Rare Case of Hidradenitis Suppurativa of the Vulva in a Retroviral Positive Postmenopausal Nigerian Woman: A Diagnostic Challenge and Review of the Literature

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Abstract

Hidradenitis suppurativa (HS) is a painful and chronic condition commonly occurring in women in the reproductive age group. A 53-year para 5^{+0}, 3years postmenopausal retroviral positive lady on highly active antiretroviral therapy (HAART) presented to our clinic with a 3-month history of a discharging ulcer on the left labium majus with ipsilateral non-tender inguinal lymphadenopathy. Surgical excision biopsy of the lesion was done and histopathological examination of excised tissue showed features of HS. The patient was treated with oral antibiotics, oral steroids and anti-inflammatory medications. Following these treatment interventions, the lesion healed well and had no recurrence over 2years of follow-up.

Keywords: Hidradenitis Suppurativa, postmenopausal women, excision biopsy.

Introduction

Hidradenitis suppurativa (HS) is a chronic suppurative and inflammatory disorder of the terminal follicular epithelium in the apocrine gland–bearing parts of the skin such as the axilla, groin, perianal and infra-mammary areas and has a pronounced impact on patients.[1,2] It is rare in healthy individuals, pre-pubertal and postmenopausal woman,[3] but three times more commonly seen in females compared to males.[1,3,4] Epidemiologic studies reported varying HS prevalence rates from 0.1% in the US to 0.8% in the Danish population, based on heterogenous measurement methods. However, a recent meta-analysis gave an overall prevalence of 0.4%. The highest prevalence rate of around 4% is found in young adults between 20-40 years. coinciding with increase in sex hormones and women of childbearing age.[1,5,6] This rate declines in older patients.[2] Though it has been reported that it is rarely seen before puberty and in the postmenopausal woman,[4] one patient survey has shown variable effects after menopause. In that study, while 39.5% of participants after menopause reported worsening of their HS symptoms, 44.2% had no change.[6]

Though it is a clinically disabling condition, it is rarely fatal except when it progresses to overwhelming systemic infection in an immune-compromised patient.[1]

The treatment of HS is complex. The treatment modalities include medical therapies such as topical and systemic antibiotics, topical antiseptics, systemic and intra-lesional corticosteroids, systemic retinoids, anti-androgen drugs and hormonal manipulations. Severe HS may require invasive surgical excision of the involved tissue and radical therapy with good outcome. Recently, biologics such as adalimumbab have been advocated.[1,2,7,8]

HS is a rare condition, especially in postmenopausal women. There are few reported cases of HS in postmenopausal women in the literature and rarely from our locality.

Case Report

A 53-year-old para 5^{+0} (5 Alive) lady who was 3 years postmenopausal and HIV positive was referred to our gynaecological clinic with a 3-month history of swelling in the left labium majus. The swelling was insidious and gradually increased in size. It was associated with pain, ulceration with a yellowish discharge which was non-fowl smelling. The left inguinal lymph nodes were enlarged but not tender.

She was a known hypertensive patient but not diabetic. She had a cervical cytology (Pap smear) test done two years prior to her presentation and was normal. She had no history of postmenopausal vaginal bleeding. There was no evidence of weight loss and she had no cough.

General examination revealed a healthy-looking lady. She was 1.60 meters tall and weighed 78kg with a body mass index (BMI)= 30.5kg/m^2. The left inguinal lymph nodes were enlarged, discrete and were not tender. Her blood pressure was 150/95mmHg. Other systemic examinations were normal. Pelvic examination revealed healthy looking clitoris, right labia majus and minus. There was ulcerative, hard, nodular and non-tender mass of about 20 by 20 mm in the lower third of the left labium majus. The vaginal capacity was normal and there were no nodular masses. The cervix was healthy looking. The uterus was approximately 8 weeks gestation size and both adnexa were free.

The initial clinical diagnosis, based on the clinical presentation and examinations, was chronic indurated vulvar ulcer secondary to syphilis to rule out vulvar malignancy. Other differential diagnosis included lymphogranuloma venereum, chancroid, donovanosis, aphthous ulcers and tuberculosis. Thus, local excision with a wide margin (excision biopsy) became necessary.

