



Trichofolliculoma on gluteal area: A unique case report

Gluteal bölgede trikofoliküloma: Özgün bir olgu sunumu

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Abstract

Trichofolliculoma is a rare benign cystic nodule, consisting of a central dilated hair follicle that radiates into the surrounding dermis. Trichofolliculoma presents as a dome-shaped, solitary, 0.5-1.0 cm diameter of papule or nodule with a central pore. The clinical differential diagnosis of trichofolliculoma includes basal cell carcinoma, milia, keratoacanthoma, trichoepithelioma, and syringoma. Histopathological differential diagnosis includes trichoepithelioma, dilated pore of Winer, pilar sheath acanthoma, and basal cell carcinoma. Trichofolliculomas usually occur on the face and less frequently on the neck and scalp. The gluteal area is an unprecedented site for trichofolliculoma although some unusual locations were observed. Herein, we report a very rare trichofolliculoma of the gluteal area which was not reported in English literature.

Keywords: Trichofolliculoma, hair follicle tumors, gluteal skin

Öz

Trikofoliküloma, merkezi dilate bir kıl folikülünden oluşan ve çevredeki dermise yayılan; nadir görülen benign bir kistik nodüldür. Merkezi gözenekli, kubbe şeklinde, soliter, 0,5-1,0 cm aralığında çapa sahip bir papül veya nodül olarak ortaya çıkar. Trikofolikülomanın klinik ayırıcı tanısı bazal hücreli karsinom, milia, keratoakantom, trikoepitelyoma ve siringomayı içerir. Histopatolojik ayırıcı tanı trikoepitelyoma, Winer'in genişlemiş dilate poru, pilar kılıf akantomu ve bazal hücreli karsinomu içerir. Trikofolikülomalar genellikle yüzde, daha az sıklıkla boyun ve sağlı deride görülür. Bazı beklenmedik bölgelerde de görülmekle birlikte, gluteal bölge trikofoliküloma için eşi görülmemiş bir bölgedir. Burada çok nadir görülen ve İngilizce literatürde bildirilmemiş olan gluteal bölgedeki trikofolikülomayı sunuyoruz.

Anahtar Kelimeler: Trikofoliküloma, kıl folikülü tümörleri, gluteal deri

Introduction

Trichofolliculoma is a neoplasm that shows a panfollicular differentiation, with male predominance, and is more common in middle-aged adults and very rare in children or infants¹. Trichofolliculoma presents as a dome-shaped, solitary, 0.5-1.0 cm diameter of papule or nodule with a central pore². Kligman and Pinkus³ reported that the core of the lesion is a keratin-filled, dilated sinus or invagination, and a grotesque follicle, lined with a keratinizing stratified

epithelium. Trichofolliculomas appear mostly on the scalp or neck. Unusually, they may be present in the external auditory canal, intranasal area, genital area, lip, or vulva⁴⁻⁸. Trichofolliculoma of the gluteal skin has not been reported in the English literature. Thus, we report this case.

Case Report

A 54-year-old male patient presented with two neighboring painful lesions on the right medial gluteal area skin. They had

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quickly increased in size over the past 2 weeks. The patient's medical record, familial background, and laboratory examinations were not remarkable. A flesh-colored 1-1.5 cm nodule with a punctum in the middle and a similar small satellite lesion immediately medial to it was observed on dermatological examination (Figure 1). Drained material was not seen when the nodule was punctured. Wide local excision was performed. Postsurgery was uneventful without recurrence in the following months.

Macroscopically the tumor was a dome-shaped gray-white nodule measuring 1.5x1x1 cm (Figure 2, 3). The surgical specimen was fixed in 10% buffered formalin, routinely processed, and embedded in paraffin. The 4 µm-thick sections were stained with hematoxylin and eosin. Histopathologic examination of the nodule displayed a central infundibular cyst with secondary hair follicles from a well-circumscribed papilla which radiated to the surrounding dermis. The central follicle contained keratinous material. Near the central cyst were a few secondary hair follicles formed of congested and hyperchromatic cells. These cells aligned in a palisading pattern. The lesion was embedded in a fibrotic stroma (Figure 4, 5). Informed consent was obtained.

