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Authors

Promer, Katherine

Pillay, Allan

Chi, Kai-Hua

et al.

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Case Report

A case of inguinal lymphogranuloma venereum imitating malignancy on CT imaging

Katherine Promer, MD^{a,*}, Allan Pillay, PhD^b, Kai-Hua Chi, MS^b, Noushin Vahdat, MD^c, Samantha S. Katz, PhD^b, Cheng Y. Chen, PhD^b, Joshua Fierer, MD^{a,c}

^aUC San Diego Health, CA, USA

^bCenters for Disease Control and Prevention (CDC), Atlanta, GA, USA

^cVA San Diego Healthcare System, CA, USA

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ABSTRACT

Lymphogranuloma venereum is a sexually transmitted infection caused by serovars L1, L2, and L3 of *Chlamydia trachomatis*. We here report a case of Lymphogranuloma venereum, confirmed by PCR testing, which mimicked malignancy on CT imaging.

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Case description

A 63-year-old man who has sex with men with HIV (viral load < 20 copies/mL, CD4⁺ 573/mL) and recurrent balanitis presented to an emergency department (ED) in San Diego, California with right groin pain and swelling. He first noted a penile lesion and right groin swelling 3 weeks prior. Syphilis serology and HSV lesion polymerase chain reaction (PCR) were negative, so symptoms were attributed to recurrent balanitis. The penile lesion diminished with fluconazole, but progressive right groin swelling and development of fever prompted a visit to the ED.

Physical exam revealed a 3 cm tender right inguinal mass and a 0.5 cm macule at the dorsal-proximal penile shaft with-

out evidence of active balanitis. CT (Fig. 1a and b) revealed multiple enhancing, enlarged right inguinal and external iliac lymph nodes, concerning for lymphoma or metastatic or inflammatory adenopathy, so the patient was admitted for further evaluation. The infectious diseases consultant considered lymphogranuloma venereum (LGV), syphilis, and chancroid as possible diagnoses, recommending empiric treatment with doxycycline 100 mg twice daily and one dose of 250 mg intramuscular ceftriaxone. Surgery performed an excisional biopsy leaving a drain in place. Grossly, the mass resembled a purulent lymph node, and histology revealed fibro-adipose tissue with marked acute and chronic inflammation but no visible nodal tissue. Neither Gram stain, Giemsa stain, nor bacterial cultures revealed a causative organism. The patient was discharged to complete 21 days of doxycycline for presumptive LGV treatment and advised to inform his most recent partners of this diagnosis. After he was discharged, the urine molecular test resulted as positive for Chlamydia (Cobas

* Corresponding author.

E-mail address: kpromer@ucsd.edu (K. Promer).

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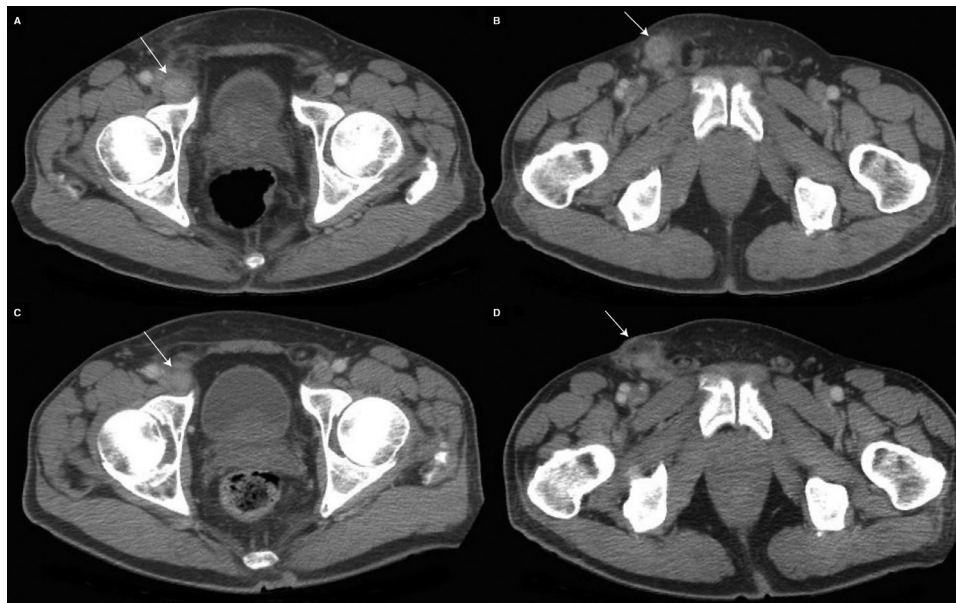


Fig. 1 – Contrast enhanced CT images of the abdomen and pelvis. (A) Axial slice of the initial CT demonstrating 2.1 cm enhancing right external iliac lymph node (arrow). (B) Axial slice of the initial CT demonstrating 2.7 cm enhancing right inguinal lymph node (arrow). (C) Axial slice of the follow-up CT demonstrating decrease in size of external iliac lymph node to 1.7 cm (arrow). (D) Axial slice of the follow-up CT demonstrating small anterior fluid collection at site of biopsy in right inguinal region attributed to post-surgical changes (arrow).

