutation in a trichoblastoma in a patient with Brooke-Spiegler syndrome[7]; however, the significance of *CYLD* mutation in the pathogenesis of trichoblastoma has not been fully elucidated.[8]

Trichoblastomas generally behave in an indolent asymptomatic manner, with malignant transformation being a rare occurrence.[9] However, complete excision of these tumors is recommended given the potential for continued slow growth if incompletely removed. In the case of our patient, the lesion was completely excised at the outset. We plan to clinically monitor for evidence of disease recurrence as part of an annual full-body examination and have noted no evidence of recurrence in more than 2 years.

Daniel Navi, MD
Barbara Egbert, MD
Kristin M. Nord, MD

REFERENCES

1. Brenn T, McKee PH. Tumors of the hair follicle. In: McKee PH, Calonje E, Granter SR, eds. *Pathology of the Skin With Clinical Correlations*. 3rd ed. London, England: Elsevier Mosby; 2005:1552-1557.
2. Jaqueti G, Requena L, Sánchez Yus E. Trichoblastoma is the most common neoplasm developed in nevus sebaceous of Jadassohn: a clinicopathologic study of series of 155 cases. *Am J Dermatopathol*. 2000;22:108-118.
3. Kim DW, Lee JH, Kim I. Giant melanotrichoblastoma. *Am J Dermatopathol*. 2011;33:e37-e40.
4. Schulz T, Hartschuh W. Merkel cells are absent in basal cell carcinomas but frequently found in trichoblastomas. an immunohistochemical study. *J Cutan Pathol*. 1997;24:14-24.
5. Swick BL, Baum CL, Walling HW. Rippled-pattern trichoblastoma with apocrine differentiation arising in a nevus sebaceus: report of a case and review of the literature. *J Cutan Pathol*. 2009;36:1200-1205.
6. Hafner C, Schmiemann V, Ruetten A, et al. *PTCH* mutations are not mainly involved in the pathogenesis of sporadic trichoblastomas [published online ahead of print June 26, 2007]. *Hum Pathol*. 2007;38:1496-1500.
7. Kazakov DV, Soukup R, Mukensnabl P, et al. Brooke-Spiegler syndrome: report of a case with combined lesions containing cylindromatous, spiradenomatous, trichoblastomatous, and sebaceous differentiation. *Am J Dermatopathol*. 2005;27:27-33.
8. Kazakov DV, Schaller J, Vanecek T, et al. Brooke-Spiegler syndrome: report of a case with a novel mutation in the *CYLD* gene and different types of somatic mutations in benign and malignant tumors [published online ahead of print February 4, 2010]. *J Cutan Pathol*. 2010;37:886-890.
9. Schulz T, Proske S, Hartschuh W, et al. High-grade trichoblastic carcinoma arising in trichoblastoma: a rare adnexal neoplasm often showing metastatic spread. *Am J Dermatopathol*. 2005;27:9-16.