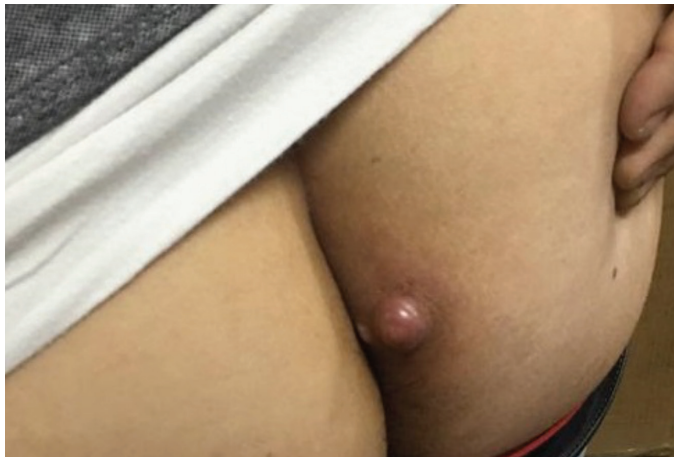


Figure 1. A flesh-colored nodule with a punctum in the middle

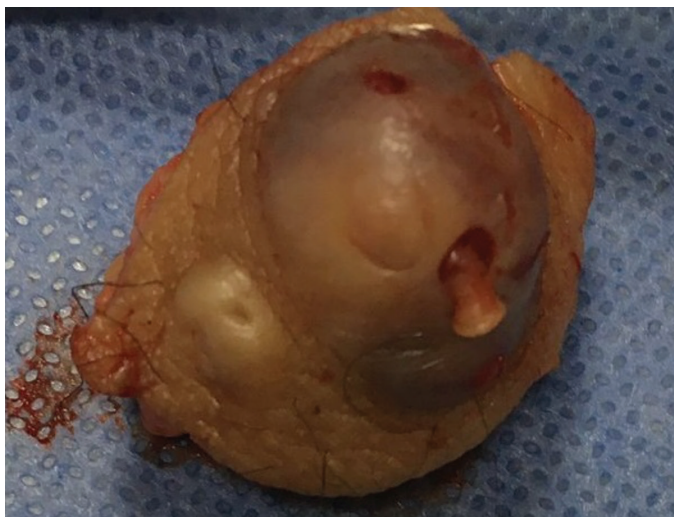


Figure 2. Excision material

Discussion

Miescher⁹ first described trichofolliculoma in 1944 as a rare benign follicular tumor. It presents as a dome-shaped, solitary, 0.5-1.0 cm diameter of papule or nodule with a central pore². Some authors considered a hair follicle component in an anomalous distribution to be a hamartoma rather than a neoplasm. It shows differentiation between a hair follicle nevus and a trichoepithelioma¹⁰. Trichofolliculomas are not part of a syndrome or are associated with a family history¹¹.

The definite etiology remained unknown and without proven relation with any systemic or dermatological diseases. However, it is believed to be caused by failed differentiation of pluripotent skin cells toward hair follicles¹². Trichofolliculomas have no malignant potential¹³ even including the single case reported with perineural invasion. Excision is curative and is usually performed for aesthetic purposes.

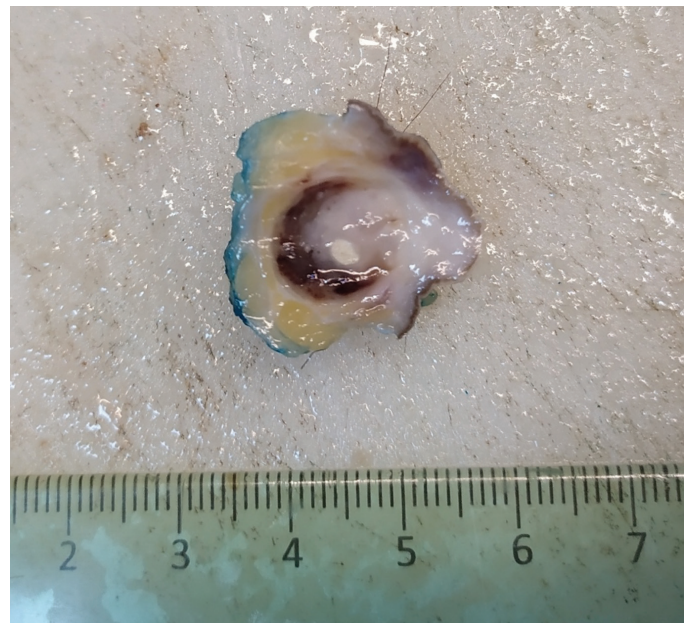


Figure 3. Dome-shaped nodule with gray-white cut surface

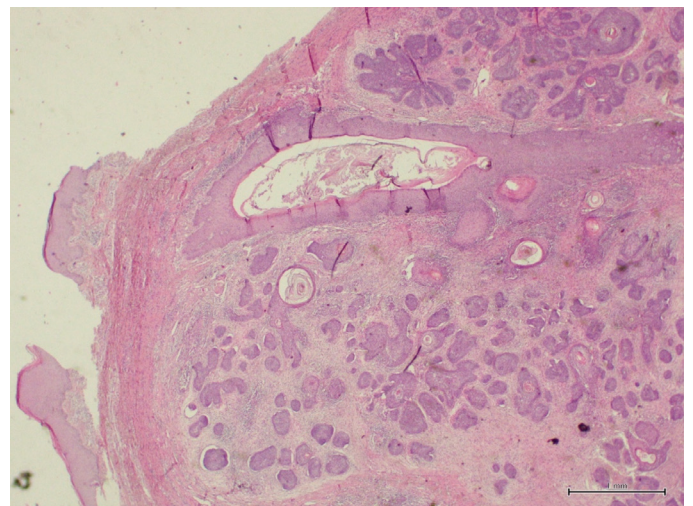


Figure 4. Central dilated hair follicle and several abortive hair follicles (x20, hematoxylin and eosin)

