

# A suspected case of lymphogranuloma venereum (LGV) suggests underdiagnosed LGV infection among Slovenian men who have sex with men

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## Abstract

Lymphogranuloma venereum (LGV) is sexually transmitted infection caused by serovars of *Chlamydia trachomatis*, mostly seen among HIV-positive men who have sex with men. The first three reports of possible LGV in Slovenia were from April to June 2015, followed by a confirmed case of LGV in August 2015. We present the case of an HIV-positive MSM that presented with an anorectal abscess, discharge, lymphadenopathy, and unusual perianal plaque. Gonococcal proctitis was assumed and he received empirical antibiotic treatment, after which only intermittent improvement occurred. After a positive test result for chlamydial infection, but without a response to azithromycin treatment, LGV was suspected. Treatment according to the guidelines was introduced. When doxycycline therapy started, rapid improvement was observed, and it was therefore assumed that the LGV infection had been successfully treated. Two similar cases with an unusual anorectal presentation and an excellent response to antibiotic therapy for LGV were observed at the same center shortly thereafter. While pointing out possible delays and limitations in diagnostic procedures at self-pay facilities, the need for better access to high quality STI management in public and in private services is emphasized. Enhanced surveillance and testing guidelines could reveal a hidden LGV epidemic among MSM in Slovenia.

**Keywords:** dermatology, proctology, lymphogranuloma venereum, MSM, private practice limitations

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## Introduction

Lymphogranuloma venereum (LGV) is a sexually transmitted infection (STI) caused by L1, L2, and L3 serovars of *Chlamydia trachomatis* (CT), and in most cases in developed and European countries it is seen among white HIV-positive men who have sex with men (MSM) (1). Patients commonly present with symptoms of proctitis (i.e., rectal pain, discharge, bloody stools, constipation, and tenesmus) (1), although reports from the UK, the Netherlands, and Germany show that approximately one-third of LGV cases are asymptomatic (2).

Since 2003, when an LGV outbreak among MSM was first reported in the Netherlands, (3) LGV has spread across other western European countries (4, 5). The first laboratory-confirmed case of LGV in Slovenia was diagnosed at the STI outpatient clinic at the Clinic for Infectious Diseases and Febrile Illnesses at the Ljubljana University Medical Center and was reported to the National Institute of Public Health (NIPH) in August 2015 (6). In addition, three possible LGV cases among HIV-positive MSM were reported to the NIPH in 2015, all treated at the private clinic Zdrav Splet for self-pay patients from April to June 2015.

## Case report

A 39-year-old HIV-positive MSM, who was not on anti-retroviral therapy (ART), was referred to our clinic by an infectologist. He had had one unprotected episode of receptive anal intercourse with one unknown HIV-positive partner 2 months earlier, and the first signs of anal discomfort were noticed 2 weeks after sexual intercourse.

At the first visit he underwent an urgent operation for a perianal abscess on the right side of the anal verge. On his left perianal region he had an unusual condylomatous plaque (Fig. 1) and bilateral inguinal lymphadenopathy was noted. Drainage of the abscess was performed and the pus was sent to the National

Laboratory for Health, Environment, and Food in Maribor for a PCR test for CT and *Neisseria gonorrhoeae* (NG) and also for isolation and culture of NG. Suspecting a gonococcal infection, we started treatment with 2 g of azithromycin orally. A rapid blood test for syphilis, an immunoassay detecting antibodies against *Treponema pallidum* (TP) (Biomerieux, France), was negative, and testing for other STIs was refused by the patient. We received the microbiology results after a few days and they were positive for CT and negative for NG. The patient did not consent to paying for suggested microbiological examinations for LGV.



**Figure 1** | Perianal abscess on the right side and condylomatous plaque on the left side, with a skin tag on the anal verge (source: B. Mlakar).

At the first follow-up after 14 days, the patient noticed only minor improvement (Fig. 2), probably mostly because the perianal abscess was drained. During a proctoscopy we found inflamed rectal mucosa with a whitish-yellow discharge, and we decided to treat him as an LGV case. We prescribed doxycycline 100 mg orally twice daily to him for 21 days in line with

the 2013 European guidelines on the management of LGV (7). We also recommended that the infectologist start ART prior to surgical removal of the perianal condylomatous plaque.



**Figure 2** | Hyperpigmented perianal condylomatous plaque, a skin tag on the anal verge, and presence of purulent and bloody anal discharge (source: B. Mlakar).

At the second follow-up visit, 4 weeks later, the patient was almost asymptomatic, and he reported no pain, normal bowel movements, and no anal discharge. However, during the proctoscopy we found a white coating on the rectal mucosa, and therefore we repeated PCR tests for NG and CT, and also for other pathogen bacteria and fungi. Only *Candida albicans* was positive, and fluconazole capsules 100 mg twice daily for 14 days were prescribed. Three weeks later, the rectal mucosa were normal at proctoscopy and the perianal plaque had also almost disappeared. The condylomatous regression was probably a result of the HIV therapy he had started 6 weeks earlier. We decided to use cryotherapy to remove the remains of this plaque instead of surgery. Later on, the patient was without signs and symptoms of an STI.