Laboratory investigations done included Full blood count which showed haemoglobin concentration of 13.3g/dl, total white blood cell count of 6,900cells / mm^3 with granulocyte concentration of 42.1%, lymphocyte concentration of 49.9% and platelet count of 264,000 cell/mm^3. The results of urinalysis, Liver function test, electrolyte, urea and creatinine levels were all within normal values. Also, the hepatitis B surface antigen, hepatitis C and VDRL tests were non-reactive. There was no microbial growth on microbiological study of the ulcer. CD4 Count was 452cells/ mm^3 and the viral load was <20copies/mm. The chest x-ray done revealed mild cardiomegaly (CTR = 54%) with mild unfolding of the aorta. There was no active lung parenchymal disease and the surrounding bony thorax was normal. Abdominopelvic ultrasonography equally revealed normal findings.

Following cardiologist's review and appropriate counseling, she subsequently had wide local excision biopsy of the vulval lesion with 2 cm free margins. The histopathological report on the specimen revealed "focus of ulceration in the epidermis with extensive areas infiltrated by a mixed population of lymphocytes, plasma cells and neutrophils underlying the dermis. Skin adnexia, hair follicles and apocrine glands are surrounded by these cells with obvious luminal contents in the apocrine glands. There is mild acanthosis of the epidermis. There was no evidence of malignancy. Features are those of Hidradenitis Suppurativa.

The histopathological figures are shown below [Figure 1 to 10]:

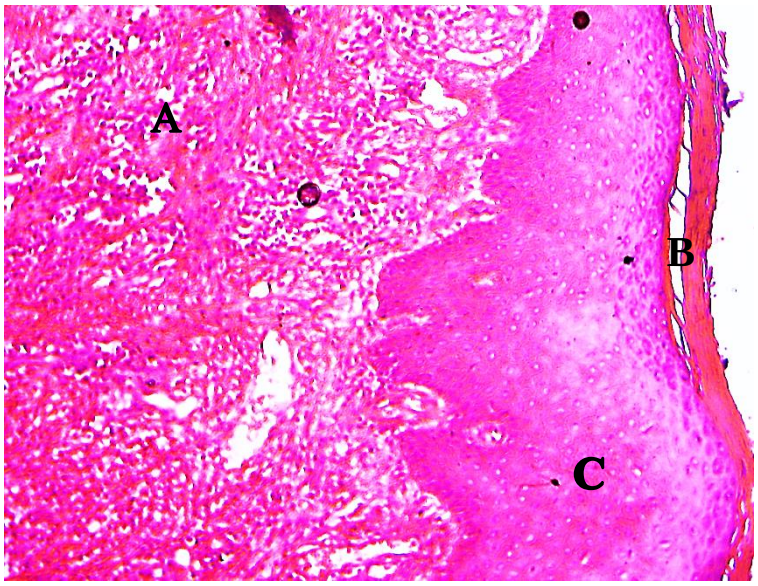

Figure 1: Hematoxylin and eosin, magnification = 100 ×.

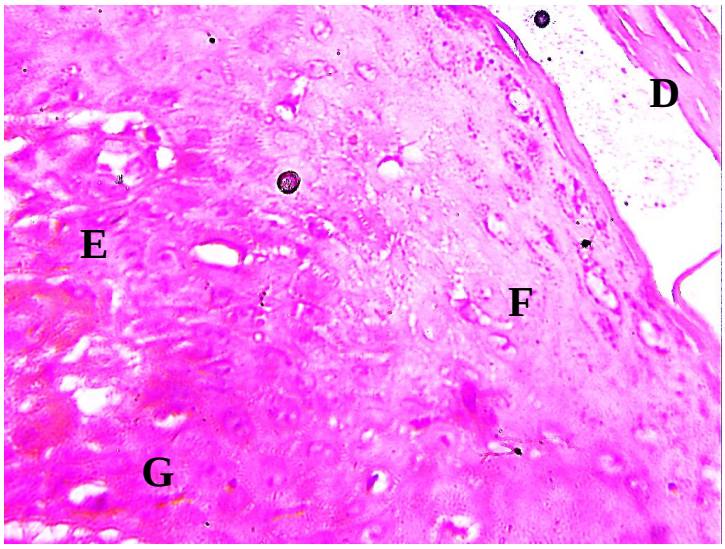

Figure 2: Hematoxylin and eosin, magnification = 400 ×.

