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What is the diagnostic process experience of patients with genital Lichen? An italian survey

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Summary ***Introduction: Genital Lichen Sclerosus (GLS) is a chronic inflammatory disease due to autoimmune events that occurs in anogenital region. It seems to affect mostly women but both the etiology and the prevalence of the disease are largely unknown. The aim of this cross-sectional study was to examine the real-world diagnostic and therapeutic experiences of patients with GLS, focusing on their perceptions and expectations regarding disease management.***

Methods: Utilizing Google Forms, we developed a questionnaire consisting of 10 items aimed at examining the diagnostic and therapeutic experiences of patients with GLS. This survey was distributed via email to all members of the Italian Association of patients with Lichen Sclerosus (LISCLEA), which includes 564 female and 216 male members. The survey was accessible for a period of 48 hours in February 2020.

Results: Of the 780 members surveyed, 280 (36.3% response rate) completed the questionnaire, comprising 226 females (80.7%), 53 males (18.9%), and 1 respondent (0.4%) who did not declared her/his gender identity. A significant 34% of respondents waited over five years for a correct diagnosis of GLS. Diagnostic challenges were frequently reported, with a majority (78%) believing that doctors' knowledge about LS is inadequate. Moreover, 63.9% expressed a need for better medical training concerning GLS, supported by calls for more research networks (42.5%) and specialized centers (26.1%). GLS had a severe impact on sexual health and relationships; 57.3% reported anxiety due to GLS, and 39% avoided intercourse because of symptoms like pain and discomfort.

The majority (95%) received local treatments, while a small percentage (5%) underwent surgical interventions such as circumcision. The diagnostic and therapeutic process was perceived as difficult by most patients (82%).

Conclusions: GLS profoundly affects patients' quality of life, causing significant anxiety, discomfort, and often hindering sexual activity. The study highlights the commonality of late diagnoses and the insufficient referral of patients to specialists, underscoring the need for greater awareness and expertise among healthcare providers. Enhancing doctor awareness and knowledge could facilitate earlier diagnosis and more effective management of GLS, thereby improving outcomes for those affected by this debilitating condition. This research advocates urgent enhancement in both medical education regarding GLS and the establishment of more specialized care pathways to better address the complexities of this disease.

KEY WORDS: *Genital; Penile; Lichen; Sclerosus.*

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INTRODUCTION

Genital Lichen sclerosus (GLS) is a chronic inflammatory skin disease with subsequent fibrosis causing the late complications. It most commonly occurs in the anogenital region and it is responsible for sexual dysfunction and urological morbidity. The precise etiopathogenesis of GLS remains controversial, (1) although genetic, autoimmune (2) and infective (such as human papillomavirus HPV) factors have been implicated. The Koebner phenomenon describes the development of lesions in previously normal skin after scratching or from areas that have undergone trauma (3-5). In men occur on the glans penis or foreskin (6). Frenulum is a particular target manifesting sclerosis or complete obliteration, constrictive lichenoid posthitis is commonly seen associated with a fibrotic preputial band causing "*hourglass*" wasting of the penile shaft. Women may develop scarring of the labia minora, labia majora and the clitoris (7). The spectrum of presentation is very wide, ranging from asymptomatic condition to disabling urinary and sexual symptoms such as itching, burning, bleeding, splitting, dysuria, and pain associated with sexual activity (7, 8) which may reduce *quality of life* (QoL) (9-11). GLS can increase anxiety and stress having a negative impact on the overall mental health (12).

GLS can occur at any age and in both sexes. The male-to-female ratio varies between 1:3 and 1:10 (13). The prevalence is approximately 1.7% in females (13) and 0.07% in males (14) although the exact prevalence is unknown. It's probably under-estimated; since a third of cases are asymptomatic (13), furthermore it is managed by many specialties as well as there's a significant under-recognition and hence under-reporting by patients and physicians. The literaure available on its prevalence and the

patients' real-life experiences are lacking and limited because many studies evaluated patients based on diagnosis alone (9). To address the knowledge gap of these topics, it would be necessary to investigate the impact of GLS on QoF and patient's perception regarding their diagnostic-therapeutic process.
The goal of this cross-sectional study is to collect data from persons having or that had a GLS about their diagnostic and therapeutic experiences evaluating the need to implement the management of these patients.

