METHODOLOGICAL CONSIDERATIONS RELATED TO EQUITY, DIVERSITY, AND INCLUSION IN CLINICAL EPIDEMIOLOGY

Promoting equity, diversity, and inclusion in surveys: insights from a patient-engaged study to assess long COVID health-care needs in Brazil

Bárbara Caldas[a,*], Margareth Portela[a], Elisabeth Stelson[b,c], Sara Singer[d], Thatiana Amaral[e], Cledir Amaral[f], Claudia Escosteguy[g], Mônica Martins[a], Carla Lourenço Tavares de Andrade[a], Letícia Soares[b], Flora Cornish[h], Meredith Rosenthal[i], Emma-Louise Aveling[i]

[a]Department of Health Administration and Planning, Escola Nacional de Saúde Pública Sergio Arouca, Fundação Oswaldo Cruz, Rio de Janeiro, Brazil
[b]Patient-Led Research Collaborative, Calabasas, CA, USA
[c]Department of Social and Behavioral Sciences, Harvard T. H. Chan School of Public Health, Boston, MA, USA
[d]Department of Health Policy, School of Medicine, Stanford University, Stanford, CA, USA
[e]Center for Health Sciences and Sports, Universidade Federal do Acre (UFAC), Rio Branco, Brazil
[f]Instituto Federal do Acre (IFAC), Rio Branco, Brazil
[g]Epidemiology Service, Hospital Federal dos Servidores do Estado, Rio de Janeiro, Brazil
[h]Department of Methodology, London School of Economics and Political Science, London, UK
[i]Department of Health Policy and Management, Harvard T. H. Chan School of Public Health, Boston, MA, USA

Abstract

Background and Objective: Long COVID (LC) refers to persistent symptoms after acute COVID-19 infection, which may persist for months or years. LC affects millions of people globally, with substantial impacts on quality of life, employment, and social participation. Ensuring access to effective, patient-centered care for LC demands evidence, grounded in inclusive representation of those affected by the condition. Yet survey studies frequently under-represent people with the most disabling disease presentations and racially and socioeconomically marginalized groups. We aimed to describe a patient-engaged approach to developing a survey to inform public LC health care and to assess its implementation in terms of enabling participation by diverse LC patients in Brazil.

Methods: Survey development was iterative, achieved through an interdisciplinary collaboration among researchers including people living with LC, and grounded in 3 guiding principles: (1) evidence-based; (2) inclusive, intersectional, and patient-centered understanding of chronic illness and research participation; and (3) sensitivity to the context of health-care access.

Results: The product of our collaboration was a longitudinal survey using a questionnaire assessing: LC symptoms; their clinical and functional evolution; and impacts on quality of life, household income, health service access, utilization, and out-of-pocket expenses. We illustrate how we operationalized our 3 principles through survey content, instrument design, and administration. Six hundred fifty-one participants with diverse LC symptoms, demography, and socioeconomic status completed the survey. We successfully included participants experiencing disabling symptoms, Black and mixed race participants, and those with lower education and income.

Conclusion: By centering patient experience, our novel, principles-based approach succeeded in promoting equity, diversity, and inclusion in LC survey research. These principles guiding patient-engaged collaboration have broad transferability. We encourage survey researchers working on chronic illness and in other contexts of marginalization and inequality to adopt them. © 2024 The Authors. Published by Elsevier Inc. This is an open access article under the CC BY-NC license (http://creativecommons.org/licenses/by-nc/4.0/).

Keywords: Long COVID; Patient-researcher partnership; Survey research; Equity; Diversity; Inclusion

1. Introduction

Long COVID (LC) affects over 400 million people globally [1], with substantial negative impacts on quality of life, employment, social participation, and costs for health-care systems and economies more broadly [2−4]. Effective care for LC is central to mitigating these costs and avoid

What is new?

Key findings
- Our patient-engaged, international collaboration designed a novel approach to developing a survey to assess long COVID health-care needs in Brazil rooted in the following: (1) diverse forms of evidence; (2) an inclusive, intersectional, and patient-centered understanding of chronic illness and research participation; and (3) context-sensitive understanding of factors shaping health-care access.
- Response and retention rates and sample demographics indicate survey participation was feasible and accessible for diverse participants, including those frequently under-represented in COVID or chronic illness research.

What is added to what is known?
- Our novel, patient-engaged approach demonstrates how to promote equity, diversity, and inclusion in survey research.