Sections of the skin showing the dermis, A, stratum corneum, B, and epidermis, C. the dermal lesion reveals tubular and acini structures lined by epithelia cells and outer myoepithelia cells. D, E, F and G is a higher magnification of the epidermis.

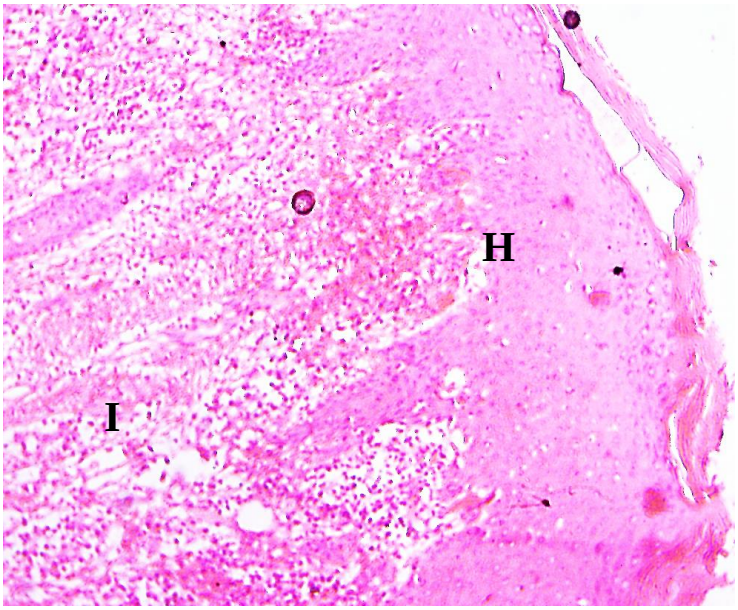

Figure 3: Hematoxylin and eosin, magnification = 100 ×.

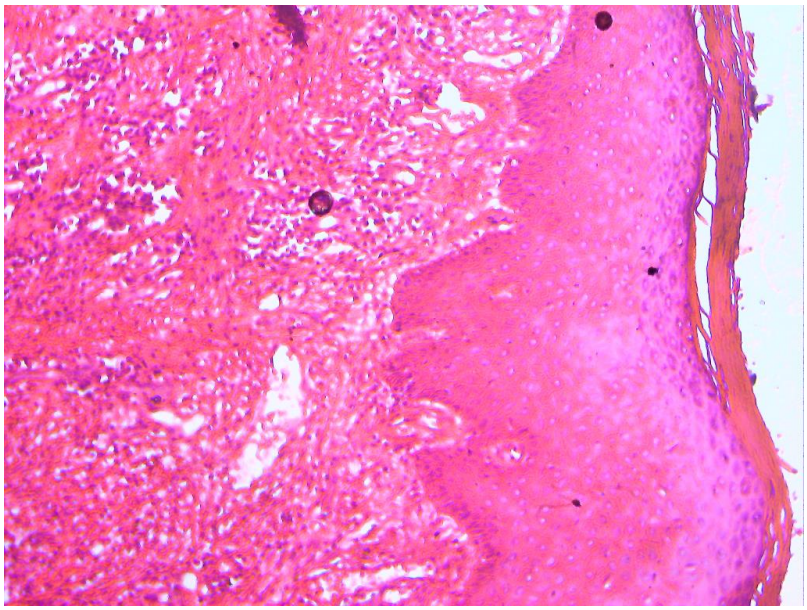

Figure 4: Hematoxylin and eosin, magnification = 400 ×.

