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Patient-reported outcome measures in a population of medically indigent patients with systemic lupus erythematosus in Puerto Rico

Diana V Rodríguez-Rivera[1], Yerania Rodríguez-Navedo[1], Mariely Nieves-Plaza[2] and Luis M Vilá[1]

Abstract

Objective: To determine patient-reported outcomes measures in indigent patients with systemic lupus erythematosus receiving their healthcare through the Puerto Rico government managed care system and compare these measures with non-indigent patients treated in a private fee-for-service setting.

Methods: A cross-sectional study was conducted in a cohort of 98 Puerto Ricans with systemic lupus erythematosus. Patients from the public group (n = 40) were treated in a university-based specialized systemic lupus erythematosus clinic and the private group (n = 58) in a community-based rheumatology practice. Demographic and clinical features and patient-reported outcomes measures per LupusPRO instrument were determined. LupusPRO captures quality-of-life measures in 12 domains. Differences among study groups were examined using chi-square, Fisher's exact, t-tests, and the Wilcoxon signed-rank test.

Results: The mean (standard deviation) age of the study population was 44.9 (12.0) years; 94 (95.9%) were women. Patients in the public setting were younger and were more likely to have renal disease and elevated anti-double-stranded DNA antibodies, and being treated with azathioprine and cyclophosphamide. Patients from the public sector were more likely to have better quality-of-life measures in the LupusPRO domains of pain/vitality and coping. No significant differences were observed for the domains of lupus symptoms, physical health, emotional health, body image, cognition, procreation, lupus medications, desires/goals, social support, and satisfaction with medical care.

Conclusion: Despite having a lower socioeconomic status and worse clinical status, systemic lupus erythematosus patients from the public sector had equal or better patient-reported outcomes measures than those treated in the private setting. This favorable outcome may be associated with the comprehensive healthcare received by these patients in a specialized lupus clinic.

Keywords

Systemic lupus erythematosus, patient-reported outcomes, quality of life, underserved population, Puerto Rico

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Introduction

Systemic lupus erythematosus (SLE) is a chronic autoimmune disorder which may involve any organ or system. SLE usually affects women of childbearing age.[1] Chronic diseases such as lupus impact patient's overall physical and emotional function; consequently, the personal, social, professional, and economic health may be impaired. Patient-reported outcomes measures (PROM) are essential to assess how patients perceive their health.[2] They provide valuable information that complements clinical assessment resulting

[1]Division of Rheumatology, Department of Medicine, University of Puerto Rico, Medical Sciences Campus, San Juan, Puerto Rico
[2]Graduate Programs, San Juan Bautista School of Medicine, Caguas, Puerto Rico

Corresponding author:
Luis M Vilá, Division of Rheumatology, Department of Medicine, University of Puerto Rico, Medical Sciences Campus, PO Box 365067, San Juan PR 00936-5067, Puerto Rico.
Email: luis.vila2@upr.edu

in a more effective care. The LupusPRO is a comprehensive quality-of-life (QOL) survey designed specifically for patients with SLE.[3–8] It measures lupus symptoms/flare, concerns about pregnancy, brain fog, lupus medicines side effects, satisfaction with care, effects on career/desires/goals, coping, and social support. LupusPRO has been validated in multiple languages, including Spanish.

In 1994, a Healthcare Reform was implemented in Puerto Rico establishing a public, managed care health system for the medically underserved population.[9] The eligibility for the Puerto Rico government insurance is determined by the annual family income adjusted for the number of individuals in the household. Those eligible have an income below poverty level according to US standards; thus, having this insurance is reflective of a lower socioeconomic status. Low socioeconomic status has been found to be related to poor outcomes in lupus patients.[10] Therefore, we sought to determine QOL using the LupusPRO instrument in SLE indigent patients receiving their healthcare through the Puerto Rico healthcare system and compare these measures with non-indigent patients treated in a private fee-for-service setting.

Methods

Patient population

A cross-sectional study was conducted in a cohort of 98 Puerto Rican patients with SLE. All patients were ⩾21 years old, had Puerto Rican ethnicity (self and four grandparents), and fulfilled the American College of Rheumatology (ACR) revised classification criteria for SLE.[11] Patients were recruited from the Lupus clinic of the University of Puerto Rico Medical Sciences Campus (UPR-MSC) in San Juan, Puerto Rico, and from a private general rheumatology practice located in San Juan, Puerto Rico. Patients were enrolled between September 2012 and August 2013. This study was approved by the Institutional Review Board of the UPR-MSC Human Research Protection Office.