Materials and methods

This cross-sectional study was conducted in collaboration with Lisclea, an Italian patient association that supports individuals affected by GLS and their caregivers. Lisclea provides a web platform filled with quality information for managing the disease. A structured questionnaire was developed using Google Forms. The questionnaire consisted of 10 close-ended questions designed to gather demographic data (age, sex, geographic area within Italy), clinical symptoms, patients' perspectives on their diagnostic and therapeutic experiences, and perceptions of doctors' awareness about GLS. The survey targeted 780 Lisclea members who were either currently living with or had previously experienced GLS. Recruitment was facilitated through the association's website, mailing list, and WhatsApp group. Participants were informed about the survey's purpose, the voluntary and anonymous nature of their participation, and the confidentiality of the responses. To ensure data integrity, the survey system allowed only one response per digital identity. The survey was made available for 48 hours during February 2020 and was presented in Italian, the native language of the participants. The questionnaire format included predominantly dichotomous or multiple-choice questions, measuring responses at nominal, categorical, and interval levels.

Results

A total of 280 questionnaires were returned from the 780 distributed, achieving a response rate of 36.3%. Not all participants answered every question, and minors reported being assisted by caregivers, in filling out the questionnaire. The respondents comprised 226 females (80.7%), 53 males (18.9%), and one individual (0.4%) who did not declare her/his gender identity, as shown in Table 1. Regarding diagnostic delays, 34% (95/280) of participants reported waiting more than five years from the onset of symptoms to receiving a correct diagnosis of GLS. Treatment approaches varied, with 95% (265/280) receiving local treatments and 5% (15/280) undergoing surgical procedures such as circumcision. A significant concern among the participants was the perceived inadequacy of doctors' knowledge about GLS, noted by 78% (219/280) of respondents. The impact of GLS on quality of life was notably severe, with 57.3% (161/280) reporting anxiety due to the condition and 39% (109/280) avoiding intercourse because of pain, discomfort, bleeding, and itching. Additionally, 21% (59/280) cited specific anxiety related to their condition. The diagnostic and therapeutic processes were considered difficult by most respondents, 30% (84/280) found it very difficult, 29% (81/280) difficult, and 23% (65/280) quite difficult, while only 17.1% (48/280) rated it as quite easy or easy. There was a strong call for better preparation among doctors, with 63.9% (179/280) advocating for improved medical training, supported by the need for new research networks (42.5%, 119/280) and specialized centers (26.1%, 73/280).

Discussion

This cross-sectional study clearly shows the criticism for patients with GLS to receive appropriate management. According to the study results, less than 20% of patients with LS of genitalia had a diagnosis within 6 months from the onset of symptoms while most of responders waited years (15), as shown in Table 2.
Furthermore 80% of responders have the perception that medical culture on GLS is suboptimal. Despite the benign nature, the GLS have an insidious course that determines serious urological and sexual morbidities. In our study more than 98% of responders reported local symptoms such as itching and discomfort and the 39% assessed that GLS made penetrative intercourse impossible. Probably,

Age of responders years	< 18	18-30	31-50	51-70	> 70	Total
Patient gender						
Female n (%)	18 (6.4%)	6 (2.1%)	71 (25.4%)	123 (44%)	8 (2.9%)	226 (80.7%)
Male n (%)	6 (2.1%)	9 (3.2%)	27 (9.6%)	11 (3.9%)	\	53 (18.9%)
Gender not declared	\	\	\	1 (0.4%)	\	1 (0.4%)
Total n (%)	24 (8.6%)	15 (5.4%)	98 (35%)	134 (48%)	8 (2.9%)	280 (100%)

Table 1.
Distribution of patients by age and gender.

Time needed for the diagnosis. Months	< 6 months	6-12 months	12-36 months	36-60 months	> 60 months	Non declared	Total
Patient gender							
Female n (%)	33 (12%)	12 (4.3%)	62 (22%)	38 (14%)	81 (29%)	\	226 (80.7%)
Male n (%)	9 (3.2%)	6 (2.1%)	21 (7.5%)	3 (1.1%)	14 (5%)		53 (18.9%)
Gender not declared	\	\	\	\	\	1 (0.4%)	1 (0.4%)
Total n (%)	42 (15%)	18 (6.4%)	83 (30%)	41 (14.6%)	95 (34%)	2 (0.7%)	280 (100%)

Table 2.
Time elapsed from the first onset of symptoms to the correct diagnosis of patients.

