CASE REPORT / OLGU SUNUMU

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An Unusual Presentation of Proliferating Trichilemmal Tumor Developing in Psoriatic Plaque: A Case Report

Psöriyatik Plakta Gelişen Prolifere Trikilemmal Tümör: Nadir Görülen Bir Olgu Sunumu

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ABSTRACT

Proliferating trichilemmal tumor (PTT) is a rare, mostly benign neoplasm which stems from the follicular outer root sheath epithelium. PTT occurs as a subcutaneous cystic nodule slowly enlarging to a larger noduler mass, and is usually localized on the scalp of elderly women. To the best of our knowledge, PTT localized on a psoriatic plaque has not been reported previously. Herein, we report an unusual case of benign PTT arising from a psoriatic plaque on the knee of a 63-year-old male patient.

Keywords: proliferating trichilemmal tumor, psoriasis, trichilemmal

Prolifere trikilemmal tümör (PTT) foliküler dış kök kılıfı epitelinden köken alan nadir, çoğunlukla benign bir neoplazmdır. PTT yavaşça büyük nodüler bir kitle olarak genişleyen subkutan kistik nodül şeklinde oluşur ve genellikle yaşlı kadınların saçlı derisinde lokalizedir. Literatürde daha önce kronik psoriazis plağı üzerinde gelişen PTT bildirilmemiştir. Bu olgu sunumunda 63 yaşında erkek hastanın diz bölgesindeki kronik psoriazis plağı üzerinde gelişen benign PTT bildirilmiştir.

ÖZ

Anahtar Sözcükler: prolifere trikilemmal tümör, psoriazis, trikilemmal kist

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Introduction

Proliferating trichilemmal tumor (PTT) is a rare, and mostly benign neoplasm differentiating towards the follicular outer root sheath epithelium. PTT is usually found on the scalp of elderly women as a long standing, and subcutaneous cystic nodule slowly enlarging to a larger noduler mass. Etiopathogenesis is unclear but it is believed that PTT originates from a pre-existing trichilemmal cyst (TC) as a complication of trauma and inflammation (1–3). Both PTT and TC show trichilemmal keratinization as a histological marker. But additionally, PTT shows extensive epithelial proliferation, variable cytologic atypia, and mitotic activity. Recent reports have described a small number of malignant proliferating trichilemmal tumors (4, 5).

To the best of our knowledge, no PTT that occurs on a psoriatic plaque has been reported previously. Herein, we report an unusual case of benign PTT arising from a psoriatic plaque on the knee of a 63-year-old male patient.

Case report

A 63-year-old-male presented with a 9-months history of a pink, dome shaped tumor on a chronic psoriatic plaque on his knee, which was gradually enlarging with episodes of bleeding in the last 2 months. He denied trauma or insect bite at this location. According to his medical history, he has had psoriasis since 25 years. He has used topical (corticosteroid, calsipotriol)

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Figure 1. Localized, exophtic, non-painful, soft consistency and ulcerated nodular lesion



Figure 2. 2x2 cm in diameter exophtic, nodular lesion developing in a chronic psoriatic plaque

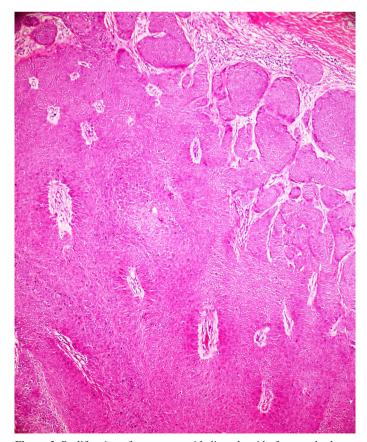


Figure 3. Proliferation of squamous epithelium devoid of a granular layer, trichilemmal keratinization at the center of the lobules, peripheral palisading of small basaloid cells X40, H&E

and systemic (cyclosporin, methotrexate, rePUVA) treatments at different times. But for the last 4 years, he left all of the treatments and follow-up. Dermatological examination showed a localized, exophytic, non-painful, soft consistency and ulcerated nodular lesion, 2×2 cm in diameter on a chronic psoriatic plaque (Figure 1, 2). The patient had no extracutaneous anomalies, and laboratory examinations revealed no significant abnormalities. The lesion was clinically diagnosed as squamous cell carcinoma and a total surgical tumor excision with a narrow margin of normal tissue was performed. Histological examination revealed the proliferation of squamous epithelium devoid of a granular layer, trichilemmal keratinization at the center of the lobules, and peripheral palisading of small basaloid cells (Figure 3). Based on the histological features, the tumor was diagnosed as PTT.

Discussion

PTT is a skin neoplasm that probably arises from a pre-existing trichilemmal cyst subsequent to trauma or inflammation; it can also occur *de novo*. Usually, the clinical presentation of PTT is a pink to reddish, long-standing, subcutaneous, cystic nodule which is described clinically as a slowly growing large solitary nodular lesion, and 90% of cases are located in the scalp. It may ulcerate, bleed, and produce a purulent discharge. Women are the most affected, and the average age of patients is 65 years (1–4). In our case, the lesion had developed in a chronic psoriatic

plaque localized on the knee of a man, and has grown specifically in the last year. The histologic features of PTT are trichilemmal keratinization areas in the center of the lobules, and formation of compact eosinophilic keratin without the granular layer. In addition, dyskeratotic cells, basaloid cells, calcification, varying degrees of mitoses and cytological atypia have been described. Occasionally, malignant transformation of PTT occurs, and the diagnosis is essentially based on histopathologic features (2–4). Histologically, malignant PTTs show severe nuclear atypia, cellular pleomorphism with atypical mitosis, dyskeratotic cells, and an infiltrating margin. Distant or lymph node metastasis have been described in malignant forms of PTT (2, 3). The treatment choice is surgical resection of the tumor with a safety margin of 1 cm, and accurate follow up. Our case highlights the importance of recognizing this unusual tumor at an uncommon location.

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