What is the implication and what should change now?
- Researcher-driven approach to survey studies should move toward an evidence-based, patient-driven, and context-sensitive approach to promote equity, diversity, and inclusion.
- Partnering with patients at all stages of research is foundational to inclusive research practices.

deepening inequalities, yet patients around the world struggle to access quality, patient-centered health care [5−7]. As scientific understanding of LC advances [8,9], evidence grounded in inclusive representation of those affected by the condition is needed to effectively translate and embed that knowledge in health-care services. It is thus essential that survey studies assessing LC symptoms, health-care needs and barriers include people experiencing the most debilitating symptoms [10,11] and those worst affected by COVID-19 and LC, including as a result of structural harms such as poverty and racism [12,13]. However, survey studies, and COVID research more broadly, frequently under-represent people with the most disabling disease presentations, racially minoritized and other historically marginalized groups facing a high burden of disease [10,11,14−16].

Over 200 potential symptoms have been associated with LC, including fatigue, cognitive dysfunction, postexertional malaise (PEM), joint and muscle pain [4]; frequent diagnoses among people with LC include dysautonomias, myalgic encephalomyelitis, mast cell activation syndrome, as well as organ and tissue damage [8]. Some people with LC experience episodic illness with fluctuating symptoms that follow a relapsing-and-remitting pattern [17]. LC can last from months to years [18], with reported recovery after 2 years as low as 7.6% [19].

In Brazil, like many countries, COVID-19 disproportionately burdened communities already contending with structural and social drivers of ill-health [13,20]. For many with LC, then, effects of the condition are compounded by barriers to accessing health care and socioeconomic precarity (eg, lack of paid time off for informal work) [21,22].

Estimates of LC prevalence in Brazil, available only since 2022, vary from 20% to 80% of people infected with COVID-19 [23−34]. Only a minority of LC studies have evaluated impact on quality of life [30] or functional status [23]; none assessed health-care needs or use, or socioeconomic impacts. To date, no study has provided population estimates for LC symptoms, and no symptom survey has included PEM, reported by a high proportion of people with LC [4,35]. Characterizing diversity in these Brazilian studies is difficult. Only one-third of papers report demographic data beyond age and sex (eg, about race, ethnicity, gender identity, schooling, or income). Those that do suggest significant limitations; for instance, participants of studies adopting an online self-administered questionnaire were >70% highly educated White females [26,33]. Such omissions undermine studies' value for promoting evidence-based equitable health system responses [36].

The aims of this paper are 2-fold: firstly, to describe a novel, collaborative approach to survey development and secondly, to assess the success of this approach in achieving a diverse sample in terms of demographic characteristics, socioeconomic status, and including participants with significantly burdensome symptoms. The survey constituted 1 part of a mixed-methods study designed to contribute evidence regarding the needs of people living with LC and recommendations to inform health care for LC within Brazil's Unified Health System (*Sistema Único de Saúde, (SUS)*). Our approach was rooted in meaningful patient partnership and 3 guiding principles: inclusion of a diverse evidence base; an inclusive, intersectional, and patient-centered understanding of chronic illness and research participation; and sensitivity to the context-specific and complex reality of health-care access. The survey was conceived when there was minimal information on LC in Brazil, a country heavily hit by the pandemic [37]. The questionnaire was administered to a representative sample of patients hospitalized with SARS-CoV-2 in SUS hospitals in the Rio de Janeiro city, using a cohort design to capture changes over time (eg, vaccination rates) (Box 1). Our survey enabled participation by diverse LC patients in Brazil, demonstrating how to promote equity, diversity, and inclusion in survey research.

2. Materials and methods

2.1. A patient-engaged collaborative approach

Our approach followed principles of participatory research [38] to promote quality [39,40] and embed equity in design, conduct, and dissemination [36,38,41]. Collaboration was international, interdisciplinary and included people living with LC. The latter included members of the Patient Led Research Collaborative, an international group of people living with LC who dedicate personal and professional expertise to advance LC research and policy [40]. Involvement of people living with LC in coinvestigator, research associate, and advisory positions influenced all

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Box 1 Key methodological characteristics of our survey study cohort design, population and sampling