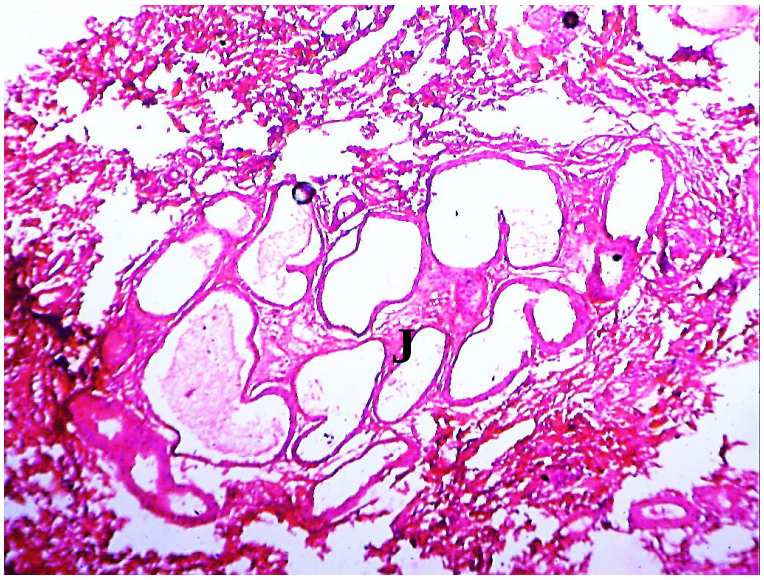

Figure 5: Hematoxylin and eosin, magnification = 100 ×.

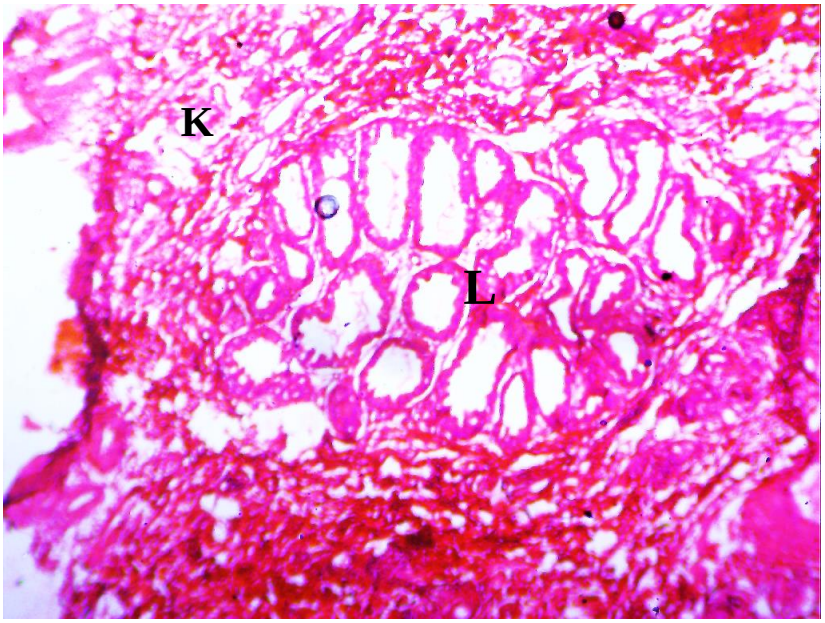

Figure 6: Hematoxylin and eosin, magnification = 100 ×.

Sections of the skin showing tubular and acini structures lined by epithelia cells and outer myoepithelia cells (L, O and J). The lesion is disposed within a stroma of connective tissue (K), in which are blood vessels

