



Genital Lichen Planus in Males: Clinical, Laboratory, and Histological Aspects of 7 Cases

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Abstract

Introduction: Genital Lichen Planus (GLP) is associated with high morbidity due to the possible progression to stenosis, genital synechia, and carcinoma, in addition to psychological impact. We propose an iconographic presentation describing the characteristics of patients with GLP and complementary tests in order to assist the diagnosis.

Materials and Methods: Retrospective study of male patients with GLP from a tertiary hospital in São Paulo, Brazil, between January 2009 and December 2019. Therein are described their clinical, histological, and laboratory aspects.

Results: Seven patients were included. The most affected site was the shaft of the penis (71%), followed by the glans penis (54%). All patients presented with Wickham's striae, with the annular form being the most common clinical form (71%). Typical histological changes were observed in 100% of cases.

Discussion: The clinical manifestations of GLP show the importance of acknowledging the epithelium involved in the clinical expression of the lesion. All histopathological tests were compatible with GLP, thereby demonstrating the importance of choosing an appropriate biopsy site.

Conclusion: This study contributes to the diagnostic elucidation of a chronic and relapsing genital disease by highlighting clinical aspects with the aid of histopathological diagnosis and interpretation of laboratory test results.

Keywords: Lichen planus; Penis; Dermatitis; Genital; Pruritus; Autoimmunity

Introduction

Lichen Planus (LP) is an autoimmune and inflammatory disease. It presents with an unpredictable, chronic, and recurrent course, affecting 0.2% to 1% of the adult population between 30 and 60 years. It is more common among females in a ratio of two women to one man, with no racial predisposition. Sporadic in character, rare are the reports of familial cases, which are of earlier onset and have a recurrent course. The disease can affect all skin types, with a predilection for the oral and genital mucosae [1,2].

The skin, appendages and mucosae play a role in protecting against aggression from external agents, such as invasion by microorganisms, exposure to allergens, and also mechanical protection against trauma, in order to ensure the integrity of the organism and its homeostasis. For this reason, the cutaneous-mucous epithelium acts like the first line of physical and immunological defense, which constitutes an essential part of the immune system. Biological processes affecting the skin's barrier function and interfering with its differentiation and repair process can lead to the development of LP [3]. The disease starts off with cellular damage to the keratinocytes in the basal layer, secondary to autoimmune stimulation. This reaction is mediated by T lymphocytes that normally do not respond to the skin. However, when they are stimulated by exogenous agents that bind to the lymphocyte receptor by a mechanism of molecular mimicry, an aggression reaction to keratinocytes is triggered, which leads to their apoptosis [4].

Its clinical forms can be divided into linear, hypertrophic, atrophic, and annular. It presents with an acute course and a predilection for the flexor surface of the wrists. A typical lesion is characterized by a purple-colored papule having a polygonal contour and flat surface surmounted

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Received Date: 21 Feb 2022

Accepted Date: 15 Mar 2022

Published Date: 25 Mar 2022

Citation:

Tamanini JM, Lellis RF, Veasey JV.
Genital Lichen Planus in Males: Clinical,
Laboratory, and Histological Aspects of
7 Cases. *Clin Case Rep Int.* 2022; 6:
1301.

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by thin, off-white lines, conferring on the lesion a lace-like pattern, also known as Wickham's striae [5]. Its clinical manifestation varies and is determined by the affected topography, epidermis' respective peculiarities, time of progression, which interferes in the thickness and color of the lesion, and by the severity of the condition, i.e. a more severe condition may progress with erosions and blisters, depending on how aggressively the basal layer has been affected. The condition may be associated with the Koebner phenomenon, as represented by the appearance of lesions following local trauma [1].

Mucosal involvement is present in 45% of males and 53% of females, with the genital region being especially affected in males [6]. LP lesions can adopt different clinical presentations according to the time of progression, location, and severity. The clinical forms of mucosal LP are classified into reticular, annular, erosive, bullous, and atrophic types, and may be the only manifestation of the disease [7]. The oral mucosa is the most affected site, followed by the genital track. However, LP can affect any mucous site, such as the eyes, tympanum, esophagus, and trachea. The symptoms vary, with pruritus being the most common symptom, reported in cases of either asymptomatic or painful lesions, or with a burning sensation. Genital Lichen Planus (GLP) is found in 25% of the male population with LP, and its morbidity is due to the possibility of it progressing with stenosis, genital synechia, and even carcinoma, in addition to the psychological impact of a sexual nature resulting from this chronic and recurrent genital dermatosis, whose diagnosis is often made late, thereby causing great anxiety in the patient [2,7].