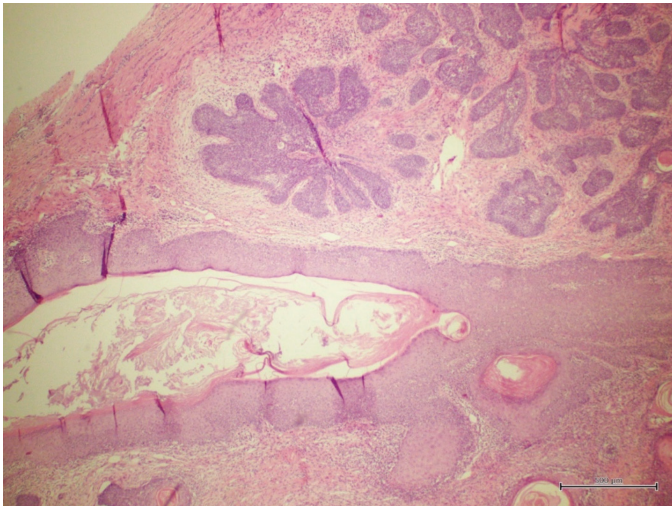


Figure 5. Hair bulbs around the central dilated hair follicles (x40, hematoxylin and eosin)

A cystically dilated keratin-filled follicle is the specific histopathologic finding of a trichofolliculoma. It is usually located in the upper dermis, with or without epidermal connection². Mature structures may generate hair shafts. Clear cells may occur in the external root sheath due to glycogen. Palisading basal cells may be seen. Collagen openly surrounds the follicles circumscribing the lesion from the dermal collagen. The stroma is fibrocytic.

A group of skin lesions has similar clinical findings to trichofolliculoma. These are basal cell carcinoma, milia, keratoacanthoma, syringoma, and trichoepithelioma. The most widespread malignant skin disease is basal cell carcinoma. It usually appears as pale pink or flesh-colored papules or nodules with branching vessels. Milia are small cysts lined by epidermis which usually occur from trauma or the use of topical drugs. They appear on the face as multiple millimetric spherical and rigid lesions. Keratoacanthoma is a dome-shaped nodule, with a central keratinous plug that is 1-2.5 cm in diameter. Trichoepitheliomas are one or more, nude colored millimetric nodules that are usually located on the face. Lastly, syringomas are multiple yellow millimetric papules mainly located on the lower eyelids.

The absence of a central pore in tissue sections can make diagnosis difficult although the microscopic findings of trichofolliculomas are quite typical. Histological differential diagnosis includes trichoepithelioma, the dilated pore of Winer, pilar sheath acanthoma, and basal cell carcinoma. Trichoepithelioma consists of nests of basaloid cells with keratinous cysts in the dermis. Tumor cells have minimal cytoplasm, large hyperstained nuclei, and a peripheral palisade. Centrally dilated follicle connected to the epithelium is seen on the dilated pore of Winer and the pilar sheath acanthoma, which are similar to the trichofolliculoma. However, immature follicles that radiated from the center are not seen¹⁴. Trichofolliculoma has well-differentiated hair follicles compared to pilar sheath acanthoma, in which underdeveloped hair follicles are seen. Outer root sheath, inner root sheath, and trichohialin granules, which are not seen in pillar sheath acanthoma, are the features seen in secondary follicles of trichofolliculoma¹⁵. Basal cell carcinoma consists of basaloid nests with peripheral palisading formations. Increased

mitotic rate, myxoid stroma, and apoptotic cells are specific findings that help differentiate from benign follicular tumors.

Trichofolliculomas usually occur on the face, and the neck and scalp are less frequent areas. Several abnormal sites, such as the ear canal, intranasal area, genital area, lips, and vulva, were reported¹⁶. However, the gluteal area is an unprecedented site for trichofolliculoma. Our case had a 1.5 cm diameter nodule and the reason for the dermatology examination was pain. To our best knowledge, this is the first report of trichofolliculoma that developed on the gluteal skin. Trichofolliculoma should still be considered in the differential diagnosis of dome-shaped and solitary gluteal nodules although very rare.

Ethics

Informed Consent: It was obtained.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: FN., E.Ş., Concept: S.C.T., C.T., Design: S.C.T., C.T., Data Collection or Processing: S.C.T., C.T., Analysis or Interpretation: S.C.T., C.T., Literature Search: S.C.T., C.T., Writing: S.C.T.

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

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