Case report of hidradenitis suppurativa localized to the face in an HIV patient

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Abstract

Hidradenitis suppurativa is a chronic, debilitating inflammatory skin disease. Case reports of individuals with hidradenitis suppurativa presenting exclusively on the face, as well as reports of individuals with HIV and hidradenitis suppurativa, are rare. Here, we present the case of an HIV-positive man who presented with hidradenitis suppurativa localized only on his face. We also review facial hidradenitis suppurativa and hidradenitis suppurativa in HIV patients.

Keywords

Hidradenitis suppurativa, face, human immunodeficiency virus

Introduction

Hidradenitis suppurativa (HS), also known as acne inversa, is a chronic, recurrent, debilitating inflammatory skin disease characterized by painful, deep-seated, inflamed papules, nodules, cysts, abscesses, and sinus tracts, mucopurulent discharge, and progressive scarring. It is a disorder of the terminal follicular epithelium and most often occurs in the axillae, sub-mammary folds, groin, perineum, buttock, and medial thighs. More rarely, HS can affect the scalp, face, neck, and back.^{1,2}

There are very few reports of HS on the face^{3–6} and only a single previously reported case of HS exclusively localized to the face.⁵ Cases of HS in individuals with HIV are also rare, with only six previously reported cases^{7–12}; however, a recent retrospective, cross-sectional study revealed a statistically significant association between HS and HIV.¹³ Here, we report on a unique case of HS affecting only the face of a man with HIV.

Case report

A 31-year-old Caucasian man presented to our clinic with a 4-month history of a solitary mass on the left mandibular area believed to be an abscess. It was slightly tender and exuded pus from several openings within the mass. He had a history of being HIV positive for 7 years. His HIV was well controlled on once daily oral Biktarvy containing bictegravir, emtricitabine, tenofovir, and alafenamide, and he had a normal CD4⁺ count and an undetectable viral load. Clinical examination showed a 5 by 5 cm erythematous mass with a rough surface and a few draining sinuses over the right mandibular area (Figure 1). A previous X-ray showed no evidence of periostitis



Figure 1. Erythematous mass over right mandibular area.

or bone destruction and he had previously seen a dentist and a dental sinus was ruled out. The differential diagnosis when seen included infectious causes such as actinomycosis, a deep

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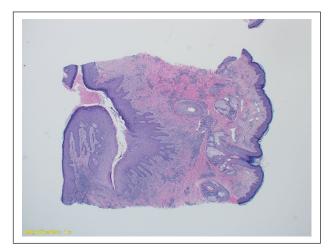


Figure 2. Squamous-lined cystic structure at the deep skin biopsy edge compatible with a sinus tract.

fungus, tuberculosis (TB), and atypical mycobacteria. Two 4-mm punch biopsies were done, one for histology and one for cultures. Skin biopsy showed a squamous-lined cystic structure at the deep biopsy edge compatible with a sinus tract (Figure 2). Periodic acid-Schiff (PAS), Grocott's methenamine silver (GMS), Ziehl-Neelsen (ZN), and Fite stains were all negative. Cultures sent for aerobic and anaerobic cultures grew a few Staphylococcus aureus but were negative for Actinomyces species. Tissue culture for deep fungus, TB, and atypical mycobacteria were all negative. He was treated with oral doxycycline 100 mg PO BID for 2 months, and when reassessed, the morphology of the lesion had markedly changed (Figure 3) to a flesh-colored indurated plaque with marked cribriform scarring. A full skin examination at this time showed no lesions in the axillae, groin, and buttocks except for a 5-mm depressed scar in the left lower abdomen which the patient attributed to an "ingrown hair."

At this time, the clinical appearance suggested HS localized to the face. An incisional biopsy from the plaque including an area of cribriform scarring was submitted for pathology and showed an acute folliculitis with deep dermal fibrosis and a lymphoplasmacytic infiltrate. All repeated cultures were negative. He was put on a course of oral isotretinoin and the plaque was injected with intralesional triamcinolone acetonide 10 mg/cc and he has a good response to this treatment.