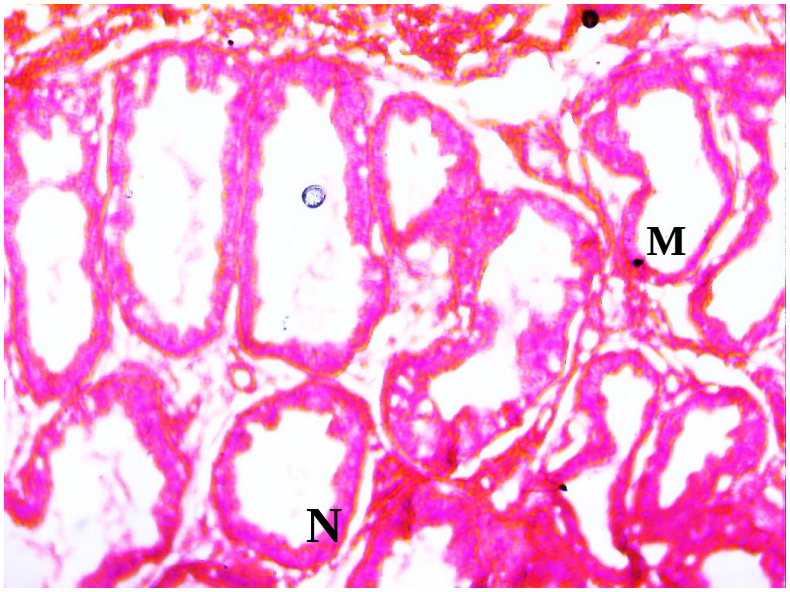

Figure 7: Hematoxylin and eosin, magnification = 400 ×.

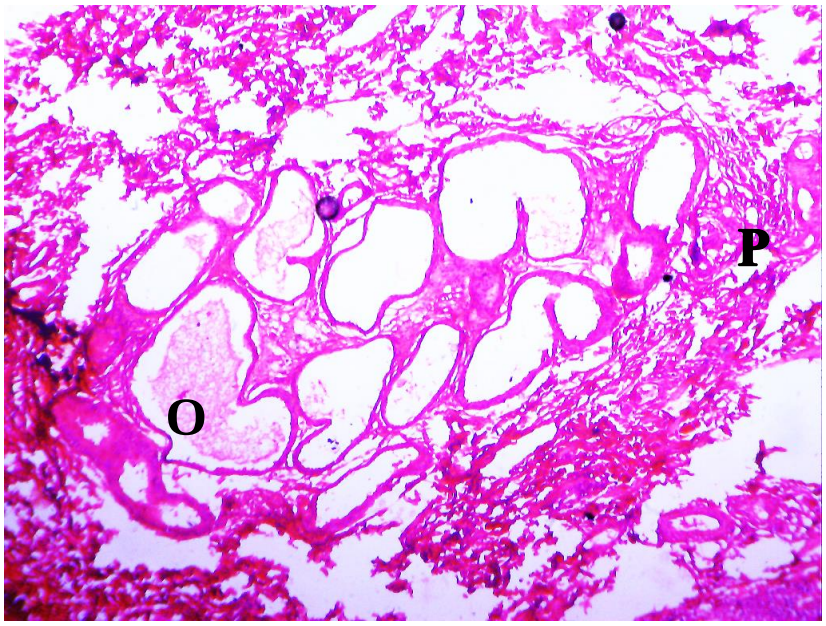

Figure 8: Hematoxylin and eosin, magnification = 100 ×.

Figures 7 and 8 revealing a higher magnification of the lesion showing cystlike structures disposed in cluster and separated by connective tissue stroma. The glandular lesions are lined by epithelia cells with myoepithelia cells disposed to the periphery. Eosinophilic fluid is present in some of the acini, O.