SLE patients had their routine visits at 3-month intervals. Additional visits were scheduled as needed according to disease activity or complications. At each visit, including the study visit, a structured clinical note was completed by the physician to gather information regarding demographic parameters, health-related behaviors, clinical manifestations, laboratory tests, pharmacologic treatment, disease activity, and disease damage. For all patients, a lupus autoantibody panel was performed at the time of SLE diagnosis. At study visit, the patient completed the Spanish LupusPRO survey.

Variables

For the analyses, patients were allocated into two groups based on how they received their healthcare. Patients who were evaluated at the Lupus Clinic of the UPR-MSC belonged to the "public group" (managed care setting) and those who received their care at the private rheumatology practice comprised the "private group" (fee-for-service setting).

Demographic parameters, lupus manifestations, serologic abnormalities, comorbid conditions, disease activity, disease damage, and lupus medications were evaluated. Demographic parameters included age and gender. Cumulative SLE clinical manifestations were determined as defined by the ACR classification criteria for SLE.[11] The following autoantibodies were determined at diagnosis: antinuclear (ANA), anti-double-stranded DNA (dsDNA), anti-Smith, and antiphospholipid antibodies. Comorbid conditions examined included arterial hypertension, diabetes mellitus, and hypothyroidism. Disease activity and damage accrual were determined at study visit. Disease activity was ascertained by the Safety of Estrogen in Lupus Erythematosus National Assessment SLE Disease Activity Index (SLEDAI)[12] and the Systemic Lupus Disease Activity Measure-Revised (SLAM-R).[13] Disease damage was assessed using the Systemic Lupus International Collaborating Clinics/ACR Damage Index (SDI).[14] The current (within 4 weeks of the study visit) and cumulative (at any time) exposures of the following therapeutic immunosuppressive/immunomodulator agents were recorded: corticosteroids, hydroxychloroquine, azathioprine, mycophenolate mofetil, and cyclophosphamide.

LupusPRO

The LupusPRO is a validated, self-administered PROM instrument for SLE patients.[3–8] It has eight health-related quality of life (HRQOL) domains: (1) lupus symptoms, (2) cognition, (3) lupus medications, (4) procreation, (5) physical health, (6) pain/vitality, (7) emotional health, and (8) body image, and four non-HRQOL domains: (1) desires/goals, (2) social support, (3) coping, and (4) satisfaction with medical care. The LupusPRO has a 5-point Likert response format, where 0 = *none of the time/not applicable*, 1 = *a little of the time*, 2 = *some of the time*, 3 = *most of the time*, and 4 = *all of the time*. Reverse scoring is required for HRQOL domains. Item scores are totaled for each domain item and the mean domain score is obtained by dividing the total score by the number of items in that domain. The mean raw domain score is transformed to scores ranging from 0 (*worst QOL*) to 100 (*best QOL*).

Statistical analysis

The statistical software STATA version 13 (STATA Corp, College Station, TX, USA) was used to perform the statistical analyses. Differences between study groups were analyzed with chi-square, Fisher's exact, Student's t-tests, and the Wilcoxon signed-rank test, as appropriate. A p-value of ⩽0.05 was considered to represent statistical significance.

Results

In all, 98 subjects were enrolled, 94 (95.9%) were women. The mean (standard deviation [SD]) age at study visit was 44.9 (12.0) years. Forty patients were treated in the public

Table 1. Demographic parameters, clinical manifestations, serologic features, disease activity, and damage accrual in SLE patients.

Characteristics	Public (n = 40)	Private (n = 58)	p-Value
Gender, female (%)	97.5	94.8	0.643
Age at study visit, mean years (SD)	39.7 (11.5)	48.5 (11.0)	<0.001
Cumulative clinical manifestations,[a] %			
Photosensitivity	82.5	86.2	0.616
Malar rash	70.0	69.0	0.913
Oral ulcers	37.5	39.7	0.830
Arthritis	77.5	69.0	0.353
Serositis	22.5	8.6	0.054
Renal disorder	55.0	24.1	0.002
Neurological disorder	7.5	8.6	1.000
Hemolytic anemia	12.5	6.9	0.345
Leukopenia	47.5	37.9	0.345
Lymphopenia	90.0	75.9	0.076
Thrombocytopenia	17.5	8.6	0.188
Serologic features, %			
Antinuclear antibodies	97.3	100.0	0.402
Anti-dsDNA antibodies	94.1	57.1	<0.001
Anti-Smith antibodies	63.2	44.4	0.211
Anti-phospholipid antibodies	20.0	34.5	0.119
Selected comorbidities			
Arterial hypertension	65.0	48.3	0.102
Diabetes mellitus	10.0	8.6	0.816
Hypothyroidism	7.5	31.0	0.005
SLAM-R at study visit, score (SD)	5.9 (2.7)	5.3 (2.9)	0.400
SLEDAI at study visit, score (SD)	1.6 (2.1)	1.4 (2.0)	0.717
SDI, score (SD)	0.8 (1.1)	1.1 (1.4)	0.365

