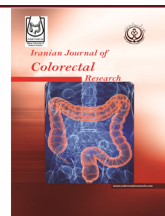


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Hidradenitis Suppurativa Complicated by Actinomycosis and Viral Hepatitis C: A Case Report

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Abstract

Introduction: Hidradenitis suppurativa (HS), also known as acne inversa, is a chronic inflammatory skin disease that is more common in women, especially during puberty and menopause. It is believed that the initial pathogenetic mechanism is hyperkeratinization of the follicular cavity, followed by follicular occlusion, expansion, and rupture. Ultimately, the presence of bacterial agents in the focus leads to a local inflammatory reaction. Many concomitant diseases and triggers have been identified, including bacterial flora, which can significantly complicate the course of HS, leading to disease chronicity and a poor response to therapy. Early diagnosis of HS leads to timely treatment; the diagnosis is based on clinical features. The appropriate treatment must be chosen depending on the stage of severity.

Case Presentation: We present a case of HS complicated by actinomycosis and viral hepatitis C. Actinomycosis is a rare diagnosis, especially in developed countries. In clinical practice, the diagnosis of actinomycosis can be difficult due to the pathogen's nonspecific clinical and pathological features, inherent difficulties in vitro cultivation, and complex histological characteristics.

Conclusion: Given the growing number of immunocompromised patients worldwide (for example, HIV-infected or hepatitis C patients) and the numerous reports of infection with actinomycetes in these conditions, actinomycosis should be considered as a possible opportunistic infection. Identifying and eliminating concomitant bacterial flora can significantly increase the chances of successful and effective therapy of HS.

Keywords: Hidradenitis suppurativa, Follicular occlusion syndrome, Actinomycosis, Viral hepatitis C

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Introduction

Hidradenitis suppurativa (HS), also known as acne inversa, is a chronic inflammatory skin disease that affects the follicular part of the pilosebaceous unit and manifests in painful nodes,

abscesses, and scars. Clinical manifestations of HS are located in areas with more receptors for sex hormones (androgens and estrogens) than other areas of the skin, as well as in areas subject to friction, for example, in the axillary or inframammary area. Nguyen et al. suggested that HS is caused by chronic

inflammation with changes in tissue architectonics, follicular hyperkeratosis, and expansion and rupture of follicles in the dermis (1).

The prevalence of HS is estimated at 1% among the general population, although the true percentage may be much higher because patients tend to hide the clinical manifestations. According to official statistics, the incidence of HS has more than doubled from 1986 (4.0 patients per 100,000) to 2008 (10.0 patients per 100,000). In 2021, the global prevalence of HS in the world was estimated at 0.00033–4.1%, resulting from improved diagnosis and awareness of skin disease (1). Clinical observations indicate that HS is three times more common in women than men (2). HS can manifest before and after puberty; however, most studies indicate that HS usually occurs after puberty (usually at the age of over 20 years), and the most severe course is seen in the third or fourth decade of life. HS sometimes occurs after menopause (3). Most patients suffer from mild or moderate symptoms, with about 4–22% experiencing a severe course of the disease (4).

Although the pathogenesis of HS is currently unclear, it is believed that the first link of the pathogenetic mechanism is hyperkeratinization of the follicular cavity, followed by follicular occlusion, expansion, and rupture. Ultimately, the presence of bacterial agents in the focus triggers a local inflammatory reaction (5). Due to this pathogenetic mechanism, the disease is also referred to as “follicular occlusion syndrome” (6). According to various clinical observations and scientific studies, the factors aggravating the course of HS are genetic, immune, hormonal, mechanical and mental stress, obesity and metabolic syndrome, and climatic factors (7-10).

It was previously believed that bacterial infections cause HS. However, recent studies call this into question due to sterile bacterial cultures from lesions (7) and the ineffectiveness of antibiotic treatment in many patients (8). In other studies, various skin microbiomes were detected in patients with HS, consisting mainly of non-pathogenic bacteria (11). For example, scientists from France studied the microbiome of 102 lesions; polymicrobial anaerobic microflorae including severe anaerobes, *Streptococcus milleri*, and actinomycetes were found in 24% of abscesses or nodules and in 87% of chronic purulent skin lesions (12). Apparently, HS develops following an inadequate immune response to commensal bacteria due to dysregulation of both the innate and acquired immune systems. Bacteria take part in initiating the immune response in HS but are not its root cause (12).

As a rule, verification of HS diagnosis is based on clinical signs, including three criteria:

1) Typical morphology (nodules, abscesses, fistula passages, and scars).

2) Localization of lesions (intertriginous areas, armpits, inframammary folds, inguinal folds, buttocks, perianal area, and perineum).

3) Recurrent and chronic course.