LP has been observed in association with other comorbidities, especially those involving immune disorders. The correlation between LP and Hepatitis C Virus (HCV) infection is well described, especially when erosive oral lesions are present [8,9]. The relationship between this dermatosis and the presence of the Hepatitis B Virus (HBV), active hepatitis A, and vaccination for hepatitis B has also been described [10]. There are reports on the relationship between the disease and ulcerative colitis, alopecia areata, dermatomyositis, morphea, lichen sclerosus, lupus erythematosus, pemphigus, and hematological neoplasms [9], corroborating the hypothesis of autoimmune etiology. Medicines, ingestion of quinine in tonic water, and allergy to the metals used in dental restorations lead to clinically indistinguishable lesions of LP, which is characterized by lichenoid eruptions, the topography, histopathology, and progression of which have particular features [11].

The diagnosis is based on clinical and histological aspects. Classic cases can be diagnosed clinically, yet an anatomopathological study is often necessary. The histological findings that are representative of LP involve keratinocyte damage (keratinocyte damage apoptosis at the dermoepidermal junction, which leads to hydropic degeneration of the basal layer with subsequent pigmentary incontinence) and lymphocytic infiltrate (superficial perivascular, and/or with a band-like arrangement, called lichenoid infiltrate), giving rise to typical changes in the epidermis and the dermoepidermal junction, such as orthokeratotic hyperkeratosis, hypergranulosis (which represents the clinical finding of striations), and acanthosis [12].

We propose an iconographic presentation describing the clinical and histological characteristics of patients with GLP treated at a tertiary hospital in the city of São Paulo/Brazil, in order to assist urologists and dermatologists to improve the diagnosis of skin lesions.

Methodology

This is a retrospective study with male patients who received

treatment at a dermatology clinic of a tertiary hospital in the city of São Paulo, Brazil, between January 2009 and December 2019. All patients having a clinical and histopathological diagnosis of genital lichen planus were included.

The following aspects were analyzed in these patients: Age and comorbidities, presence of extragenital involvement of the skin and/or mucosa, time of progression, location, clinical form, and lace-like pattern (Wickham's striae, or the lack thereof) of the genital lesion.

The findings were included on a Microsoft® Excel (version 15.33) spreadsheet in order to compare the clinical and histological data with each other.

Results

Seven patients were included, with a median age of 30.5 years and a mean age of 39.42 ± 16.53 years. The lesion's median time of progression was six months, and the lesion's mean time of progression was 27.71 ± 48.72 months. Four patients (57%) presented with some comorbidity, and the locations on the genital were: Glans penis (57%), prepuce (28%), balanopreputial sulcus (42%), penis shaft (71%), and penis base (14%), with only one patient having a lesion in only one single anatomical site of his genital. All patients had a lace-like pattern corresponding to Wickham's striae, and the clinical forms present on the genital were: Annular (71%), hypertrophic (14%), and erosive (14%). Two patients (14%) had concomitant extragenital involvement of the skin and one patient, of the oral mucosa. Patients' clinical aspects are illustrated in Figure 1, and their clinical data are shown in Table 1.

In regard to histological aspects, all patients presented with keratinocyte apoptosis at the dermoepidermal junction (100%), with six cases (85%) having been observed in isolation and, in one patient (14%), in a confluent manner. In all cases (100%) hydropic degeneration of the basal layer, acanthosis, hypergranulosis, and orthokeratotic hyperkeratosis were present. Lichenoid superficial perivascular lymphocytic infiltrate and pigmentary incontinence were also observed in all patients (100%). Band-like lymphocytic infiltrate was found in six cases (71%) in varying degrees; and in two cases (14%), only an outline of this finding was observed; whereas in three cases (42%) this finding was observed in a more robust manner. Histological aspects are shown in Table 2 and Figure 2 illustrates the findings described.

In relation to laboratory tests, HIV, hepatitis C, hepatitis B, Anti-Nucleus Factor (ANA) serologies and a non-treponemal test for screening syphilis (VDRL) were conducted. Three patients (42%) had undergone complete laboratory screening, two (28%) had incomplete laboratory tests; and another two (28%) had undergone no laboratory screening tests. Three patients (42%), who were coincidentally those having all laboratory test results, tested negative for all aspects surveyed. One patient (14%) tested negative for HIV, hepatitis and VDRL, but his other parameters had not been tested. One patient (14%) had hepatitis C and syphilis, coincidentally, with no other tests having been performed. Patients' laboratory results are shown in Table 3.

Discussion

Genital lesions are a common complaint in a physician's office, and most doctors and patients tend to instinctively associate those with sexually transmitted infections. However, most complaints are actually due to inflammatory diseases such as balanitis, eczema,

Table 1: Description of the clinical aspects of seven cases of genital lichen planus.