SD: standard deviation; dsDNA: double-stranded DNA; SLAM-R: Systemic Lupus Activity Measurement-Revised; SLEDAI: Safety of Estrogen in Lupus Erythematosus National Assessment SLE Disease Activity Index; SDI: Systemic Lupus International Collaborating Clinics/American College of Rheumatology Damage Index.
[a]Per American College of Rheumatology classification criteria for SLE.

setting and 58 patients received their care in the fee-for-service setting. Table 1 depicts the demographic parameters, clinical manifestations, serologic features, disease activity, and damage accrual in SLE patients. SLE patients in the public setting were younger (39.7 versus 48.5 years, $p = 0.003$) and were more likely to have renal disease (55.0% versus 24.1%, $p = 0.002$) and elevated anti-dsDNA antibodies (94.1% versus 57.1%, $p < 0.001$) than patients seen at the private sector. On the other hand, patients from the private sector were more likely to have hypothyroidism (31.0% versus 7.5%, $p = 0.005$). No significant differences were observed for gender, other clinical or serologic SLE manifestations, arterial hypertension, diabetes mellitus, disease activity (by SLAM-R or SLEDAI instruments), and damage accrual.

The pharmacologic therapy of SLE patients is shown in Table 2. SLE patients from the public setting were more likely to be treated with azathioprine (current use: 22.5% versus 5.2%, $p = 0.010$ and cumulative use: 52.5% versus 25.9%, $p = 0.007$) and cyclophosphamide (cumulative use: 35.0% versus 10.3%, $p = 0.003$). No significant differences were observed for corticosteroids, hydroxychloroquine, or mycophenolate mofetil exposure.

Table 2. Immunosuppressive/immunomodulator treatment in SLE patients.

Medications	Public (n = 40), %	Private (n = 58), %	p-Value
Current			
Corticosteroids	67.5	48.3	0.059
Hydroxychloroquine	80.0	75.9	0.629
Azathioprine	22.5	5.2	0.010
Mycophenolate mofetil	20.0	14.3	0.179
IV cyclophosphamide	0.0	1.7	1.000
Cumulative (at any time)			
Corticosteroids	95.0	89.7	0.466
Hydroxychloroquine	97.5	100.0	0.408
Azathioprine	52.5	25.9	0.007
Mycophenolate mofetil	30.0	19.0	0.205
IV cyclophosphamide	35.0	10.3	0.003

IV: intravenous.

The scores of the LupusPRO domains are shown in Table 3. Overall, the best scores were attained for the domains of satisfaction with medical care, body image, and lupus medications, whereas the worst scores were observed

Table 3. Scores of the LupusPRO domains in SLE patients (0 = *worse quality of life* (QOL), 100 = *best QOL*).

LupusPRO domains	All patients (n = 98), mean	Public (n = 40), mean	Private (n = 58), mean	p-Value
Health-related QOL				
SLE symptoms	66.5	69.3	64.4	0.350
Cognition	69.0	74.4	65.3	0.125
Lupus medications	82.1	83.4	81.2	0.626
Procreation	91.1	89.1	92.5	0.647
Physical health	81.8	83.4	80.7	0.524
Pain/vitality	64.6	72.0	59.1	0.017
Emotional health	66.0	67.0	65.3	0.760
Body image	83.5	85.4	82.2	0.493
Non-health-related QOL				
Desires/goals	70.1	72.8	68.2	0.442
Social support	81.5	86.9	77.8	0.127
Coping	79.5	87.2	74.1	0.012
Satisfaction with medical care	95.1	96.7	94.1	0.055

QOL: quality of life; SLE: systemic lupus erythematosus.

for the pain/vitality, emotional health, and SLE symptoms domains. Patients from the public sector had better QOL measures in the pain/vitality (72.0 versus 59.1, $p = 0.017$) and coping (87.2 versus 74.1, $p = 0.012$) domains than patients from the private group. No significant differences were found for the other domains.

