CASE REPORT

First case of "bubonulus" in L2 lymphogranuloma venereum

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Since 2003, an ongoing lymphogranuloma venereum (LGV) proctitis outbreak has been reported in industrialised countries with a new variant designated L2b. Only men who have sex with men (MSM) are affected and most are HIV co-infected; delayed or incorrect diagnoses are frequent. We report a rare clinical case of LGV primary stage called "bubonulus" with penile adenopathy and secondary local acute lymphoedema in an MSM seropositive man. This is the first case described of this clinical presentation with a L2b genovar strain, occurring immediately after use of a sex toy. It suggests that this presentation is favoured by host immunity and behavioural factors. These factors may also play a part in the new outbreak of LGV.

Lymphoedema of the external genitalia is a symptom encountered in many acquired conditions include neoplastic, reactive, granulomatous and infectious disorders such as lymphogranuloma venereum (LGV). Since 2003, an ongoing LGV proctitis outbreak has been reported in industrialised countries with a new variant designated L2b. Only men who have sex with men (MSM) are affected and most are HIV co-infected; delayed or incorrect diagnoses are frequent. We report a rare clinical case of an LGV primary stage called "bubonulus" with secondary penile lymphoedema in an MSM seropositive man.

CLINICAL CASE

A 49-year-old MSM, HIV positive patient on HAART (highly active antiretroviral therapy), presented in September 2006 with a painless non-inflammatory brutal oedema of the penis with phimosis. He described three episodes of secondary syphilis in the past years (the last one treated a year ago with benzathine benzylpenicillin 2.4 MU) and gave a history of multiple sexual partners (40 per year) with "serosorting" and inconsistent condom use. The lesions appeared three days after a vigorous episode of "cock ring" use, used for a few hours with an anonymous partner.

Examination found three non-inflammatory centimetric subcutaneous nodular formations along the dorsal face of the penis with a soft, painless, circumferential oedema and several non-purulent ulcerations of the glans (fig 1). There was no scrotal oedema and no inguinal adenopathy. There were no rectal symptom and rectal examination was normal.

The biological assessment showed a CD4+ count at $50 \times 10^6/l$ and an undetectable HIV viral load. *Haemophilus ducreyi*, *Treponema pallidum*, and herpes simplex virus were not identified in ulcer swab using polymerase chain reaction (PCR) and immunofluorescence. *Chlamydia trachomatis* infection was demonstrated by PCR in the ulceration. Serovar L2b was typed by restriction fragment length polymorphism of omp1 gene amplified by polymerase chain reaction (PCR-RFLP genotyping) confirming LGV.

Syphilis serology showed a stable VDRL titre at 32 and *C trachomatis* serology showed high levels of IgG (more than 512)

and positive IgA. A transcutaneous echography with Doppler ultrasound found three non-collected subcutaneous lymph nodes, 5–7 mm, of the dorsal face of the penis without other vascular, pelvic, or urinary tract abnormalities.

The patient was treated with oral doxycycline 200 mg/day for 3 weeks with complete disappearance of ulcerations and lymph nodes at day 15.

DISCUSSION

LGV is a systemic sexually transmitted disease caused by *C trachomatis* strains belonging to the L1, L2, or L3 genotype.

This lymphotropic organism leads to the development of thrombolympangitis/perilymphangitis, and lymphadenitis, immunity of the host limiting the progression without eliminating it. Most of the damage is caused by the immune delayed cellular response, chronic oedema, and lymphatic fibrosis compromising the local vascularisation of the skin or the mucous membrane, leading to the traditional presentation: genital ulceration and buboes. The primary stage of the disease can rarely be associated with a lymphangitis of the dorsal penis, with formation of a large and tender lymphangial nodule classically called "bubonulus".^{1,2} Lymphangitis can be accompanied by local oedema, which may produce phimosis. This is the case with our patient. Later, long standing oedema, fibrosis, and lymphogranulomatous infiltrate can result in elephantiasis with "esthiomene."^{3,4}