4800 CT/NG test [Roche Molecular Diagnostics, Pleasanton, CA, USA]), further supporting the diagnosis.

Fever and groin pain improved, but on day 10 of doxycycline, he returned to the ED with diffuse abdominal pain. Repeat CT (Fig. 1c and d) demonstrated enhancing right inguinal lymph nodes, smaller but still concerning for lymphoma or metastatic disease. Abdominal pain resolved, and CT findings were attributed again to LGV. The patient was discharged and continued to improve on doxycycline treatment.

A formalin-fixed paraffin-embedded (FFPE) sample and 2 fresh tissue specimens, refrigerated for a week prior to freezing, were shipped on dry ice to the Centers for Disease Control and Prevention for LGV PCR and genotyping. Genomic DNA was extracted using the ChargeSwitch gDNA Mini Tissue Kit (Thermo Fisher Scientific, Waltham, MA, USA) and tested for LGV and non-LGV *C. trachomatis* using a real-time quadriplex PCR assay [1]. All 3 specimens tested positive for LGV. Nested PCR amplification of the *ompA* gene and sequencing of the amplicon using the BigDye Terminator v3.1 Cycle Sequencing Kit and an ABI 3130xl Genetic Analyzer (Applied Biosystems, Foster, CA, USA) [2] identified the L2b-variant.

Discussion

Data on the incidence of LGV in San Diego County had been limited in the years preceding this 2017 case. The 9 reported cases in 2012 and the fewer than 5 reported cases per year between 2013 and 2016 likely underestimate true incidence. Most reported LGV cases in men who has sex with men

present as proctitis with rectal bleeding, tenesmus, and abdominal pain; a genital lesion with tender lymphadenopathy is less common [3].

Several testing options exist for the diagnosis of LGV. Genital, rectal, and lymph node specimens can be tested for *C. trachomatis* by culture, direct immunofluorescence, or nucleic acid amplification tests [4,5]. Commercial direct immunofluorescence kits using antimajor outer member protein antibodies are *C. trachomatis*-specific, while those with antilipopolsaccharide antibodies cross-react with nontrachomatis chlamydia, and nonchlamydial bacteria [6]. Serology may also support the diagnosis of LGV in the appropriate clinical context [4,5]. Lastly, PCR testing on unstained FFPE or fresh tissue can differentiate LGV strains from other serovars of *C. trachomatis* [1,7].

PCR has been used to detect LGV in rectal, bubo aspirate, penile lesion, urethral, and urine specimens [8–10]. An ideal specimen for PCR should be frozen immediately at -80°C or stored only 1–2 hours at 4°C prior to freezing. In this case, genotyping revealed the L2b-variant, the major serovar implicated in the 2003 European LGV outbreak and subsequently in North America [11–13]. Genotyping can be used for molecular epidemiology to establish transmission chains [14]. A series of 5 atypical LGV cases presenting with painful, edematous genital ulceration caused by L2b suggests that this variant is more virulent than other genotypes [15].

Characterization of LGV imaging findings is limited to 3 case reports with CT images depicting cervical or rectal tissue involvement resembling cancer [16–18]. In our case, a correct clinical diagnosis was made by the infectious diseases consultant through history, physical exam, and appropriate

laboratory testing. The surgical biopsy and hospitalization may have all been avoided had management focused on an infectious etiology rather than pursuing further work-up of possible malignancy. This case, however, emphasizes that LGV should be included in the differential diagnosis of unilateral inguinal lymphadenopathy and provides the opportunity to demonstrate the CT appearance of inguinal LGV, which may imitate malignancy.

Conclusion

Imaging is generally not required to diagnose LGV and, as this case illustrates, does not distinguish from lymphoma or metastatic adenopathy. Therefore, particularly in a patient with appropriate risk factors, LGV should be included in the differential diagnosis along with metastasis, lymphoma, and other infectious or inflammatory etiologies.

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Disclaimer

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