In the short period of 3 months, two additional MSM patients visited our clinic with complaints of anal pain, discharge, and lymphadenopathy. The first patient was HIV-positive and was receiving ART. In the second patient, coinfections with HIV and syphilis were discovered. The syphilis was treated at our clinic, and he was referred to an infectologist for induction of ART. Both had had anal intercourse within 1 month prior to examination. The examination revealed anal discharge and mucosal inflammation. We assumed gonococcal proctitis, and rectal swabs for PCR and culture for NG were taken; both patients received immediate antibiotic therapy with azithromycin and ceftriaxone. There was only mild clinical improvement and the microbiological results for NG were negative. They were offered additional testing for CT and LGV, but both of them, because they were self-pay pa-

tients, refused. Because of the persistence of lymphadenopathy and discharge, they were empirically treated with doxycycline for probable LGV infection. After the treatment, both had no further anorectal symptoms and signs, and their lymph nodes reduced to normal size.

## Discussion

We report on a possible LGV case suspected in an HIV-positive MSM that presented with common clinical manifestations for LGV and responded well to the treatment for LGV. A laboratory test for CT was performed and was found to be positive, but LGV was not laboratory-confirmed because the self-pay patient did not consent to paying for suggested additional microbiological examinations. Because the anorectal signs and symptoms were a common clinical manifestation of LGV and reacted only to antibiotic therapy, as prescribed according to the 2013 European guideline on the management of LGV (7), we concluded that LGV infection was probable and had been treated successfully. In addition, two additional suspected cases presented and were successfully treated at our clinic as LGV, even though LGV infection was not confirmed. All three cases were reported to NIPH as probable LGV in 2015. That same year, the only laboratory-confirmed and first proven case of LGV in Slovenia was reported from a public STI outpatient clinic (6).

Our report shows how long the diagnostic and therapeutic process can be when there are limitations on microbiological diagnosis because self-pay patients with signs and symptoms of STI do not consent to paying for testing for all of the most common STIs. Because our outpatient facility is known to be MSM-friendly, patients often do not want to be referred to the public outpatient STI clinics, where appropriate diagnosis, including necessary microbiological examinations, and relevant treatment can be provided and paid for within mandatory health insurance coverage.

For better access to high-quality STI case management, access to public and private specialized STI outpatient services without a referral from a primary healthcare level should be considered when the future sexual and reproductive national health strategy is prepared. Moreover, some reports of one-third of LGV cases being asymptomatic (2) should concern us, along with the fact that in most cases the sexual partners of infected patients remain unidentified. Therefore, enhanced LGV surveillance, together with national LGV testing guidelines, might reveal a hidden LGV epidemic among MSM in Slovenia.

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## References

1. Hamill M, Benn P, Carder C, Copas A, Ward H, Ison C, et al. The clinical manifestations of anorectal infection with lymphogranuloma venereum (LGV) versus non-LGV strains of *Chlamydia trachomatis*: a case-control study in homosexual men. *Int J STD AIDS*. 2007;18:472-5.
2. Saxon C, Hughes G, Ison C, UK LGV Case-Finding Group. Asymptomatic lymphogranuloma venereum in men who have sex with men, United Kingdom. *Emerg Infect Dis*. 2016;22:112-6.
3. Goetz H, Nieuwenhuis R, Ossewaarde T, Thio B, van der Meijden W, Dees J. Preliminary report of an outbreak of lymphogranuloma venereum in homosexual men in the Netherlands, with implications for other countries in western Europe. *Euro Surveill*. 2004;8:2367.
4. Savage EJ, van de Laar MJ, Gallay A, van der Sande M, Hamouda O, Sasse A, et al, on behalf of the European Surveillance of Sexually Transmitted Infections (ESSTI) network. Lymphogranuloma venereum in Europe, 2003–2008. *Euro Surveill*. 2009;14:19428.

5. Sary G, Sary A. Lymphogranuloma venereum outbreak in Europe. *J Dtsch Dermatol Ges.* 2008;6:935-40.
6. Matičič M, Klavs I, Videčnik Zorman J, Vidmar Vovko D, Kogoj R, Keše D. Confirmed inguinal lymphogranuloma venereum genovar L2c in a man who had sex with men, Slovenia, 2015. *Euro Surveill.* 2016;21:30129.
7. De Vries HJC, Zingoni A, Kreuter A, Moi H, White JA. 2013 European guideline on the management of lymphogranuloma venereum. *J Eur Acad Dermatol Venereol.* 2015;29:1-6.