Keratoacanthoma Seen with Hidradenitis Suppurativa: A Case Report

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Abstract

Hidradenitis suppurativa (HS) is considered a primary disease of the hair follicle. The chronic occlusion of the follicular pilosebaceous unit and an associated immune response appears to be the main causes. The chronic, active, and poorly controlled disease may lead to several complications such as scars, contractures, lymphedema, osteomyelitis, and squamous cell carcinoma (SCC). We report here a 33-year-old male with HS who developed keratoacanthoma while on secukinumab treatment. The tumor representing 2 weeks of evolution in an area affected by HS (lower abdomen) was followed up after histopathological confirmation. Almost complete spontaneous regression was observed at the subsequent visits. As far as we are aware, solitary keratoacanthoma associated with HS has not been previously described. Our case shows that squamous differentiation is not limited to SCC and can develop from any scar tissue outside the anogenital region in patients with HS. Thus, the case presented here emphasizes the necessity of careful examination in scar areas as well as inflammatory lesions in HS.

Keywords: Biological therapy, hidradenitis suppurativa, keratoacanthoma, squamous cell carcinoma

Introduction

Hidradenitis suppurativa (HS) is a chronic, recurrent, inflammatory skin disease characterized by deeply located painful nodules and abscesses, often result in sinus tracts and scarring. It frequently occurs after puberty, affecting 1% of the population. The chronic occlusion of the follicular pilosebaceous unit and an associated immune response appears to be the main causes.[1] The chronic, active, and poorly controlled disease may lead to several complications such as scars and squamous cell carcinoma (SCC).

Case Report

A 33-year-old male with a diagnosis of HS since 2012 applied to our clinic. Physical examination revealed multiple inflamed, tender, discrete papules and nodules with linear scars and fistulas. He was previously treated with systemic antibiotics and isotretinoin. He had 30 pack-years of smoking and genital human papillomavirus (HPV) infection. He was included in an ongoing study and started on biological therapy (secukinumab). With the initiation of secukinumab, he achieved clinical remission. After 4 months of therapy, a new (2 weeks old) 1×1.5 cm dome-shaped nodular lesion with a crateriform hyperkeratotic center in the intersection area of the abdomen and the pubic fold was seen [Figure 1a]. An incisional biopsy was taken with the suspicion of keratoacanthoma (KA) or SCC. Almost complete spontaneous regression was observed at the subsequent visit [Figure 1b] and KA diagnosis was established with biopsy [Figure 2].

Discussion

KAs, accepted as well-differentiated SCC, are mostly solitary, rapidly growing dome-shaped nodules containing central keratin plug. There is only one previous case

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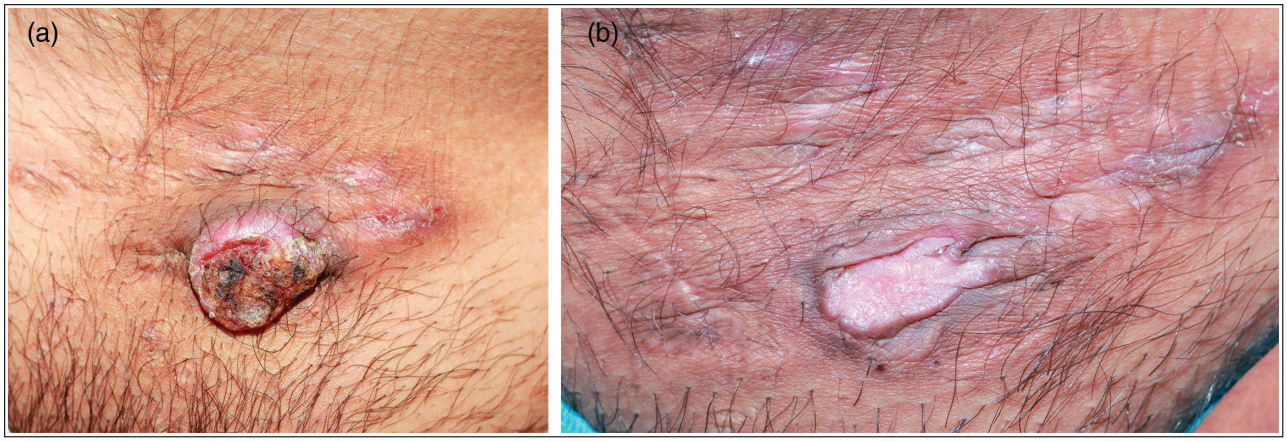