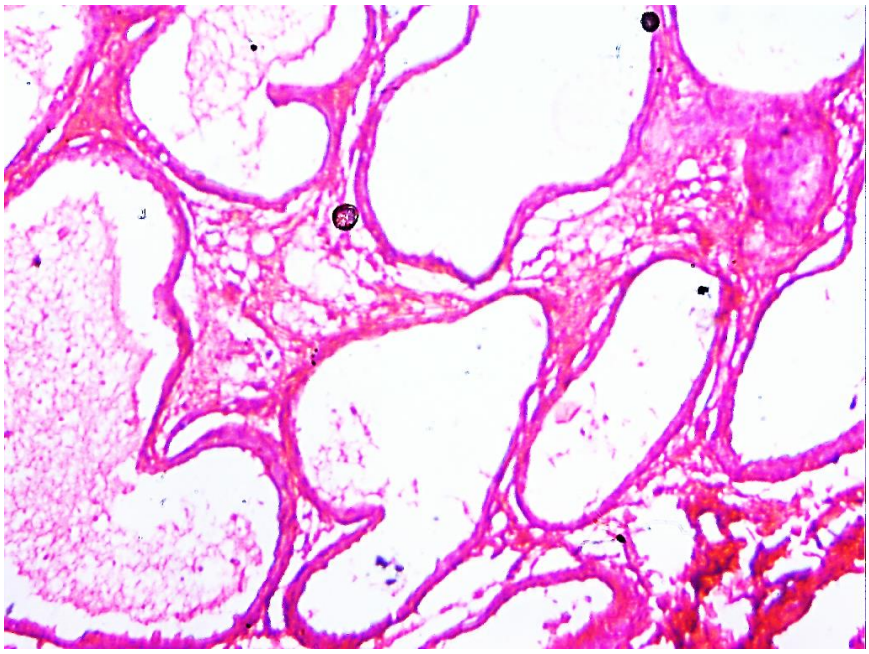

Figure 9: Hematoxylin and eosin, magnification = 100 ×.

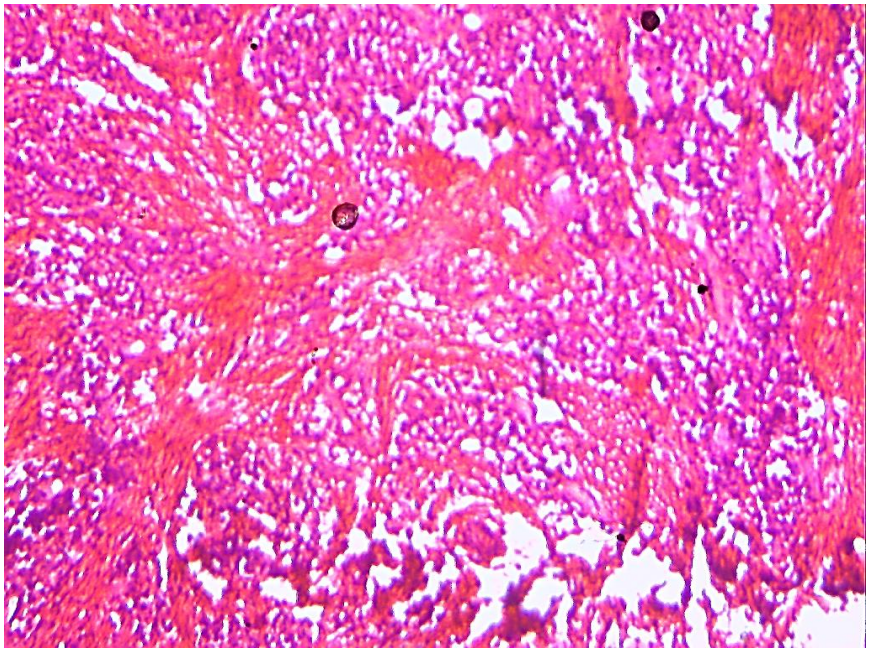

Figure 10: Hematoxylin and eosin, magnification = 100 ×.

Thus, our initial clinical suspicions of syphilitic chancre or vulvar malignancy were ruled out. Patient was counselled on the histopathological findings along with possible modes of further management. She received systemic antibiotics: intravenous cefuroxime 750gm 8-hourly for 72 hours and thereafter converted to oral cefuroxime 500gm 12-hourly for 10 days and metronidazole 400mg 8-hourly for 10days; steroids: tablet prednisolone 10mg 12-hourly for 5days. This was later scaled down to 5mg 12-hourly for 5 days and finally 5mg daily for another 5days and anti-inflammatory therapy (tablet diclofenac sodium 50mg 12-hourly for 7 days), along with her antiretroviral and antihypertensive medications. On discharge and review of her histopathological report, antibiotics were modified to metronidazole, clindamycin and rifampicin for 16 weeks. There was good clinical response and she had no recurrence of the vulvar lesion during a two-year period of follow-up.