Discussion

The advent of patient-centered health brought a turning point in modern medicine, leading to the development of PROM.[15] Health information is traditionally gathered from the patient and interpreted by the physician. Nowadays, with the introduction of PROM, we are able to obtain data directly from the patient using structured methods. These measures yield valuable information (e.g. QOL, disability) that is vital for patient evaluation and management.[3] Our study is the first that compares PROM in SLE patients receiving their healthcare through public and private settings in Puerto Rico. We found that the public group, despite having lower socioeconomic status and poorer disease status, had similar or better PROM than patients seen in the private sector.

Overall, our patients (public and private) reported better PROM when compared to other ethnic groups, including Philippine, Turkish, and Chinese lupus patients.[3,6,8] The relatively mild disease severity of our group, together with other factors such as cultural and healthcare delivery, could explain these differences.

Some demographic and clinical features differed between our study groups. Lupus patients in the public sector were younger and were more likely to have renal disease and elevated anti-dsDNA antibodies. Anti-dsDNA is a surrogate marker of disease activity and damage and is associated with lupus nephritis.[16] As expected, patients from the public were more commonly treated with azathioprine and cyclophosphamide as both drugs are regularly used for the treatment of lupus nephritis.[17]

Our study revealed significant differences in PROM between the study groups. Patients from the public sector reported better coping than the private group. These results contrast with data of prior studies which have consistently found that poverty is associated with worse disease outcomes.[10] Furthermore, decreased coping skills have been observed in underprivileged patients with chronic diseases such as diabetes.[18] We also found that patients in the public group reported better QOL in the pain/vitality domain of the LupusPRO than patients from the private setting. A plausible explanation is that patients from the private group were more likely to have hypothyroidism which is associated with tiredness/fatigue and a wide spectrum of rheumatic and pain syndromes.[19] The pain and lack of vitality experienced by the private group may explain their coping difficulties as high levels of pain are associated with increased anxiety, depression, and worse health-related QOL.[20]

The fact that SLE patients from the public group had their healthcare delivered in a specialized lupus clinic could explain why they had similar or better PROM than patients from the private group. The Lupus Clinic of the UPR-MSC was established in 2002 and was designed to promote expert care for patients with lupus, to train future rheumatologists, and to facilitate lupus research. This clinic provides expert collaborative care from experienced and dedicated board-certified rheumatologists who specialize in the diagnosis and treatment of lupus. Our specialized physicians work closely with nephrologists, pulmonologists, dermatologists, hematologists, and high-risk obstetricians and gynecologists as well as other healthcare professionals such as dedicated nurses and pharmacists. Similar to our experience, Aisiku et al.[21] found that sickle cell disease patients receiving their care in specialized clinics had higher overall satisfaction than those seen in regular clinics.

Some limitations of our study should be addressed. First, this is a cross-sectional study and as such has the limitations inherent to this type of design. Second, the study sample

size was relatively small. Third, relevant sociodemographic information such as educational level was not gathered. Fourth, comorbidities that may affect PROM such as neuropsychiatric disorders and fibromyalgia syndrome were not ascertained. Nonetheless, patients with these conditions were not excluded from the study. Fifth, none of the participants had a disease flare or high levels of disease activity at study visit; thus, we could not evaluate well the association of disease activity with PROM. Finally, data on patient's knowledge or education about SLE or access to support groups were not determined.

In summary, our study compared PROM in a population of Puerto Ricans with SLE. Although patients in the public sector had a worse clinical status, they reported similar or even better PROM than patients from the private group. This favorable outcome may be the result of the comprehensive healthcare received by these patients in a specialized lupus clinic.

Specialized clinics are an important tool as they provide high-quality care leading to effective clinical interventions, and consequently better PROM. Future studies may contemplate administering the LupusPRO instrument to a larger population, and at different time intervals to monitor whether changes in disease activity affect PROM. Finally, the introduction of PROM into clinical practice seems promising and may be used as an essential instrument to improve overall patients' health.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Ethical approval for this study was obtained from the University of Puerto Rico Medical Sciences Campus Institutional Review Board (approval number/ID 7510112).

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Informed consent

Written informed consent was obtained from all subjects before the study.

