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Squamous cell carcinoma of the perineum masquerading as necrotizing hidradenitis suppurativa

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Abstract

Many cases of superinfected hidradenitis suppurativa (HS) involve multiple species of bacteria, but gas-producing infections are rare and can complicate the clinical picture. Additionally, recognizing squamous cell carcinoma (SCC) as a complication of longstanding HS is imperative. Herein, we present a unique case of a severe emphysematous HS that was initially mistaken for Fournier gangrene and eventually diagnosed as superinfected SCC.

Keywords: hidradenitis suppurativa, condyloma, squamous cell carcinoma

Introduction

Hidradenitis suppurativa (HS) is a condition of chronic inflammatory follicular occlusion of the folliculosebaceous unit and apocrine glands most often in the axillae, inguinal, and anogenital regions, but also sometimes involving the paramammary and buttock regions. It is characterized by deep, painful nodules that become abscesses with draining sinus tracts, bridged scars, and double-ended pseudo-comedones. To date, it has been established that longstanding HS can evolve into squamous cell carcinoma (SCC) and that concurrent HPV may increase risk for SCC [1]. Additionally, SCC associated with HS follows a particularly aggressive course with

high risk of metastasis and high mortality [2]. However, no guidelines exist regarding screening for SCC in patients with HS.

Although hidradenitis suppurativa (HS) can present acutely with secondary infection requiring surgical debridement, we present a rare case of a particularly severe emphysematous HS that was initially mistaken for Fournier gangrene and eventually was diagnosed pathologically as squamous cell carcinoma in a background of koilocytic change. Herein, we recommend that surgically excised tissue from patients with HS have histological examination as a screening method for SCC.

Case Synopsis

A 47-year old morbidly obese man with a 10-year history of hidradenitis suppurativa (HS) was admitted for a one-month history of worsening pain and drainage from a chronic non-healing perineal wound. The patient reported having previously seen multiple outpatient physicians at different facilities for this condition. He was not being followed regularly in a dermatology clinic. Because of his social situation, he was using the emergency department as his primary form of care. The patient also endorsed a 200 pound weight gain over the past three years, which had exacerbated his perineal HS. It was in the context of this complex social history



Figure 1. Bilateral axillae with dormant sinus tracts and double-headed comedones.

that this patient presented for the first time to our institution's emergency department where he was admitted to the general surgery service. Consultations were placed for the plastic surgery, urology, infectious disease, and dermatology departments.

On exam, the patient was afebrile, weighed 478 lbs., and was tearful. The bilateral axillae and inguinal regions had scarred sinus tracts and double-headed comedones without active lesions (**Figure 1**). On the perineum, the patient had a 6-7 cm exophytic mass with expression of a malodorous greenish-yellow purulent discharge (**Figure 2**); the lesion was mildly tender to palpation without subcutaneous crepitus.

CT of the abdomen/pelvis revealed tissue stranding from the scrotum to proximal thigh with gas bubbles in the subcutaneous soft tissue of the groin. Concern for Fournier gangrene prompted initiation of broad-



Figure 2. On the perineum, the patient had a 6-7 cm exophytic mass with expression of a malodorous greenish-yellow purulent discharge (left). Post-excision of exophytic mass revealing diagnosis of squamous cell carcinoma (right).

spectrum antibiotics and urgent surgical debridement, which involved removal of a 29.5 x 13.0 x 4.0 cm mass that was sent for histological evaluation. Culture results revealed multiple facultative organisms (*Klebsiella*, Enterococci, *E. Coli*), non-predominant Group A strep and *Pseudomonas*, but no anaerobes and did not meet criteria for Fournier gangrene.

