

ELEPHANTIASIS OF THE PENIS REVEALING LYMPHOGRANULOMA VENEREUM. A CASE REPORT AND LITERATURE REVIEW

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Abstract

Introduction. Lymphogranuloma Venereum is a sexually transmitted disease due to *Chlamydia trachomatis* belonging to groups L1 - L3. The transmission can be direct by sexual contact or indirect. The disease is endemic in tropical and subtropical areas of Africa, Asia and the Caribbean. It occurs most often in the form of inguinal adenitis satellite of the initial chancre may go unnoticed. Genital elephantiasis is a complication.

Observation. We report the case of a patient aged 46 years having as antecedent treated pulmonary tuberculosis in 2005 and then for a relapse in 2006. He had unprotected intercourse and had since 2005 a swelling of the penis with the presence of fistulas. Clinical examination revealed bilateral inguinal lymph nodes. The biological and radiological assessment made allowed to withhold the diagnosis of Lymphogranuloma Venereum. Adequate medical treatment was introduced. The evolution was favorable.

Conclusion. Sexually transmitted infections *Chlamydia trachomatis* in Lymphogranuloma Venereum must be screened to avoid their complications. The prognosis is favorable if the treatment is early. Elephantiasis is irreversible. The research and possible treatment of sexual partners are important elements in the management of these patients.

Keywords: Lymphogranuloma Venereum, Nicolas-Favre disease, *Chlamydia trachomatis*, sexually transmitted Infection, Genital elephantiasis.

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INTRODUCTION

Nicolas-Favre disease, originally described by Wallace in 1833, has been defined as an entity clinical and pathological by Durand (a pupil of Nicolas and Favre) in 1913. It corresponds to Lymphogranuloma Venereum (LGV) which is a sexually transmitted infection (STI) rare affecting mainly the lymphatic vessels genital starting point, but the locoregional tropic, due to serovars L1, L2 or L3 of *Chlamydia trachomatis*. In tropical countries in Africa, the proportion of genital ulcers which can be attributed to Lymphogranuloma Venereum varies between 1% and 10% [1].

We report a case of LGV of the penis revealed during an elephantiasis.

PATIENT AND OBSERVATION

It's an old patient of 46 years having as antecedent treated pulmonary tuberculosis in 2005 and then for a relapse in 2006 (declared cured), smoking cessation Chronicle at the rate of 20 packets/year weaned with occasional alcoholism. He had unprotected sexual intercourse. The patient presented since 2005 a swelling of the penis increasing volume with fistula and from pus and rebel urine to prescribed treatments.

Clinical examination found swelling of the penis with presence next to many erosions to purulent surface and variable size (1-2 cm of long axis) and bilateral inguinal lymph nodes (figure 1). A comprehensive review was conducted. Searching of BK (Koch's Bacillus) in sputum, in the urine by gene type Polymerase Chain Reaction (RT-PCR) amplification reactions and pus from fistulas of the

rod came back negative. A radiological complement made including an ultrasound of the soft parts which was overall infiltration of the wall of the penis measuring 15 mm thick with a normal aspect of the corpora cavernosa and the corpus spongiosum. Cystogram intra-venous retrograde and urinary (UCRM) showed urethral and peri-uretrales lesions earlier distal 4 cm approximately with the presence of multiple fistulas of the anterior urethra in showerhead without stenosis. The posterior urethra had a late appearance with a moderate opening of the bladder neck on voiding shadow.

The biopsy of a corpus cavernosum objectified chronic fibro-inflammatory redesigns without signs of specificity. The spongy body biopsy revealed a hyperplastic fleshy bud. The STI (sexually transmitted Infection) including identification of HIV (Human Immunodeficiency Virus), hepatitis B and C and syphilis was negative. Serology by micro-immuno-fluorescence of Chlamydia trachomatis was positive. MRI (Magnetic Resonance Imaging) showed an infiltration of the subcutaneous tissue around the spongy body compatible with Nicolas Favre disease and the existence of bilateral inguinal lymphadenopathy (figure 2). A surgical biopsy of the penis confirmed the diagnosis.

Before these arguments clinical and bacteriological diagnosis of Lymphogranuloma Venereum was retained and the patient received doxycycline 200 mg/day for 2 months, colchicine 1 mg/day for 2 months and twice-daily washing with water and soap with eosin spray application. The evolution was favorable despite the persistence of a swelling moderate of the penis.



Fig. 1 : Elephantiasis of the penis and fistula



Fig. 2: MRI: Infiltration of the subcutaneous tissue around the spongy body and presence of air bubbles.

