SKIN

BRIEF ARTICLE

Two Cases of Mycetoma-like Hidradenitis Suppurativa

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ABSTRACT

Few cases of hidradenitis suppurativa (HS) mimicking mycetoma have been reported in the literature. Herein, we discuss two male patients (57 and 38 years old) with Crohn's disease presenting with chronic, painful gluteal nodules, draining sinus tracts, and scarring concerning for mycetoma, metastatic Crohn's disease, or HS. Biopsies were consistent with HS with no evidence of Crohn's disease. Triple wound cultures for both patients were negative. However, one patient's (57 M) fungal culture was positive for Aspergillus sclerotiorum, but a referral to Infectious Disease concluded that concern for Aspergillus mycetoma was low. Following thorough workup, both patients were diagnosed with HS and were started on adalimumab for dual Crohn's and hidradenitis therapy. The first patient (57 M) had already experienced improvement of his gluteal lesions on this regimen at his one-month follow-up appointment. The second patient (38 M) had been prescribed adalimumab in early 2024 but only received one injection due to delivery scheduling challenges. He was recently reinitiated on the medication. Overall, we highlight two cases of HS presenting similarly to mycetoma and treated with adalimumab.

INTRODUCTION

Hidradenitis suppurativa (HS) is a chronic follicular occlusive disease primarily affecting intertriginous regions such as axillae, groin, inframammary area, perianal region, and perineal region. Disease spectrum ranges from mild recurrent papules, pustules, and nodules to severe, chronic abscesses with draining sinus tracts and significant scarring.[1,2] Historically, HS was thought to be related to apocrine glands, but recent studies suggest the disease stems from occlusion of follicular portions of folliculopilosebaceous units.[2,3] Metastatic Crohn's disease is a rare Crohn's manifestation in which skin lesions form noncontiguous with the gastrointestinal tract, mimicking skin conditions like HS. Although metastatic Crohn's disease is considered in differential diagnoses for HS, few reports exist of HS presenting similarly to mycetoma.[4,5,6] This report examines two cases where HS presented with characteristics resembling mycetoma.

CASE REPORT

Case A

A 57-year-old Black male with history of Crohn's disease presented with lesions on his right buttocks present for 1-2 years. Past treatments included intermittent antibiotic courses and wound care, with a recent course of clindamycin completed two months before his clinic visit. The patient had previously been treated with Infliximab for Crohn's disease but had discontinued it 6-12 months prior to this evaluation. The rash began as a small right buttock lesion and expanded across the gluteal region. Physical examination revealed a chronic wound on the right buttock and intergluteal cleft, characterized by skin breakdown, indurated plaques, and draining nodules (**Figure 1A**). Differential diagnoses included actinomycetoma, eumycetoma, metastatic Crohn's disease, and HS, though HS was considered less likely. A biopsy indicated marked pan-dermal acute, chronic, and granulomatous inflammation consistent with HS. No definitive evidence of infection was found on multiple levels and histochemical stains. Biopsy lacked well-formed granulomas, making cutaneous Crohn's disease unlikely. Triple tissue cultures (AFB, fungal, bacterial) were negative, but fungal skin swab grew Aspergillus sclerotiorum. Despite the positive Aspergillus culture, the infectious disease specialist deemed risk of Aspergillus mycetoma low, suggesting Aspergillus sclerotiorum might be a contaminant. The patient was started on adalimumab for dual Crohn's and HS therapy. At one-month follow-up, his gluteal lesions showed noticeable improvement (**Figure 1B**).