Case	Patient's Characteristics			Extragenital Involvement		Genital Involvement		
	Age (years)	Time of progression (months)	Comorbidities	Cutaneous	Mucosa	Location	Clinical Form	Striations
A	30	2	Peripheral venous insufficiency. Occasional use of dipyron	Disseminated throughout the body. Sparing the scalp and palmoplantar region	Jugal mucosa (erosive form)	Glans penis, prepuce	Annular	Present
B	43	3	Genital dyskeratotic acanthoma	Absent	Absent	Balanopreputial sulcus, Penis shaft	Annular	Present
c	27	120	Absent	Absent	Absent	Penis shaft	Hypertrophic	Present
D	45	3	Absent	Absent	Absent	Glans penis, Balanopreputial sulcus, Prepuce	Annular	Present
E	73	24	Absent	Absent	Absent	Glans penis, Penis shaft	Erosive	Present
F	27	36	Epilepsy	Absent	Absent	Glans penis, Balanopreputial sulcus, Prepuce	Annular	Present
G	31	6	Cocaine user	Disseminated throughout the body	Absent	Penis shaft. base	Annular	Present

Table 2: Description of the histological aspects of seven cases of genital lichen planus.

Case	Epidermal Histological Changes					Dermal Histological Changes		
	Orthokeratotic hyperkeratosis	Hypergranulosis	Acanthosis	Keratinocyte apoptosis at the dermoepidermal junction	Basal layer hydropic degeneration	Lymphocytic infiltrate		Pigmentary incontinence
						Pervascular superficial lichenoid	"Band-like" arrangement	
A	Present	Present	Present	Confluent	Present	Present	Outline	Present
B	Present	Present	Present	Isolated	Present	Present	Present	Present
C	Present	Present	Present, hyperplastic	Isolated	Present	Present	Absent	Present
D	Present	Present	Present	Isolated	Present	Present	Outline	Present
E	Present	Present	Present	Isolated	Present	Present	Absent	Present
F	Present	Present	Present	Isolated	Present	Present	Present	Present
F	Present	Present	Present, hyperplastic	Isolated	Present	Present	Present	Present

Table 3: Laboratory test results from the seven cases of genital lichen planus.

Case	Serology					ANF
	HIV	Hepatitis C	Hepatitis B (HbsAg)	Hepatitis B (Anti-Hbs)	Syphilis (VDRL)	
A	-	-	-	-	-	-
B	-	-	NP	NP	-	NP
C	NP	NP	NP	NP	NP	NP
D	NP	NP	NP	NP	NP	NP
E	NP	+	NP	NP	+	NP
F	-	-	-	-	-	-
G	-	-	-	-	-	-

"-": Non-Reagent; "+": Reagent; "NP": Not Performed

and lichenoid diseases [6,12,13]. In a study conducted in the United Kingdom with 226 patients receiving treatment over a three-year period, only ten received a clinical diagnosis (i.e. without a biopsy) of genital lichen planus [6]. In another study also carried out in the United Kingdom with 600 patients, in turn, 7.3% were diagnosed with genital lichen planus, of which only 11 were confirmed with a biopsy [14]. Such data put into perspective the findings demonstrated herein, in which the seven cases were clinically diagnosed and confirmed by means of a histopathological examination.

Patients with cutaneous lichen planus present with genital involvement in 1.5% to 3% of cases [2]. On the other hand, GLP patients concomitantly have extra-genital manifestations of the disease on the skin in about 20% of cases [15]. Of the cases herein presented,

only two patients had extra-genital skin lesions, a proportion that was similar to that described in the literature. Concomitant genital lesions and in the oral mucosa are rare and found in only 1.36% of cases [2] of those seven patients, only one had his oral mucosa affected.

The most common clinical presentation of GLP is annular and preferably on the glans penis [1]. Of the cases shown here, five are annular; one is erosive, and one is hypertrophic. Of the annular cases, four involved the glans penis and one affected the shaft of the penis; other clinical presentations, in turn, affect the shaft of the penis. This shows the importance of epithelium involvement for the clinical expression of the disease, since the glans is composed of a semi-mucosa of non-keratinized, stratified flat epithelium and the shaft of the penis, in turn, is composed of keratinized, stratified



Figure 1: Clinical aspect of the seven cases of genital lichen planus.