Histopathological examination of the specimen showed extensive areas of inflammation consistent with HS as well as an infiltrating and moderately differentiated keratinizing SCC with high Ki67 index extending to the surgical margin, but there was no

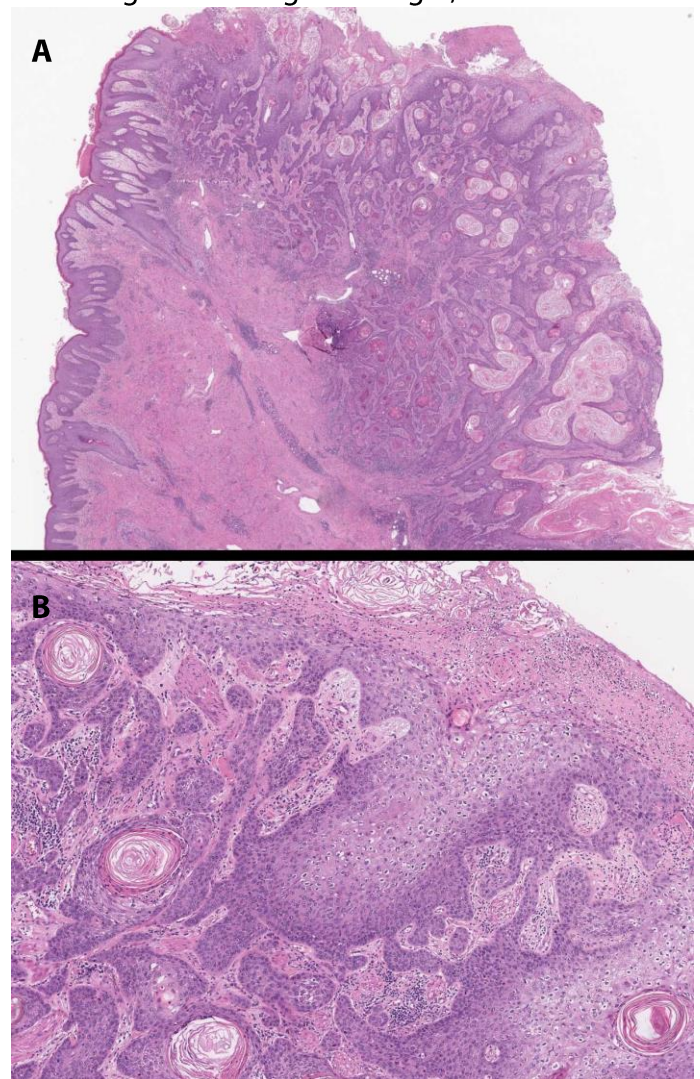


Figure 3. Histologic sections show an ulcerated, markedly proliferative squamous proliferation emanating from hyperplastic epidermis, **A)** 20x. **B)** Close examination shows infiltration of nests and fronds of atypical squamous cells into surrounding inflamed and desmoplastic stroma. Examination of more superficial keratinocytes shows the suggestion of koilocytic/HPV-related change, 100x. H&E.

evidence of lymphatic or perineural invasion (**Figure 3**). Notably, some areas were associated with koilocytic change and P16 reactivity, suggestive of concurrent condyloma (**Figure 4**). Repeat biopsies of the surgical margins were negative. Therefore, the oncology consultant recommended avoiding radiation while the patient's wound was actively healing, but to consider radiation therapy and possibly adjunct cetuximab after wound healing was achieved. The patient was transferred to an outside hospital for continued wound care. Unfortunately,

two months into his hospital stay, the patient was noted to have a perineal polypoid growth on exam and palpable left inguinal lymphadenopathy. A biopsy was done that showed recurrent SCC. CT of the abdomen and pelvis showed inguinal lymphadenopathy, felt to be either reactive or neoplastic. After discussion at tumor board, the patient has started neoadjuvant chemotherapy with paclitaxel, ifosfamide, and cisplatin with the goal of shrinking the tumor prior to complete resection with lymph node dissection and adjuvant therapy.

