

A case of late stage LGV in a woman in Europe

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Background

- Lymphogranuloma venereum (LGV) is a systemic sexually transmitted infection caused by *Chlamydia trachomatis* serovars L1-L3.
- Chronic progressive lymphangitis in women with inguinal and pelvic lymphadenitis which leads to chronic oedema, sclerosing fibrosis of subcutaneous tissue, elephantiasis and chronic genital ulceration, is called "esthiomene".
- We report the first case of 'esthiomene' due to *Chlamydia trachomatis*, L2 serovar, in Europe.

Case report

A 32-year-old, caucasian Spanish woman. Nothing to mention about her personal past and medical history. She had a Spanish stable partner for 6 years and she had not traveled abroad.



Lymphoedema of vulva, affecting mons pubis, labia majora and minora with several fistulous openings connected to each other. The lower third of the vagina showed mucosal fibrosis with a cobblestone appearance. Overall, there was a greater damage on the left side

Case report

Age 2009	Age 2010	Age 2011
<ul style="list-style-type: none"> • Bartholino's abscess due to <i>Streptococcus agalactiae</i> 	<ul style="list-style-type: none"> • January: Vulvar abscess with multiple fistulas that improved with clindamycin. • Six months later the patient was readmitted for vulvar abscess and multiple fistulas. A scanner was performed which showed no collections. Treatment was initiated with doxycycline 100 mg/12 hours and azithromycin 500mg/24 hours, for 3 months, which resulted in the closure of fistulas. • Later on, and on several occasions, due to reappearance of fistulas, she was treated with azithromycin and doxycycline, which resulted in an improvement without definitive closure. 	<ul style="list-style-type: none"> • September: the patient was referred to Tropical Medicine Unit of Hospital Universitario Central de Asturias for a reappearance of a vulvar abscess that was drained in the emergency department. • Partner asymptomatic

Laboratory

- Blood hematology and chemistry were normal
- Polymerase chain reaction and culture for CMV, herpes virus 1 and 2, varicella human papillomavirus L1 virus, mycobacterias, and bacterial cultures, were negative.
- Hepatitis B, C, HIV, HTLV and syphilis antibodies: negative.
- Quantiferon: negative

Other parameters

- Pelvic magnetic resonance:
- Inflammation of the soft tissue on the left labia majora and minora, with some collection, extending to the vaginal and periurethral vestibule bulb without clearly visible fistulas tracks, and bilateral inguinal inflammatory lymphadenopathy.
- Colonoscopy did not show significant findings.

Anatomic pathology

- Discrete epidermal hyperplasia, dermis with a patchy inflammatory infiltrate consisting mostly of plasma cells and lymphocytes, forming **granulomatous-looking structures with foamy histiocytes and peripheral lymphocytes without central caseosis.**
- Giemsa, Ziehl-Nielsen, Warthin-Starry and Grocott staining rule out the presence of acid-fast bacilli, fungal structures, and spirochetes but it shows a **granular staining in the cytoplasmic histiocytic content.**

DIAGNOSTIC

- ***Chlamydia trachomatis* was detected by PCR** (Cobas Taqman CT system; Roche Diagnostic Systems, Branchburg, NJ). Genotyping and alignment of amplicon sequences (Clustal-W2 program) **identified a L2 genotype, no b.**

Evolution.

The patient was diagnosed with LGV in tertiary stage and treated with a combined therapy that included doxycycline 100 mg/12 h and azithromycin 500 mg/24h for 3 months without cure. Surgical resection was required to remove the affected tissues (mons pubis, labia majora and minora, Bartholin gland right and infiltrate vaginal mucosa), proceeding with direct closure of incisions. After the surgery she was treated with doxycycline 100 mg/12 h and azithromycin 500 mg/24h intravenously for the first 3 weeks and orally for 3 months. The patient recovered completely, and PCR for *Chlamydia trachomatis* from a new vulvar biopsy, performed three, six and twelve months after the surgery, was negative.