Figure 1: (a) A dome-shaped nodular lesion with a crateriform and hyperkeratotic center, measuring 1 × 1.5 cm in the intersection area of the abdomen and the pubic fold where patient has previous HS scars. (b) Ongoing assessment showed almost complete spontaneous regression of the lesion 12 weeks after the biopsy. The hyperkeratotic center was already cleared, and the lesion was flattened

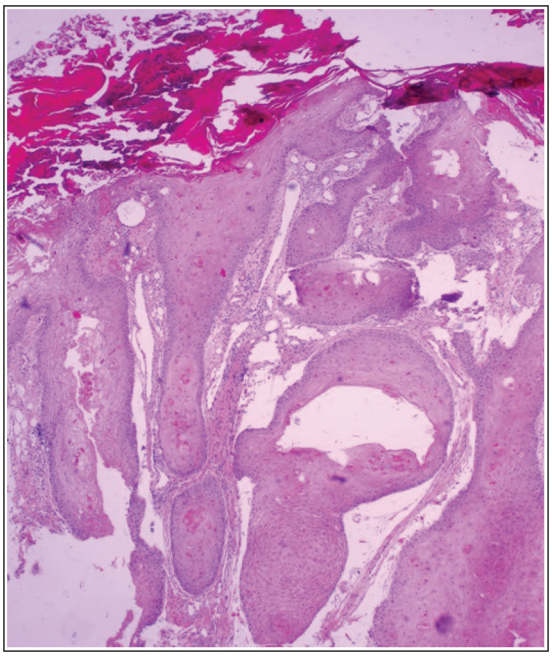

Figure 2: Biopsy shows some dysplastic cells with mitosis, absence of invasion, and nest and clusters of proliferating well-differentiating squamous epithelium (H&E X200).

report of co-occurrence of HS and KA published in the literature.[2] However, in that report, the patient had Dowling-Degos disease and multiple KAs along with HS. Fenske *et al*.[2] suggested that the association might be attributed to the common etiopathogenetic mechanism and a single underlying defect of the abnormal epithelial proliferation of the pilosebaceous apparatus as KAs also arise from hair follicles like HS. This underlying defect could be abnormal Notch signaling, which is an important protein for normal follicle development and skin appendages. Impaired Notch signaling disrupts hair follicle homeostasis and structure, leading to an inflammatory immune response like in HS.[3] Furthermore, Notch was shown to act as an epidermal tumor suppressor in nonmelanoma skin cancers, including SCC.

Another pathogenetic mechanism could be the concept of the immunocompromised cutaneous district.[4] Cutaneous scars are vulnerable sites for the development of neoplasms and dysimmune reactions. Caccavale *et al*.[4] suggested that the immunological behavior of a scarred area is different from that of the rest of the body. Thus, the scar area's destabilization could be a predisposing factor to tumors in HS. A recent literature review found 85 cases of SCC arising on scars in HS, mainly in men and in the anogenital region.[5]

The development of squamous tumors is likely multifactorial. Other well-described risk factors are smoking and HPV infection.[5] Smoking was reported to downregulate Notch signaling in airway epithelial cells, the pathway involved in HS and nonmelanoma skin cancers.[5] Thus, the effects of smoking may augment preexisting impairment of Notch signaling, which may increase susceptibility to SCC.[3]

Furthermore, according to Jourabchi *et al*.,[5] consideration should be given to the increasing use of biological immunosuppressants in HS and the association between chronic immunosuppression and tumors. Thus, secukinumab therapy might be an accelerating factor in our patient along with all these risk factors and pathomechanisms.

Our case shows that squamous differentiation is not limited to SCC and can develop from any scar tissue outside the anogenital region in patients with HS. Thus, the case presented here emphasizes the necessity of careful examination in scar areas as well as inflammatory lesions in HS.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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