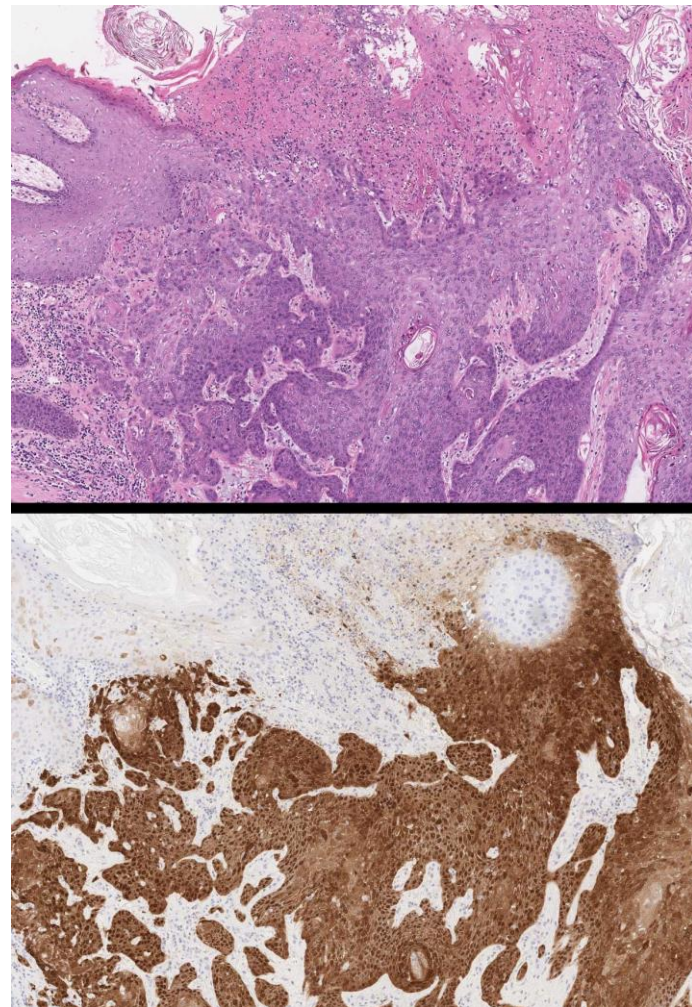


Figure 4. H&E and p16 immunohistochemistry (upper and lower pane): Histologic sections show ulcerated squamous cell carcinoma with a markedly proliferative and infiltrative profile, to include single tumor cell invasion into inflamed and desmoplastic stroma. Adjacent more normal appearing hyperplastic epidermis is observed at the left ulcer edge (upper pane, 100 \times). Performed p16 immunohistochemistry shows strong staining within tumor cells and supports a viral (HPV)-related association (lower pane – p16 immunohistochemistry, 100 \times).

Case Discussion

HS is a condition of chronic inflammatory follicular occlusion typically within intertriginous areas. It is characterized by double-headed comedones and deep, painful nodules that become abscesses with draining sinus tracts and scars. A recent review reported 80 cases of longstanding HS evolving into SCC and discussed HPV as a potential risk factor [1]. Additionally, SCC associated with HS appears to follow a particularly aggressive course with a high risk of metastasis and mortality [2]. Given the degree of infiltration and the presence of p16 reactivity seen on the patient's biopsy, the pathologist favored that the patient's SCC was associated with high-risk HPV, or arising from a pre-existing condyloma [3]. The patient did endorse a history of multiple warts in the inguinal and perineal area over the past 20 years. Another possibility is that the patient's SCC arose from a background verrucous carcinoma, such as Buschke-Lowenstein type giant verrucous carcinoma [4].

Based on the authors' experience as a referral center with an HS clinic, we recommend the following for patients with severe longstanding HS: When necrotizing soft tissue infection is suspected in the perineum, surgical exploration should not be delayed while awaiting culture results. Surgically excised tissue should be evaluated histopathologically to rule out SCC. Obtain patient's smoking history and history of condyloma; both are associated with severe HS and future SCC development. Consider screening for depression, as

HS patients may have underlying psychological distress that may lead to secondary weight gain, aggravation of HS, and complication of the evaluation and treatment. And lastly, in patients with morbid obesity or steady weight gain, consider early referrals for nutrition or bariatric surgery consultations. Higher body mass index can hinder a patient's ability to keep HS areas clean and potentially predispose patients to secondary infection.

Conclusion

Many cases of infected HS involve multiple species of

bacteria, but gas-producing infections are rare and can complicate the clinical picture. In cases in which necrotizing soft tissue infection is suspected, surgical exploration should not be delayed while awaiting culture results. Additionally, the occurrence of squamous cell carcinoma in longstanding hidradenitis suppurativa is a well-recognized, although rare, phenomenon, and debrided tissue should be sent for histopathological screening. It is crucial to consider SCC in areas of non-healing wounds in HS patients to prevent significant morbidity and mortality.

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