Lymphogranuloma Venereum with a Persistent Genital Ulcer and Lymphadenopathy as the only Symptoms

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Abstract

Genital ulcers can be caused by many infections. This case report describes a patient with a persistent genital ulcer and lymphadenopathy. Although the patient had no other symptoms, and urine samples and rectal swabs were negative for Chlamydia trachomatis (CT), lymphogranuloma venereum (LGV) was diagnosed as the cause of his genital ulcer. Not recognizing LGV infection as a cause of a genital ulcer can have severe consequences for both public health, by allowing further transmission of the infection, as for the affected individual by resulting in clinical complications.

Keywords: Lymphogranuloma venereum; LGV; Genital ulcer; Lymphadenopathy; HIV positive; Chlamydia trachomatis

Introduction

Genital ulcers can be caused by bacterial and viral sexually transmitted infections (STIs), including Treponema pallidum (primary syphilis), Haemophilus ducreyi (chancreoid), Klebsiella granulomatis (granuloma inguinale), Chlamydia trachomatis (lymphogranuloma venereum) and Herpes simplex virus. Although the patient had no other symptoms, and urine samples and rectal swabs were negative for Chlamydia trachomatis (CT), lymphogranuloma venereum (LGV) was diagnosed as the cause of his genital ulcer. Not recognizing LGV infection as a cause of a genital ulcer can have severe consequences for both public health, by allowing further transmission of the infection, as for the affected individual by resulting in clinical complications.

Case Report

A 33-year-old HIV positive man presented to our dermatology clinic with a penile lesion. The lesion had been present for two months and was painful and enlarged despite therapy with locally applied miconazole cream and fusidic acid cream. The patient reported regular anonymous unprotected receptive and insertive oral and anal sex with men; his last sexual contact had been some days prior to visiting our clinic. He did not report fever, chills, fatigue, rash, urethral discharge, dysuria, or rectal symptoms.

The patient had been receiving a fixed-dose combination of emtricitabine, tenofovir, elvitegravir and cobicistat during the past 1.5 years to treat HIV infection. The HIV infection was well-controlled and there were no symptoms associated with immunodeficiency; the HIV RNA level (viral load) in plasma was undetectable and a recent CD4+ T-cell count was 1014 cells/mm3. The patient's medical history further included rectal gonorrhoea, rectal and urethral chlamydial and anogenital warts.

On physical examination, an ulcer measuring 2 mm in diameter was present at the coronal sulcus on a pre-existing adhesion between the prepuce and glans (Figure 1). The lesion was slightly indurated and painful to palpation. There was unilateral inguinal lymphadenopathy on the left side, without fluctuation. The remainder of the physical examination was unremarkable. Swabs were collected from the penile lesion and sent for routine bacterial culture and, for Herpes simplex virus (HSV) and Treponema pallidum polymerase chain reaction (PCR). Furthermore a urine sample was collected for Neisseria gonorrhoea (GO) and Chlamydia trachomatis (CT) PCR, and a serological test was performed to determine the presence of Treponema pallidum Ig. All test results were negative.
On the second visit, 5 days later, the genital ulcer and lymphadenopathy still persisted. Therefore new penile ulcer swabs were collected and tested for Varicella zoster virus and CT using real-time PCR. The penile ulcer swab tested negative for HSV and Varicella zoster virus, but positive for CT. The CT-positive swab was further tested using an in-house pmpH real-time PCR to differentiate between an LGV and non-LGV type infection. This assay confirmed the swab to be an LGV type infection. Using high resolution multilocus sequence typing (hr-MLST) we were able to determine the CT genovar strain, which was L2b. This is known to be the most common LGV type [4]. In addition, a rectal swab was collected for CT and GO testing, but were both negative. The urine sample for CT was repeated, but remained negative. The patient was treated with doxycycline 100 mg twice daily for 21 days. On clinical follow-up, two weeks after the completion of doxycycline treatment, the patient reported no pain, and the penile ulcer had resolved completely and there was no inguinal lymphadenopathy.

Discussion

This patient presenting with a persistent and painful genital ulcer was diagnosed with inguinal LGV. LGV is rare and can cause ulceration, but mostly anorectal. The patient had no other clinical manifestations of CT such as urethritis or proctitis. Although there have been reports of LGV infections in Europe presenting with genital ulcer disease in MSM, most LGV infections in MSM are associated with proctitis. De Vrieze et al. found 1.2% anorectal LGV positivity and 0.6% inguinal LGV positivity among MSM in the Netherlands [5]. In a UK-study Ward et al. described a prevalence of LGV in MSM of 0.90% in the rectum and 0.04% in the urethra [6]. This underscores that the mode of transmission of LGV within the MSM community still remains enigmatic. Perhaps the paucity of penile lesions caused by LGV infections is due to failing to recognise cases in insertive partners, causing these missed LGV infections to contribute to further transmission of LGV. Recently an alternative gastro-intestinal transmission route via ano-oral contact has been suggested [7]. Unfortunately, in our case we could not trace any sexual partners to establish the source of infection. In addition, the painful genital ulcer persisted for a total period of three months, an unusually long period. The patient, although HIV infected, had normal CD4 counts and no signs of immunodeficiency.

Conclusion

This case demonstrates that, although rare, LGV infections should be considered as a possibility in the differential diagnosis of persistent painful genital ulcer disease, especially in MSM and patients presenting with lymphadenopathy, even when immunocompetent. And if urine samples and rectal swabs are negative for CT, a diagnosis of LGV infection should not be rejected as a cause of a genital ulcer. The primary lesion, usually a small and painless papule or ulcer, may remain subclinical. Not recognising LGV infection as a cause of a genital ulcer can have severe consequences for both public health, by allowing further transmission of the infection, as for the affected individual by resulting in clinical complications, such as permanent local oedema, phimosis, lymphatic fibrosis, lymphogranulomatous infiltrate and elephantiasis of the genitalia. Therefore testing for LGV infection should be considered in the diagnostic work-up of patients with genital ulcers, even if other clinical manifestations of CT infections are absent, and in particular when syphilis and other microbial causes (STIs) have been excluded.

References


Figure 1: Ulcer measuring 2 mm in diameter at the coronal sulcus on a pre-existing adhesion between the prepuce and glans.