Case Report
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Multiple pilar sheath acanthomas on the scrotal region

Skrotal bölgede çok sayıda pilar kılıf akantomu

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Abstract

Pilar sheath acanthoma is an uncommon, benign follicular hamartoma that frequently presents as an asymptomatic, skin-colored papule or nodule with central opening. Pilar sheath acanthoma generally appears on the upper lip of elderly patients, although a few cases have been described on other locations such as lower lip and cheeks. In this article, we present a 62-year-old male who presented with multiple pilar sheath acanthoma on the scrotal and anal regions.
Keywords: Benign follicular hamartoma, pilar sheath acanthoma, pilar neoplasm

Öz

Pilar kılıf akantomu sıklıkla asemptomatik, merkezi açıklıkla birlikte seyreden deri renginde papül ya da nodül şeklinde görülen nadir, benign foliküler hamartomdur. Pilar kılıf akantomu genellikle yaşlı hastalarda üst dudakta görülmesine rağmen çok az olguda alt dudak ve yanak gibi diğer yerlerde de tarif edilmiştir. Bu yayında 62 yaşında skrotal ve anal bölge üzerinde çok sayıda pilar kılıf akantomu bulunan bir erkek hasta sunulmaktadır.
Anahtar Kelimeler: Benign foliküler hamartom, pilar kılıf akantomu, pilar neoplazma

Introduction

Pilar sheath acanthoma is an uncommon, benign follicular hamartoma that was first described by Mehregan and Brownstein[1] in 1978. Although a few cases have been described on other locations such as lower lip and cheeks, these lesions generally appear on the upper lip in elderly patients[2]. They frequently present as an asymptomatic, clinically a comedo-like small skin-colored papule or nodule with a central opening[2].

We present a 62-year-old male with multiple pilar sheath acanthoma on the scrotal and anal regions. To the best of our knowledge, this is the first reported case of a pilar sheath acanthoma presenting on the scrotum and anal region.

Case Report

A 62-year-old male presented with the complaints of multiple, asymptomatic nodules on the scrotal and anal regions for 10 years. Dermatological examination revealed multiple skin-colored nodules with central openings, ranging from 1 mm to 1 cm in diameter (Figure 1). Provisional diagnoses included lymphangioma circumscriptum and trichofolliculoma. Excisional biopsy taken from the scrotal lesion revealed dilated hair follicle with acanthotic epithelium in the center of the area and a lobular keratinocyte mass around the hair follicle radially extending into the dermis. There were no hair formations. The cells were round to polyhedral with peripheral palisading (Figure 2).

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Discussion

Pilar sheath acanthoma is a rare, benign follicular hamartoma. Clinically it is characterized by a small, solitary, skin-colored papule or nodule, 5-10 mm in diameter with a central pore-like opening plugged with keratin. Pilar sheath acanthoma typically affects middle-aged and elderly patients[3]. Usually, these lesions are localized on the head and neck, particularly around the upper lip[2]. In the literature, pilar sheath acanthoma localized on the nasolabial fold, cheek, forehead, postauricular area and earlobe has been documented as case reports[2-7].

In our case, a 62-year-old male presented with skin-colored multiple nodules with central openings in the scrotal and anal regions. Neoplasms arising from an infundibular part of hair follicles are inverted follicular keratosis, trichilemmoma, dilated pore of Winer, tumor of follicular infundibulum, and pilar sheath acanthoma. Superficial nature of growth, connection with the epidermis, pore-like opening, proliferation of outer sheath epithelium, infundibular keratinization, and connection with the pilosebaceous structure are the common histological features of these diseases[3]. Mehregan and Brownstein[1] described pilar sheath acanthoma as being less mature than dilated pore of Winer, but more mature than the tumor of follicular infundibulum[3].

Figure 1. Multiple skin-colored nodules with central openings on the scrotal region

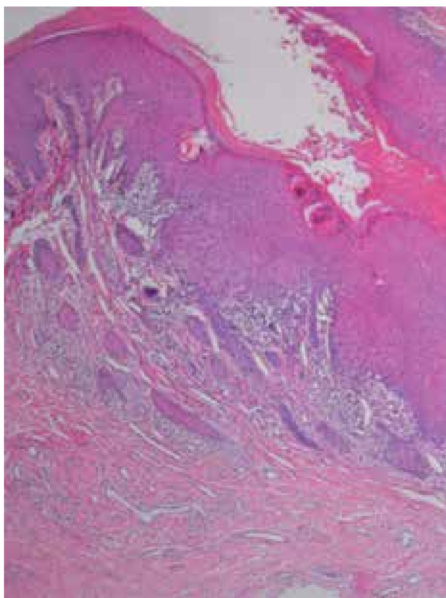

Figure 2. Dilated hair follicle with acanthotic epithelium in the center of the area and lobular keratinocyte mass around the hair follicle which is radially extending to the dermis. There are no hair formations. The cells are round to polyhedral with peripheral palisading (hematoxylin&eosin, original magnification at 40x)