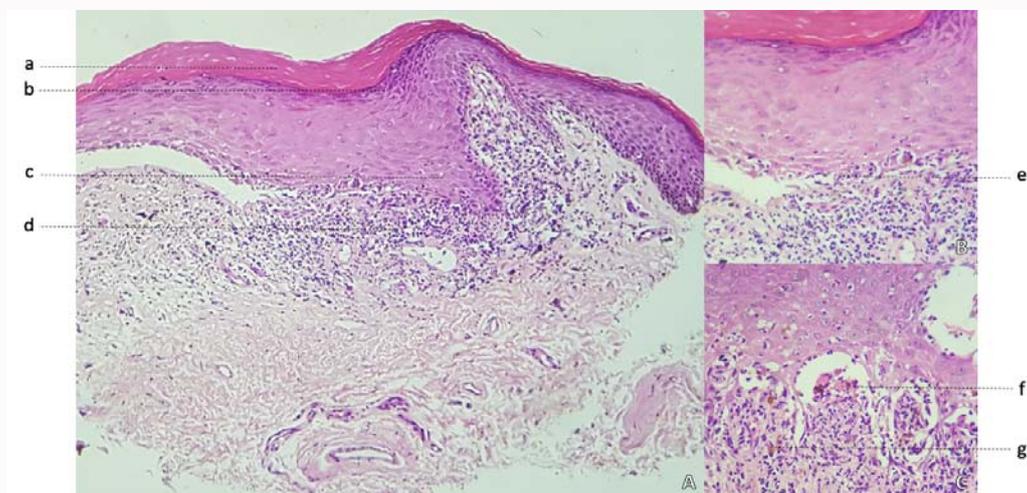


Figure 2: Histopathological examination (hematoxylin-eosin) from a case of genital lichen planus, show the findings described. A (x100 magnification): orthokeratotic hyperkeratosis (a), hypergranulosis (b), acanthosis (c), and lichenoid superficial perivascular lymphocytic infiltrate (d). B (x400 magnification): hydropic degeneration of the basal layer (e). C (x400 magnification): keratinocyte apoptosis at the dermoepidermal junction (f), and pigmentary incontinence (g).

flat epithelium skin [16]. Elakis and Hall [17] in 2017 analyzed the presence of genital dermatosis and the circumcision status and noted that the incidence of changes is significantly higher in uncircumcised patients: Among the 386 patients studied, 16 had GLP, 67% of who were patients with an intact foreskin. The presence of foreskin promotes the formation of an intertriginous region between the inner prepuce, the balanopreputial sulcus and the glans penis, which favors inflammatory processes in this region due to the accumulation of substances [17]. This situation favors a disorder of epithelial differentiation, already proven to be related to genital lichen planus in previous molecular studies [3]. All cases presented in this study are of uncircumcised patients, a common situation in Brazil [18].

Lichen planus can be diagnosed clinically in classic cases, although biopsy is useful for confirming the diagnosis and is necessary in less typical presentations. Histology shows a characteristic “sawtooth” acanthosis pattern in epidermal hyperplasia, hyperparakeratosis with thickening of the granular cell layer and vacuolar alteration of the basal layer of the epidermis, with intense infiltration (mainly T cells) in the dermal-epidermal junction that can cause in apoptosis of some keratinocytes [1]. All the cases studied had the elements described. In case No. 2 in particular, whose clinical presentation was erosive

genital lichen planus, the biopsy was not performed on the ulcerated region, but rather on the perilesional skin, where intact epidermis and the clinically characteristic violet lace-like were found. Thus, the histological findings were compatible with the classic description, since the epidermis was represented in the sample sent to the laboratory. It is worth mentioning that performing the biopsy on the lace-like pattern (Wickham’s striae) is important in order to make any sign of hypergranulosis more evident in the histopathological exam [5].

In regard to the patients’ laboratory test results, only three patients underwent a thorough investigation, i.e. had their HIV, Hepatitis B and C, ANF serologies investigated tested for syphilis by means of a non-treponemal test. Of the five patients whose Hepatitis C serology was investigated, only one tested positive. Such laboratory investigation is carried out in lichen planus cases, since there are studies demonstrating a significant causal relationship between Hepatitis C virus and lichen planus [8]. In a study conducted with 66 patients with lichen planus who had their Hepatitis C serology investigated, 7.5% tested positive vs. 0.69% of 44,947 blood donors from the same institution, which demonstrate a clear and statistically higher prevalence of infection in patients with lichen planus when

compared to blood donors at the same hospital [8]. It is worth mentioning that the clinical erosive form of the mucosa is the one that is most related to positivity for hepatitis C [8], a situation that was confirmed in the cases presented herein, since the only positive serology for the hepatitis C virus was the very case involving genital erosive clinical presentation.

Conclusion

The clinical aspects of dermatosis of the penis encompass multiple medical specialties. This study contributes to the field of research on genital dermatosis by presenting the clinical, histological and laboratory test aspects and discussing the findings and comparing them with the aspects described in the literature.

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