A resurgence of LGV has been observed since 2004 in several industrialised countries, becoming an increasingly important public health issue.⁵

This outbreak concerns essentially MSM, with gastrointestinal symptoms—bloody proctitis, purulent discharge, and buboes, with a high rate of HIV seropositivity and other sexually transmitted diseases. Serovar L2b is generally found.^{6,7}

Our patient is part of this new outbreak population but presents with this rare "bubonulus" appearance. High rates of *C trachomatis* Ig G levels suggest an old infection. Can modified lymphatic drainage of the infected site after use of the cock ring (with or without HIV modified immune response) be an explanation for these acute localised buboes?

For instance, it is still unclear why rectal symptoms are found more frequently in the new LGV MSM cases. Is the appearance of LGV a new outbreak or has it been always present undiagnosed? Some studies suggest that LGV has always been present but was unreported,^{8,9} and the paucity of penile lesions is consistent with failing to recognise cases in insertive partners.

Ward *et al* recently described 19% prevalence of sex toy use and 32% of fisting in the LGV population but with missing data⁷ and some authors showed the possibility of a high rate of asymptomatic⁶ and urethral cases.¹⁰ Our patient provides an additional argument to suggest that host factors (HIV infection, behavioural factors such as sex toys) can influence clinical

Abbreviations: HAART, highly active antiretroviral therapy; LGV, lymphogranuloma venereum; MSM, men who have sex with men; PCR, polymerase chain reaction

Figure 1 Appearance of penis showing subcutaneous nodular formations, oedema, and non-purulent ulcerations of the glans.

presentation, mimicking a new rectal outbreak in a slow epidemic.

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NS was responsible for clinically managing the patient, reviewed the literature, and wrote the manuscript; SB clinically managed the patient and

reviewed the manuscript; BB was responsible for the laboratory diagnosis; RV is head of Department of Dermatology and Venereology and critically commented on the manuscript.

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REFERENCES

- 1 **Perine PL**, Stamm WE. Lymphogranuloma venereum. In: Holmes KK, et al, eds. *Sexually transmitted diseases*. 3rd ed. New York: McGraw-Hill, 1999:424.
- 2 **Favre M**, Hellerstrom S. The epidemiology, aetiology and prophylaxis of lymphogranuloma inguinale. *Acta Derm Venereol* 1954;**34**:(suppl 1).
- 3 **Kumaran MS**, Gupta S, Ajith C, et al. Saxophone penis revisited. *Int J STD AIDS* 2006;**17**:65–6.
- 4 **Gupta S**, Ajith C, Kanwar AJ, et al. Genital elephantiasis and sexually transmitted infections revisited. *Int J STD AIDS* 2006;**17**:157–66.
- 5 **Nieuwenhuis RF**, Ossewaarde JM, Gotz HM, et al. Resurgence of lymphogranuloma venereum in western Europe: an outbreak of Chlamydia trachomatis serovar L2 proctitis in the Netherlands among men who have sex with men. *Clin Infect Dis* 2004;**39**:996–1003.
- 6 **Van der Bij AK**, Spaargaren J, Morre SA, et al. Diagnostic and clinical implications of anorectal lymphogranuloma venereum in men who have sex with men: a retrospective control case study. *Clin Infect Dis* 2006;**42**:186–94.
- 7 **Ward H**, Martin I, Macdonald N, et al. Lymphogranuloma venereum in the United Kingdom. *Clin Infect Dis* 2007;**44**:26–32.
- 8 **Quinn TC**, Goodel SE, Mkrtichian E, et al. Chlamydia trachomatis proctitis. *N Engl J Med* 1981;**305**:195–200.
- 9 **Spaargaren J**, Schachter J, Mancada J, et al. Slow epidemic of lymphogranuloma venereum L2b strain. *Emerg Infect Dis* 2005;**11**:1787–8.
- 10 **Herida M**, Kreplack G, Cardon C, et al. First case of urethritis due to Chlamydia trachomatis genovar L2b. *Clin Infect Dis* 2006;**43**:268–9.

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