The histopathological appearance of pilar sheath acanthoma has some differences from trichofolliculoma and dilated pore of Winer. Pilar sheath acanthoma is histopathologically characterized by a central, cystic invagination arising from the epidermis and following the axis of a previous hair follicle. The cyst wall is acanthotic, with small horn cyst. Although it is easy to see the mass of the compact cornified material, detecting a terminal or a vellus hair often requires multiple sections[8]. A dilated follicle or cystic lesion that contains vellus hairs is a characteristic of a typical trichofolliculoma, as well as many incomplete follicular structures bracing out from the central cavity[8]. The hair follicles in trichofolliculoma are more differentiated compared to those in pilar sheath acanthoma. Some structures that are usually seen in secondary follicles of trichofolliculoma such as an outer root sheath, inner root sheath, and trichohyaline granules are not seen in pilar sheath acanthoma. Hair shafts in central cavity are not seen in the pilar sheath acanthoma; the fibrovascular stroma is also absent[3]. Proliferation of connective tissue and sebaceous gland can alter the picture; folliculosebaceous cystic hamartoma is now accepted as an involuting lesion, while sebaceous trichofolliculoma is simply rich in sebaceous glands[8].

In dilated pore of Winer, the wall may show minimal thickening, sometimes in a papillomatous pattern and with increased melanin. Also, a large follicle with a dilated central cavity filled with cornified material is found[8]. But in pilar sheath acanthoma, the wall is thicker and more lobularly arranged[7].

Since pilar sheath acanthomas are benign neoplasms, they do not necessitate further treatment. If the patient preferred to remove these lesions for cosmetic reasons, surgical excision, electrodessication or curettage could be applied.

Although pilar sheath acanthomas are found almost exclusively on the upper lip, isolated cases have been reported on the forehead, cheek, earlobe and postauricular area. Our patient appears to be the first documented case with multiple pilar sheath acanthomas presenting on the scrotal and anal regions. We believe that the knowledge about pilar sheath acanthoma will be getting better with increasing number of publications about the disease.

Ethics

Informed Consent: Consent form was filled out by all participants**.**

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: Belkız Uyar, Concept: Belkız Uyar, Oya Nermin Sivrikoz, Design: Belkız Uyar, Oya Nermin Sivrikoz, Data Collection or Processing: Belkız Uyar, Oya Nermin Sivrikoz, Analysis or Interpretation: Belkız Uyar, Oya Nermin Sivrikoz, Literature Search: Belkız Uyar, Writing: Belkız Uyar.

Conflict of Interest: No conflict of interest was declared by the authors.

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References

1. Mehregan AH, Brownstein MH: Pilar sheath acanthoma. Arch Dermatol 1978;114:1495-7.
2. Ackerman AB, Viragh PA, Chongchitant N: Pilar sheath acanthoma. Neoplasn with Follicular Differentiation. Ed. Ackerman AB, Viragh PA, Chongchinant N. Philadelphia, Lea and Febiger, 1993;509-32.
3. Bavikar RR, Gaopande V, Deshmukh SD: Postauricular pilar sheath acanthoma. Int J Trichology 2011;3:39-40.
4. Vakilzadeh F: Pilar sheath acanthoma. Hautarzt 1987;38:40-2.
5. Choi YS, Park SH, Bang D: Pilar sheath acanthoma - report of a case with review of the literature. Yonsei Med J 1989;30:392-5.
6. Smolle J, Kerl H: Pilar sheath acanthoma-A benign follicular hamartoma. Dermatologica 1983;167:335-8.
7. Kushner JA, Thomas RS, Young RJ: An unusual location of a pilar sheath acanthoma. Int J Trichology 2014;6:185-6.
8. Rütten A: Adnexal tumors. Braun Falco’s Dermatology. Ed. Braun-Falco O, Burgdorf WHC, Plewig G, Wolff HH, Landthaler M. 3th ed. Italya, Springer Medizin Verlag Heidelberg. 2009;1377-96.