References

1. Mills JA. Systemic lupus erythematosus. *N Engl J Med* 1994; 330(26): 1871–1879.
2. Jolly M, Pickard AS, Block JA, et al. Disease-specific patient reported outcome tools for systemic lupus erythematosus. *Semin Arthritis Rheum* 2012; 42(1): 56–65.
3. Navarra SV, Tanangunan RM, Mikolaitis-Preuss RA, et al. Cross-cultural validation of a disease-specific patient-reported outcome measure for lupus in Philippines. *Lupus* 2013; 22(3): 262–267.
4. Jolly M, Toloza S, Block J, et al. Spanish LupusPRO: cross-cultural validation study for lupus. *Lupus* 2013; 22(5): 431–436.
5. Bourré-Tessier J, Clarke AE, Mikolaitis-Preuss RA, et al. Cross-cultural validation of a disease-specific patient-reported outcome measure for systemic lupus erythematosus in Canada. *J Rheumatol* 2013; 40(8): 1327–1333.
6. Kaya A, Goker B, Cura ES, et al. Turkish lupusPRO: cross-cultural validation study for lupus. *Clin Rheumatol* 2014; 33(8): 1079–1084.
7. Bourré-Tessier J, Clarke AE, Kosinski M, et al. The French-Canadian validation of a disease-specific, patient-reported outcome measure for lupus. *Lupus* 2014; 23(14): 1452–1459.
8. Mok CC, Kosinski M, Ho LY, et al. Validation of the LupusPRO in Chinese patients from Hong Kong with systemic lupus erythematosus. *Arthritis Care Res (Hoboken)* 2015; 67(2): 297–304.
9. Mayor AM, Vilá LM, De La Cruz M, et al. Impact of managed care on clinical outcome of systemic lupus erythematosus in Puerto Rico. *J Clin Rheum* 2003; 9(1): 25–32.
10. Trupin L, Tonner MC, Yazdany J, et al. The role of neighborhood and individual socioeconomic status in outcomes of systemic lupus erythematosus. *J Rheumatol* 2008; 35(9): 1782–1788.
11. Hochberg MC. Updating the American College of Rheumatology revised criteria for the classification of systemic lupus erythematosus. *Arthritis Rheum* 1997; 40(9): 1725.
12. Petri M, Buyon J and Kim M. Classification and definition of major flares in SLE clinical trials. *Lupus* 1999; 8(8): 685–691.
13. Bae SC, Koh HK, Chang DK, et al. Reliability and validity of systemic lupus activity measure-revised (SLAM-R) for measuring clinical disease activity in systemic lupus erythematosus. *Lupus* 2001; 10(6): 405–409.
14. Gladman DD, Goldsmith CH, Urowitz MB, et al. The Systemic Lupus International Collaborating Clinics/American College of Rheumatology (SLICC/ACR) damage index for systemic lupus erythematosus international comparison. *J Rheumatol* 2000; 27(2): 373–376.
15. Fung CH and Hays RD. Prospects and challenges in using patient-reported outcomes in clinical practice. *Qual Life Res* 2008; 17(10): 1297–1302.
16. Vilá LM, Molina MJ, Mayor AM, et al. Clinical and prognostic value of autoantibodies in Puerto Ricans with systemic lupus erythematosus. *Lupus* 2006; 15(12): 892–898.
17. Castro-Santana LE, Colón M, Molina MJ, et al. Efficacy of two cyclophosphamide regimens for the treatment of lupus nephritis in Puerto Ricans: low vs. standard dose. *Ethn Dis* 2010; 20(1 Suppl. 1): S1-116–S1-121.
18. Walker AF, Schatz DA, Johnson C, et al. Disparities in social support systems for youths with type 1 diabetes. *Clin Diabetes* 2015; 33(2): 62–69.
19. Tagoe CE. Rheumatic symptoms in autoimmune thyroiditis. *Curr Rheumatol Rep* 2015; 17(2): 5.
20. Waldheim E, Elkan AC, Pettersson S, et al. Health-related quality of life, fatigue and mood in patients with SLE and high levels of pain compared to controls and patients with low levels of pain. *Lupus* 2013; 22(11): 1118–1127.
21. Aisiku IP, Penberthy LT, Smith WR, et al. Patient satisfaction in specialized versus nonspecialized adult sickle cell care centers: the PiSCES study. *J Natl Med Assoc* 2007; 99(8): 886–890.