DISCUSSION:

Chlamydia trachomatis is the most common bacterial cause of STIs in humans [2]. It is transmitted almost exclusively by sexual way, and like all other STIs, it affects mainly the young and sexually active people. The transmission may be occasionally non-sexual, by personal contact, objects or in laboratory accidents [5]. There is a common association with HIV seropositivity [3]. LGV is still endemic in Asia, Africa, South America and parts of the Caribbean, where it is a major cause of morbidity [5,7]. Since 2003, an outbreak of LGV (serovar L2) of venereal origin documented among gay men in Western Europe and Australia [8,9]. It is most common in the second and third decades when sexual activity higher.

It occurs most often in the form of adenitis inguinal satellite of the initial chancre that can pass unnoticed [6]. Clinical manifestations depend on the site of inoculation and its evolution is divided into three parts. Primary, genital, injury is ephemeral and often unnoticed. It appears, after an incubation period of 3 to 30 days in the form of a papule, a pustule or painless genital erosion. It is usually located in the gland, vagina or lips, possibly to the cervix. The secondary, often inguinal and/or femoral, unilateral lesion in 2/3 of the cases is the Lymphadenopathy or bubo. It occurs 10 to 30 days after the appearance of the lesion and fistulise spontaneously to the skin in several pertuis, in showerhead. The tertiary stage, not necessarily following the stage of Bubo, is often described as the genito-ano-rectal syndrome. Genital elephantiasis, genital Lymphedema, proctitis and colitis have been reported [1.11]. Our patient was supported late in the stage of fistula and elephantiasis of the penis in relation to lymphatic obstruction.

Acute LGV is almost always associated with systemic manifestations such as fever and blood Leukocytosis, but it is rarely associated with systemic complications such as meningoencephalitis. Genital elephantiasis by lymphatic impairment, strictures, fistulas of the penis, urethra, rectum are late complications, appearing after years of latency [5].

Iterative punctures of the bubon must be performed up to its collapse. Incision and drainage of the abscess are contraindicated. The most frequent violations are lesions of the ano-genital sphere to inguinal Lymphadenopathy, haemorrhagic proctitis or penile chancre type. In the absence of treatment, scaling can be done towards the abcedation, the fistulisation and lesions of genital fibrosis later [4].

In a small proportion of cases, LGV in humans can be the appearance of chronic lesions progressive, infiltrating, ulcer or fistula of the penis, urethra, or scrotum. The associated lymphatic obstruction can cause elephantiasis as in our case. When a urethral narrowing occurs, it usually sits on the posterior urethra and translates dysuria [5]. In our case, the patient had a stenosis of the anterior urethra with the presence of several fistula to the UCRM.

The diagnosis of Chlamydia trachomatis infection is easy thanks to molecular biology testing and serology. Because of its intracellular nature, culture is rarely used compared to molecular biology techniques that are more sensitive. The bacteriological diagnosis is obtained from the ganglionic pus samples, collected by lymph node puncture, or swabs of genital ulceration. Chlamydia trachomatis can be identified by cell culture (prerogative of highly specialized laboratories) or polymerase chain reaction. PCR (Polymerase Chain Reaction) has become the reference method and recently, real-time PCR Multiplex tests have been developed for the rapid detection of Chlamydia trachomatis and its serotype specific [10]. This test in our patient was positive.

Diagnosis requires the genotyping of Chlamydia trachomatis: serovar L. Serology may be considered as an additional positive diagnosis argument when it shows a seroconversion or a significant increase in antibody titer at 15 days interval [1].

The treatment of LGV does not only to cure the infection but also to prevent complications. Doxycycline 200 mg/day in one to 2 daily catch for 21 days is the classical treatment of first-line. Our patient was given doxycycline at dose of 200 mg/day for 2 months. Second-line treatment is erythromycin 2 g/day in 4 daily catch for 21 days. Azithromycin is effective and would allow a shorter duration of treatment, but its use is not well codified in this indication [1].

The follow-up must be regular up to the healing that can take between three and six weeks. About spontaneously favorable developments in eight weeks is also possible, but the disease can relapse. The search for other STIs must be systematic. Sexual partners who had a contact at risk in the months preceding the onset of symptoms should be examined, diagnosed, and treated [1].

CONCLUSION

LGV is experiencing a resurgence since 2003. This is the default diagnosis and therefore treatment with no treatment of sexual partners. Primary prevention is paramount. Diagnosis is based on the evidence of the bacteria in infected tissues allowing adequate support avoiding complications.

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