Discussion

HS can occasionally affect the face when simultaneously presenting in more typical sites (e.g. axillae, groin); yet, reports of HS exclusively occurring on the face are exceedingly rare. A review of the literature revealed only one case in which a 30-year-old man had a 2-year history of HS involving the preauricular areas and no other areas. He was treated with doxycycline and topical retinoids and clindamycin gel, with



Figure 3. Flesh-colored indurated plaque with marked cribriform scarring.

some improvement. There are two other case reports of predominantly facial HS but in one the posterior neck was involved as well as the jawline and, in the other case, the anterior neck was also involved along with the jawline, with this case said to mimic severe cystic acne.^{4,6}

Reports of HS in immunocompromised patients, and specifically those with HIV, are also unusual. There are six published case reports in which HS occurred in an individual with HIV (Table 1). Five of these six individuals (83.3%) were male and the mean age was 36.2 ± 14.0 years, ranging from 13 to 47 years of age. The mean duration with HS was 5.4 ± 7.7 years, with at least three of these patients suffering from long-standing, severe refractory HS.

The patient detailed here represents only the second report of HS restricted exclusively on the face and the seventh case report of an HIV-positive individual with HS; this is also the only case of HS localized to the face in an HIV patient. The affected sites among the seven reported individuals with HIV and HS were broadly similar to HS in the general population; although, the occurrence at less-characteristic sites seems to be slightly more common. For instance, in three of the seven cases (42.9%), face was involved (Table 1).

The precise etiology and pathogenesis of HS remains unknown; however, a number of predisposing factors, such as obesity and smoking, have been suggested to have a role. ^{14,15} Interestingly, with regard to HIV infections, Deng et al. ¹³ found that patients with HIV are over six times more likely to have a concomitant diagnosis of HS when compared with those without HIV. The increased release of

Rankin and Haber 3

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Source	Sex	Age (years)	Duration	Location	Severity	Treatments failed	Treatment
Alecsandru et al. ⁷	Male	47	"Long- standing"	Axilla, gluteal, perianal, thoracic, abdominal regions	Severe refractory	Erythromycin stearate, minocycline, cefuroxime, doxycycline, topical clindamycin	Infliximab, topical clindamycin
Khambhati et al. ⁸	Male	35	2 years	Face, thighs, gluteal region, and axilla	_	-	Doxycycline, ibuprofen, dapsone
Husein-ElAhmed et al. ⁹	Male	47	18 years	Inguino-scrotal, perineum, and gluteal areas	Severe refractory	Isotretinoin, tetracycline, erythromycin, cephalexin, ciprofloxacin	Infliximab, neodymium- yttrium-aluminum- garnet laser therapy
Prabhu et al. ¹⁰	Male	13	I month	Right axilla	-	-	Antiretroviral therapy (zidovudine lamivudine, nevirapine)
Dhadke et al. ¹¹	Male	"Middle- age"	10 days	Chest, neck, shoulder, axilla, and right cheek	_	Local antibiotic washes, amoxicillin and clavulinic acid, prednisolone	_ , ,
Molina-Leyva and Badiola ¹²	Female	39	12 years	Both axillae	Severe refractory	Doxycycline, clindamycin with rifampin, surgical interventions	Adalimumab
Rankin and Haber, current case	Male	31	4 months	Right jawline	Moderate	Doxycycline	Isotretinoin, intralesional triamcinolone acetonide

pro-inflammatory cytokines associated with HIV, including tumor necrosis factor- α (TNF- α) and interleukin-6 and -18, was also thought to contribute to HS.¹⁶

HS is notoriously difficult to manage and, for individuals with HIV and HS, treatment options can be more limited in an attempt to not increase the immunodeficiency. In the general population, severe or recalcitrant HS cases often require systemic therapies, such as oral antibiotics, retinoids, steroids, immunosuppressives, and biologics. Anti-TNF-α agents, including infliximab and adalimumab, have demonstrated efficacy for HS in patients without HIV. Infliximab has also been used successfully to manage HS in two HIVpositive patients;^{7,9} although, in one of these cases, the therapy may have contributed to a reduced CD4⁺ lymphocyte count and increased viral load.7 Adalimumab has additionally been used to manage HS in one individual with HIV and demonstrated sustained, complete clinical response without infectious complications. 12 In our patient, the HS was relatively mild and localized to the right mandibular area. He was initially treated with oral doxycycline, and subsequently has responded well to a course of oral isotretinoin along with intralesional triamcinolone acetonide injections.

The case reported here is unusual because of the restriction of HS to the face and because the patient was HIV positive. Prompt recognition and initiation of treatment may have slowed the progression of the disease and limited decreases in

the quality of life. We suggest that clinicians be aware that HS can occur exclusively on the face and, in these situations, may suggest an infectious etiology or dental sinus. We also encourage more reports of HS in HIV patients to better estimate the incidence of these two chronic diseases coexisting.

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Informed consent

Informed consent has been obtained from the patient by the authors.

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