Case B

The second patient was a 38-year-old Black male with a history of Crohn's disease, for which he had undergone a diverting colostomy, as well as type 2 diabetes mellitus, malnutrition, and chronic gluteal

Figure 1. Patient A (57 M). **(A)** Initial clinic visit. Right buttock and gluteal cleft with well-defined, indurated, infiltrated plaque, scattered erythematous pustular nodules with sinus tracts and active drainage. **(B)** Follow-up after one month of adalimumab.

wounds persisting for nearly a decade which were previously diagnosed as HS. He had undergone multiple hospitalizations due to complications from his comorbidities and had only received a formal diagnosis of Crohn's disease in early 2024. Dermatology was involved in managing his chronic gluteal wounds during multiple hospitalizations. A biopsy in late 2022 showed significant dermal and lymphoplasmacytic inflammation consistent with HS or chronic wound, with no organisms visible on special stains. Despite multiple rounds of antibiotics and steroids, along with incision and drainage, the patient's condition improved minimally. In early 2024, dermatology initiated adalimumab treatment, but only one injection was administered as the patient struggled to attend follow-up dermatology appointments. In mid-2024, he was hospitalized for hyperglycemia concerning for diabetic ketoacidosis, during which dermatology was reconsulted for the same gluteal lesions previously seen. His worsening gluteal wounds, now draining and showing scarred nodules with sinus tracts (**Figure 2**), led to re-evaluation of his HS diagnosis. The differential included HS, mycetoma, botryomycosis, and metastatic Crohn's Disease. Biopsy was consistent with chronic wound, with one sinus tract visualized on biopsy suggestive of HS. No organisms were observed on PAS or FITE stains, and there was no evidence of Crohn's disease. Triple tissue cultures were negative. After discharge, the patient was reinitiated on adalimumab for dual Crohn's and HS therapy, and he has received one dose of the medication.

Figure 2. Patient B (38 M) during hospitalization. Bilateral buttocks with firm, tender, scarred nodules, sinus tracts coalescing into well-defined plaques, and purulent drainage.

DISCUSSION

The presentation of HS with features resembling mycetoma is rarely described in literature. Clinically, HS typically affects the axillary, mammary, inframammary, and inguinal regions,[2] whereas mycetoma is commonly observed on the feet and hands.[7] Although gluteal involvement can occur in both conditions, it is less prevalent in mycetoma than in the aforementioned areas.[4,5]

Mycetoma pathogenesis involves subcutaneous fungal or bacterial infections, particularly in immunocompromised individuals. Lesions begin as painless subcutaneous nodules that can develop into tumors or abscesses. Sinus tracts may form, draining purulent material or grains from causative organisms.[7]

In Case A, high concern for mycetoma stemmed from the patient's pustular, draining nodules, immunosuppressed status, and the fungal skin swab growing Aspergillus sclerotiorum. Despite Aspergillus being a potential mycetoma pathogen, Aspergillus sclerotiorum is more frequently a contaminant than an opportunistic pathogen. Biopsy results, along with the infectious disease team's assessment, suggested HS as the more likely diagnosis. Improvement in lesions following a month of adalimumab therapy further supported this.

Similarly, Case B presented a challenging scenario with HS-like features and complications from comorbidities. Biopsy was consistent with chronic wound. One sinus tract seen on biopsy was consistent with HS, with no organisms or signs of infection seen on special stains. Triple tissue cultures, collected as part of the complete diagnostic workup, were negative, so mycetoma was deemed less likely than HS. The patient's response to adalimumab is still in early stages, with only one dose administered to date.

Interestingly, both patients had Crohn's disease, raising the possibility of metastatic Crohn's. However, the diagnosis was excluded as biopsies were consistent with HS without evidence of Crohn's. HS is a rare extraintestinal manifestation of Crohn's disease, yet the conditions are strongly associated.[8,9] Patients with HS have a significantly higher risk of developing Crohn's, with studies showing relative risk nine times higher than the general population.[10] Incidence of HS among Crohn's patients ranges from 15-26%.[11,12] Both conditions share overlapping pathogeneses and clinical features, including dysfunctional Th1 inflammatory responses, sinus tract formation, scarring, granulomatous inflammation, and response to anti-TNF-α therapy.[8,9]

CONCLUSION

This case series highlights two novel instances of male patients with Crohn's disease presenting with chronic gluteal wounds initially concerning for mycetoma but ultimately diagnosed as HS. Both patients were treated with adalimumab, with early promising outcomes observed in one case. This report underscores importance of a comprehensive diagnostic approach when faced with atypical HS presentations.

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