Table 3.
Details about the healthcare professionals who performed the diagnoses.

Who performed the diagnosis	Gynecologist	Dermatologist	Self diagnosis using internet	Plastic Surgeon	General Practicer	Urologist	Andrologist	Non healt-care professional	Total
Patients gender									
Female n (%)	137 (49%)	62 (22%)	11 (3.9%)	4 (1.4%)	2 (0.7%)	\	\	10 (3.6%)	226 (80.7%)
Male n (%)	\	25 (8.9%)	5 (1.8%)	4 (1.4%)	2 (0.7%)	11 (3.9%)	2 (0.7%)	4 (1.4%)	53 (18.9%)
Gender non declared	1 (0.4%)	\	\	\	\	\	\	\	1 (0.4%)
Total n (%)	138 (49%)	87 31%)	16 (5.7%)	8 (2.9%)	4 (1.4%)	11 (3.9%)	2 (0.7%)	14 (5%)	280 (100%)

the remarkably high impact of GLS on sexual health and quality of life is partly due to a selection bias. Since other studies investigating the burden of GLS in national populations are lacking, our results are difficult to compare. Of course, the study is not devoid of limitations. The population studied is composed of people that joined a patient's association probably because the impact of GLS in their life. The high number of non-responders was maybe due to the short time the survey was available. However, the 95 patients that waited 5 years before being diagnosed with genital LS correspond to 34% of responders and to the 12% of all the Lisclea members. This data is important by itself because it underlines that a considerable number of patients were not able to be correctly diagnosed for a very long period. Of note, GLS changes the genitalia architecture in a time dependent mode. *Cooper et al.* showed in their study of 327 women that a delay in diagnosis of 2 years or less was associated with less scarring at diagnosis (16). If we considered that clinical diagnosis of GLS date back to the mid-twentieth century and could be suspected with a simple medical examination of genitalia (17) it seems clear that the disease is not recognized because it is not known by physicians.
Treatment options for GLS are various and range from pharmacological to surgical (18), depending on patient factors, the response of previous treatments, the severity and location of disease (19). Proper care can lead to recovery from the disease symptoms however it is crystal clear that any treatment could be given till the correct diagnosis is done. The ability of the survey receiver to understand and answer the questions was not demonstrated, however the questions were very simples and expressed in the mother tongue of the investigated population. Since patients are increasingly actively involved in health care and patient empowerment in the health system is becoming reality, surveys help health care professionals to gather meaningful data that can improve management of diseases. The present study clearly indicated the need to implement the diagnostic and therapeutic process of patients with GLS in Italy. This would ultimately permit us to better understand the real disease burden of and its impact on patients' quality of life.

Conclusions

This study highlights significant gaps in diagnosing and managing GLS, affecting patients' sexual and psychological well-being. With less than 20% of patients diagnosed within six months of symptom onset and 80% finding the medical community's knowledge insufficient, there is a clear need for improved education and training for healthcare providers. The severe consequences of delayed diagnosis necessitate urgent improvements in the diagnostic process and a shift towards more patient-centered management strategies. To address these issues, it is crucial to develop comprehensive, multidisciplinary approaches that include targeted educational initiatives, expanded research, and specialized centers. This will help mitigate the disease's impact and move towards a more informed and proactive healthcare framework for GLS patients.

Declarations

Ethical approval: *All procedures conducted in this study complied with the Institution and National Research Committee's ethical standards, the 1964 Declaration of Helsinki, and its subsequent amendments or equivalent ethical standards. Participants were informed about the survey's purpose, the voluntary and anonymous nature of their participation, and the confidentiality of the responses.*

Availability of data and material: *The data that support the study's findings are available from LISCLEA, but there are restrictions on their availability because they were used under license for the current study and thus are not publicly available. However, the authors' data are available upon reasonable request and with the permission of LISCLEA.*