Discussion

Hidradenitis Suppurativa is a chronic inflammatory and debilitating disease affecting the apocrine gland - bearing skin and causes painful abscesses and nodules that can eventually progress to interconnected sinus tracts, scarring and contractures of the skin.[9]

The aetiopathogenesis of hidradenitis suppurativa remains unclear. Some studies have shown an association between hidradenitis suppurativa and obesity, genetic predisposition, cigarette smoking and family tendency[1]. However, there was no positive history of these in our patient. Others have shown sex predilection.[10] Available reports show that it is a multifocal disease where lifestyle, immunological processes, genetics and hormonal predisposition may promote follicular hyperkeratosis, dilatation and rupture leading to the development of chronic tissue inflammation.[11] Moreso, it is also reported that atrophy of the sebaceous glands is followed by an early lymphocytic inflammation and hyperkeratosis of the pilosebaceous unit, which eventually leads to hair follicle destruction and granuloma formation, with secondary superimposed bacterial infections.[12] It is commoner in women in the reproductive age group. However, in the face of an immune-compromised condition, hidradenitis suppurativa may occur at the extremes of age,[1] as exemplified in this case and as reported by Macca et at that people with HIV are particularly susceptible to developing HS with the characteristic involvement of atypical sites, such as face or thigh due to HIV-related immunosuppression.[13] Similarly, Deng et al. found a six-fold higher rate of HS diagnosis in patients with HIV compared with those without HIV.[14]

The diagnosis is mainly clinical. The hallmark of hidradenitis suppurativa is inflammation of the apocrine gland-bearing regions causing painful boils. The severity is determined by the degree to which the lesions progress to abscesses, sinus tracts and scarring and is based on Herley's category 1-3.[15] In this postmenopausal woman where hidradenitis suppurativa presented as a hard, ulcerated, painful vulvar nodule, the clinical condition posed a diagnostic challenge. Hence, in such atypical clinical scenario, bacteriological studies and biopsy of the suspected lesion was resorted to, for accurate diagnosis of hidradenitis suppurativa. Hence, excision biopsy with wide free lesion margin was done for this patient based on the suspicion of malignancy amidst other differentials.

Differential diagnosis of this clinical condition depends on the anatomic location. In the vulva, these include, vulvar carbuncles, syphilitic ulcer, lymphogranuloma venereum, Crohn's disease, vulvar acne, keloidalis nuchae cyst and vulvar malignancy.

The treatment modalities include life style modifications, medical, surgical and radiotherapy approaches depending on the stage of the disease. Weight reduction, reduction of alcohol consumption and avoiding rubbing of affected skin are important life style modifications.[9] There are various modalities of medical management which include topical, systemic and intralesional therapies using anti-inflammatory, antibiotics, steroids in addition to anti-androgens. These have been shown to improve the quality of life and reduce the recurrence of symptoms.[9] The choice of medical option in an individual case is influenced by stage of the disease, the clinical experience of the managing physician, availability of the various treatment modalities as well as available results in case series.[9] Recent available reports have advocated the use of systemic immunosuppressive agents for patients with severe form of the disease, with a favorable outcome. Surgery is reserved for patient with poor response to medical treatment and/or patients with late-stage disease or extensive scarring, with little or minimal option of incision and drainage due to high rate of recurrence.[9] Our patient had systemic antibiotics, steroids and anti-inflammatory agents following surgical excision biopsy, based on diagnostic dilemma, with good clinical response without recurrence within two years of follow up without resorting to immunosuppression. This probably shows that surgical excision is superior to simple incision and drainage in the care of such patients.

Conclusion

Hidradenitis Suppurativa is a chronic skin lesion that may pose a lot of diagnostic dilemmas in low resource settings and requires a multi-modal approach especially when it occurs in immune-compromised postmenopausal women. Though it is commoner in women in the reproductive age group, in the face of an immune-compromised condition, hidradenitis suppurativa may occur at the extremes of age, as exemplified in this case and as previously reported by other workers.

Conflict of interest

The Authors declare that there are no conflicts of interest

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