Early diagnosis of HS leads to timely treatment. The diagnosis of HS is based on clinical features, and appropriate therapy should be chosen depending on the Hurley stage of severity. At stage I (single abscesses without fistula formation), 1% clindamycin is prescribed locally; intralesional corticosteroids and puncture aspiration of the contents of the nodules are also indicated. At stage II (recurrent abscesses with fistulous passages and scars), systemic antibiotics of the tetracycline group (doxycycline, minocycline), systemic retinoids (acitretin), sulfones, combined oral contraceptives, spironolactone are prescribed. At stage III (multiple interconnected fistula passages and abscesses), targeted biological therapy is indicated (adalimumab, infliximab, ustekinumab, or anakinra); if these agents are not available, short courses of systemic glucocorticosteroid therapy are recommended (e.g., prednisone 40–60 mg/day).

As a clinical example, we present a case of HS complicated by actinomycosis and viral hepatitis C.

Case Presentation

Patient A, born in 1978, was hospitalized in the surgical department on December 14, 2022, with complaints of purulent discharge, redness, and swelling of formations in the axillary area, the perineal area (Figure 1), and on the left breast. The patient had suffered from HS since 2005 when this diagnosis was first established. She noted frequent disease relapses, biopsies, and drainage of abscesses in the axillary, inguinal, and perianal areas. She received various groups of antibacterial drugs, with temporary clinical improvement. Tuberculosis was denied. The patient has had viral hepatitis C since 2016 (inactive phase). The family history and allergy history were negative.



Figure 1: Clinical manifestations of hidradenitis suppurativa in the inguinal area and perineum.

At the time of this hospitalization, the patient was experiencing another relapse. On 12/14/2022, a wide autopsy was performed along with excision of scar deformities and drainage (Figure 2). Examination of the wound discharge for microflora and sensitivity to antibiotics found: *Actinomyces turicensis* and *Peptostreptococcus spp.* There are no criteria for interpreting the sensitivity of these microorganisms (according to EUCAST 2022), meaning that treatment should be carried out empirically. Microflora growth was detected. In the tests for viral hepatitis, HBsAg was not detected, though HCV AT M+G was detected. Tests for HIV and syphilis returned negative. The final diagnosis was HS complicated by actinomycosis and viral hepatitis C.

Discussion

Actinomycosis is a chronic disease caused by *Actinomyces* bacteria. For a long time, actinomycetes were mistakenly considered fungi, but after studying their morphology and biological properties, there was a reason to attribute these microorganisms to bacteria (13). The most common etiological species are *A. israelii*, *A. naeshundii*, *A. odontolyticus*, *A. viscosus*, *A. meyeri*, and *A. gerencseriae* (14). Actinomycetes are found in healthy people in the oral cavity, lacunae of the tonsils, and mucous membranes of the gastrointestinal tract (13).

Actinomycosis is rare; the disease is seen in approximately 1:300,000 of the global population annually. It is much more common in men than in women (3:1), except for pelvic actinomycosis. The disease may occur in all age groups but mostly affects those aged 20–50 years (13).

Skin/soft tissue lesions are rare and may result from trauma, surgery, or the spread of infection from other organs (14). Currently, it is assumed that actinomycosis occurs much more often in people with weakened immunity than in healthy people (14). Cutaneous actinomycosis is usually a secondary disease that occurs either as a result of spreading from a deeper focus of infection or hematogenic spreading from an extra-cutaneous (often pulmonary) source (15). Cutaneous actinomycosis often occurs in the perianal area and is clinically manifested by the presence of indurative infiltrates, fistulous passages, and rough scars; therefore, given the localization, significant difficulties arise in the differential diagnosis between actinomycosis and HS (16).

The presented clinical case demonstrates, firstly, the importance of prescribing adequate therapy corresponding to the severity of the disease. The patient was not prescribed the targeted biological therapy that she needed. Instead, multiple surgical excisions and short courses of antibacterial drugs were given, contributing to the progression of this disease, the development of branched fistula passages, rough scars, as well as frequent recurrence and a significant decrease in the quality of life. It seems quite likely



Figure 2: Patient with hidradenitis suppurativa after surgical excision of the lesions in the inguinal area and perineum.

that viral hepatitis C and actinomycosis are the results of multiple surgical interventions. Secondly, this clinical case demonstrates the importance of bacterial seeding from lesions.

Overall, actinomycetes are rare microorganisms that can significantly aggravate the course of HS. Despite contradictory data on the involvement of bacterial agents in the pathogenesis of HS, identifying and eliminating concomitant bacterial flora may significantly increase the chances of successful and effective therapy and help achieve long-term remission. Hence, in clinical cases such as the one described, appropriate antibacterial treatment should be administered before or in parallel with biological drugs.

Ethics

The study was approved by the ethics committee of Sokolovskaya Asya and Bocharova Valentina (Russia). (No_271). Authors state that verbal informed consent by the patients was obtained.

Author Contribution

Study concept and design: Khryanin A.A. Analysis and interpretation of data: Sokolovskaya A.V., Bocharova V.K. Drafting of the manuscript: Khryanin A.A., Bocharova V.K. Critical revision of the manuscript for important intellectual content: Khryanin A.A., Sokolovskaya A.V.

Conflict of interest: None declared.

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