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Authors' contributions: *SL, study concept, data analysis and interpretation, manuscript original drafting, statistical analyses; MR, data analysis and interpretation, contribution to manuscript writing and editing; LDM, data analysis and interpretation, contribution to manuscript writing and editing; LB, data analysis and interpretation, contribution to manuscript writing and editing; EP, data analysis and interpretation, contribution to manuscript writing and editing; MB, data analysis and interpretation, contribution to manuscript writing and editing; MR, data analysis and interpretation, contribution to manuscript writing and editing; TC, data analysis and interpretation, contribution to manuscript writing and editing; GL, study concept, data analysis and interpretation, contribution to manuscript writing and editing; AP, study concept, data analysis and interpretation, manuscript original drafting, statistical analyses All the authors read and approved the final version of the manuscript and agreed to be accountable for all aspects of the work.*

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QUESTIONARIO

Tempo di raccolta Lunedì 10 - Giovedì 13 febbraio 2020

Se non specificato è possibile inserire una sola risposta.

1. Lasuaetà è?
   - ❑ > 70
   - ❑ 51-70
   - ❑ 31-50
   - ❑ 18-30
   - ❑ 0-17 (genitori per i bambini o di supporto per gli adolescenti)

2. Lei è
   - ❑ Femmina
   - ❑ Maschio

3. Quanto tempo ha convissuto con il Lichen sclerosus prima di avere la diagnosi?
   - ❑ > 5 anni
   - ❑ 1 anno
   - ❑ < di 6 mesi
   - ❑ 2 anni
   - ❑ 3 anni
   - ❑ 5 anni
   - ❑ > 6 mesi
   - ❑ 4 anni

4. In quale Regione vive?
   - ❑ Lazio
   - ❑ Lombardia
   - ❑ Veneto
   - ❑ Emilia Rom.
   - ❑ Sicilia
   - ❑ Puglia
   - ❑ Liguria
   - ❑ Toscana
   - ❑ Campania
   - ❑ Marche
   - ❑ Piemonte
   - ❑ Sardegna
   - ❑ Abruzzo
   - ❑ Friuli VG
   - ❑ Calabria
   - ❑ Umbria
   - ❑ Basilicata
   - ❑ Trentino
   - ❑ Molise

5. Chi ha fatto per primo la diagnosi?
   - ❑ Ginecologo
   - ❑ Dermatologo
   - ❑ Autodiagnosi
   - ❑ Urologo
   - ❑ Chir. plastico
   - ❑ Altro special.
   - ❑ Altro

- ❑ MMG
- ❑ Genitore
- ❑ Andrologo
- ❑ Coniuge

6. Supporto da parte dei medici?
- ❑ Scadente
- ❑ Sufficiente
- ❑ Buona
- ❑ Ottima

7. Quali tra queste conseguenze legate al Lichen sclerosus sono più importanti per lei? (fino a 3 risposte)
- ❑ Prurito
- ❑ Rende impossibile il rapporto
- ❑ Lacerazioni cute
- ❑ Bruciore
- ❑ Ansia
- ❑ Disagio relazionale
- ❑ Dolore
- ❑ Calo del desiderio
- ❑ Altro
- ❑ Sanguinamento
- ❑ Limita fortemente il mio lavoro
- ❑ Mi impedisce di praticare sport

8. Come giudicherebbe il suo percorso di diagnosi e cura finora?
- ❑ Abbastanza difficile
- ❑ Difficile
- ❑ Molto difficile
- ❑ Abbastanza semplice
- ❑ Semplice

9. Quando le è stata fatta diagnosi di Lichen, qual è stato il percorso proposto?
- ❑ Creme a base di cortisone + creme a base di vitamine e altri comp.
- ❑ Creme a base di cortisone
- ❑ Terapia rigenerativa con PRP e lipofilling
- ❑ Terapia in crema + terapia orale
- ❑ Mantenere un controllo nel tempo
- ❑ Altro
- ❑ Circoncisione
- ❑ Chirurgia plastica della vulva
- ❑ Di consultare un centro specializzato
- ❑ Dilatazioni uretrali
- ❑ Terapia locale con acido ialuronico

10. Quali sono gli aspetti da migliorare più rapidamente?
- ❑ Formazione dei medici sulla patologia
- ❑ Percorsi di ricerca
- ❑ Accessi rapidi nei Centri dedicati
- ❑ Centri specializzati
- ❑ Facile identificazione dei Centri
- ❑ Maggiore attenzione delle aziende farmaceutiche
- ❑ Facilitazioni sul posto di lavoro per visite e terapie
- ❑ Campagne di formazione e sensibilizzazione
- ❑ Altro