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Chronic Lymphogranuloma Venereum Proctocolitis Masquerading as Inflammatory Bowel Disease

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Abstract

Rectal infection with the L1, L2, and L3 serovars of *Chlamydia trachomatis* can cause lymphogranuloma venereum (LGV) proctocolitis, particularly among men who have sex with men (MSM). Symptoms of this sexually transmitted infection include anal pain, rectal bleeding and discharge, tenesmus, constipation, and fever. Clinicians should consider LGV when there is a history of receptive anal intercourse and symptoms of proctocolitis. A positive nucleic acid amplification test (NAAT) on a rectal sample is diagnostic. This report describes a man with HIV and chronic proctocolitis in whom the diagnosis of LGV was delayed because the clinical picture mimicked inflammatory bowel disease.

Keywords

lymphogranuloma venereum, proctocolitis, inflammatory bowel disease, nucleic acid amplification test

Introduction

Lymphogranuloma venereum (LGV), a sexually transmitted infection caused by the L1, L2, and L3 serovars of *Chlamydia. trachomatis*, is best recognized as a cause of femoral or inguinal lymphadenopathy, sometimes with a genital ulcer or papule at the site of inoculation.[1] The disease occurs most frequently in resource-limited tropical and subtropical areas of the world.[2] However, since 2003, there have been outbreaks of LGV causing hemorrhagic proctocolitis among men who have sex with men (MSM), many of whom are co-infected with HIV, in resource-rich settings, including Europe and North America.[3-6] Symptoms reflect inflammation of the rectum often without femoral and inguinal lymphadenopathy. Endoscopic findings are nonspecific but include ulcers, mucosal friability, masses, or polyps, and mucopurulent exudate.[5] The atypical clinical presentation, unawareness of clinicians and patients regarding the disease, and lack of a routine diagnostic test for LGV serovars of *C. trachomatis* have contributed to a delay in diagnosis and have caused misdiagnosis. Left untreated, rectal infection with the L1, L2, and L3 serovars can lead to rectal fistulae and strictures.[1]

Case Presentation

A 38-year-old man with HIV and chronic constipation presented with a 6-month history of rectal bleeding, discharge, fecal urgency, and tenesmus. He denied fever, chills, night sweats, and weight loss. HIV infection was diagnosed 7 years earlier, at which time the absolute CD4-T-lymphocyte count was 425 cells/μL (normal 720-1348 cells/μL). Antiretroviral therapy was initiated immediately. Medication adherence was excellent, and the HIV viral load was consistently undetectable during routine follow-up. His most recent CD4 count was normal. Past medical history was significant for latent syphilis of unknown duration, treated appropriately with benzathine penicillin about 6 months earlier. He has sex with other men, including receptive anal intercourse, but he did not have any sexual contact over the preceding last year. Family history is significant for colon cancer in maternal uncles and maternal grandmother.

Colonoscopy showed multiple shallow, irregular, non-bleeding ulcers, and excavated lesions in rectum and anus. Multiple cold forceps biopsies were performed for histology. A single sessile polyp in the sigmoid colon was removed piece-meal. Histopathology of the rectal ulcer showed moderately active colitis with ulceration and non-necrotizing granulomas. The sigmoid polyp showed the same. No

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organisms were seen with Gomori methenamine silver (GMS), periodic acid-Schiff (PAS), and acid-fast stains. Immunohistochemical stains for cytomegalovirus (CMV), herpes simplex virus (HSV), and spirochetes were negative as well.

Mesalamine enemas were prescribed for the provisional diagnosis of inflammatory bowel disease (IBD). Despite this therapy, he continued to have rectal bleeding, foul-smelling rectal discharge, and fecal urgency over the next 6 months. At a follow-up appointment in the infectious diseases clinic, we obtained a rectal swab for *C. trachomatis* testing by transcription-mediated amplification (TMA), a type of nucleic acid amplification test (NAAT), which resulted positive. TMA for *Neisseria gonorrhoeae* was negative. Rapid plasma reagin was non-reactive. We treated him with doxycycline 100 mg twice a day for 21 days for LGV proctocolitis. His symptoms improved after 1 week and completely resolved by the end of therapy.

Discussion

LGV is predominantly a disease of lymphatics. When the primary site of infection is the external genitalia, which sometimes causes an ulcer or papule, invasion and destruction of lymphatic tissue causes painful, erythematous inguinal, and femoral lymphadenopathy that can suppurate and rupture (buboes).[7] Enlargement of lymph nodes above and below the inguinal ligament causes the characteristic "groove sign." An anorectal syndrome can occur when the rectum or anal canal is the primary site of inoculation. This syndrome presents as hemorrhagic proctocolitis without inguinal and femoral lymphadenopathy because infection proximal to the dentate line drains into deeper lymph nodes in the pelvis.

The clinical diagnosis of LGV proctocolitis is challenging because of the nonspecific clinical presentation, endoscopic appearance, and biopsy findings. *N. gonorrhoeae*, HSV, and syphilis are other sexually transmitted infections that can infect the rectum. Furthermore, as illustrated by our case, LGV and IBD can present similarly. The endoscopic abnormalities in LGV include mucopurulent exudates, friable mucosa, multiple ulcers, erosions, granulation tissue, and inflammatory masses in the rectum and colon.[8] Histopathological findings consist of intense mixed inflammatory infiltrates, crypt abscess, and crypt distortion.[5] Diagnostic confusion can lead to ineffective therapies and inappropriate invasive procedures.[8] When evaluating patients with anorectal symptoms, providers should obtain a careful sexual history and should be aware that both HIV-negative and HIV-positive MSM, as well as heterosexuals who practice receptive anal intercourse, are at risk for LGV proctocolitis.

Commercially available NAATs, including those that use TMA, are highly sensitive and specific for the diagnosis of rectal infections due to *C. trachomatis*.[9] However, these tests, while positive in LGV infections, are unable to differentiate between LGV serovars and non-LGV serovars of *C. trachomatis*. PCR-based methods developed for this specific purpose are not routinely available.[10] In the appropriate epidemiologic setting, the Centers for Disease Control and Prevention (CDC) recommends obtaining a rectal swab from all patients with proctocolitis to test with a commercial NAAT for *C. trachomatis*.[1] A positive result is sufficient for the presumptive diagnosis of LGV. Taking it one step further, even before the test result is available, the CDC recommends presumptively treating patients with risk factors and signs and symptoms of proctocolitis for LGV. The recommended treatment for LGV is doxycycline 100 mg orally twice a day for 21 days.[1] Azithromycin 1 g orally once weekly for 3 weeks is an alternative.

Conclusion

We aim to shed light on similarities between LGV proctocolitis and IBD, as both present as chronic inflammation of lower gastrointestinal tract. Clinical features, endoscopic findings, and histopathologic features cannot differentiate LGV from IBD. History of receptive anal intercourse and failure to respond to IBD treatment were clues to the diagnosis of LGV proctocolitis in our case. A NAAT for *C. trachomatis* from a rectal swab is the diagnostic test of choice. Doxycycline for 3 weeks is the treatment of choice.

Declaration of Conflicting Interests

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Disclosures

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Ethical Approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed Consent

Verbal informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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