Our survey aimed to provide estimates of LC symptoms after discharge; assess clinical and functional evolution of LC symptoms; and assess impacts on quality of life, household income, health service access, utilization, and out-of-pocket expenses. To achieve a sample allowing population estimates among the population of discharged patients hospitalized for COVID-19 in SUS hospitals in Rio de Janeiro city, key features of our study design included.
- Cohort study to capture data retrospectively (eg, data related to prehospitalization period, such as vaccination, comorbidities, quality of life, occupation) and prospectively (eg, clinical evolution, quality of life, changes in occupation and income) in two rounds (waves) of administration.
- Target population: People aged at least 18 years who were discharged from SUS hospitals for COVID-19 in Rio de Janeiro, from December 2020 to November 2022.
- Two-stage sampling approach (hospital and patient-level) designed to achieve statistical representation of the target population: At the hospital stage, we selected hospitals with the highest volume of hospitalizations, and from each hospital targeted a number of patients proportional to that hospital's volume of hospitalizations. At the patient stage, for each hospital, eligible patients were stratified by cohort (24, 18, 12, and 6 months since discharge) and randomly selected.
- Database: The Severe Acute Respiratory Syndrome Surveillance System (SIVEP-Gripe), which aggregates data collected on severe acute respiratory infection cases in Brazil including hospitalizations for COVID-19; we were granted access to the identified database.

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stages of our survey design. Our team of researchers from Brazil, USA, and UK was interdisciplinary (encompassing social psychology, health-care improvement, health economics, social work, community health and mobilization, epidemiology, and various clinical fields), and included academics and practitioners with significant expertise in the local health system.

Over 7 months, we engaged in an iterative process of drafting, reviewing, discussing, and refining survey content, instrument design, and administration (Supplementary file 1). The international team interacted in English; hence Brazilian team members translated the first survey draft into English and reverse translated the final version to Portuguese. Full discussion of the capacities, resources, and skills required to facilitate productive collaboration within such a diverse team are beyond the scope of this paper. Our collaboration was grounded in a commitment to recognition and respect for diverse perspectives and ways of knowing [42]. Nonetheless, this iterative process entailed many robust discussions, and required that the team learn from moments where perspectives clashed [43].

Ethics were a priority in our approach, guiding decisions at every stage. Ethical approval was granted by relevant Institutional Review Boards in Brazil and USA. Brazilian ethical rules did not permit offering compensation for participation. Beyond ethical considerations, 3 guiding principles shaped our decision-making and survey development.

2.2. Principles shaping survey development

2.2.1. Evidence-based

We grounded the survey in evidence that was inclusive of different types of knowledge from diverse locations, including the patient-led research seminal to recognizing and understanding the condition. We used the rapidly growing literature on LC [44] (eg, about symptoms, LC patient experiences) as well as relevant, validated measures not specific to LC (eg, to assess health-related quality of life) in a systematic and transparent way to inform our study [45]. Since much of this evidence is global North−centered, in our case "evidence-based" required synthesizing relevance and significance of this literature for the Brazilian context.

2.2.2. Inclusive, intersectional, and patient-centered understanding of chronic illness and research participation

We aimed to generate findings that could promote accessible, patient-centered LC care which is responsive to individual needs and preferences and to the diversity of identities and living situations that shape these needs [46]. Thus, we drew on the Episodic Disability Framework [17,47] to emphasize a holistic understanding of living with a chronic condition, one which accounts for: fluctuations and unpredictability in symptoms and severity day-to-day and over time [17]; the effects of disability on socioeconomic

inclusion; and the role of larger political, social, economic, and historical factors in shaping illness and health-care experiences. This intersectional lens, attending to interacting identities, social positions, and structural harms, was equally central to recognizing how these factors shape not only vulnerability to and experiences of LC, but may also constitute barriers to research participation that we sought to mitigate in our design [48,49].

2.2.3. Context-sensitive understanding of factors shaping health-care access

Treatment and management possibilities for a chronic condition are contextually contingent and shaped by characteristics of health-care systems and socioeconomic conditions [50]. These factors influence judgments made by patients and professionals about medical intervention and care [51], and furnish or deprive people of the social, cultural, and economic resources needed to access and navigate care, even within a universal public health-care system like SUS [50–53]. From this perspective, information needed to promote patient-centered care for LC extends beyond narrow clinical assessment of symptoms and estimates of their prevalence, requiring context-sensitive understanding of if and how people with LC use and experience health-care services.

3. Results

The product of our iterative collaboration was a longitudinal survey, administered twice, using a questionnaire with 184 questions assessing: LC symptoms; their clinical and functional evolution; impacts on quality of life, household income, health service access, utilization, and out-of-pocket expenses (Supplementary file 2). Administration by interviewers followed a decisiontree such that some questions were contingent on prior responses, for example, fewer questions applied if participants reported not needing health care in the last 6 months. We used the Research Electronic Data Capture platform (REDCap®) to support administration and record results. The first wave was administered November 2022 to August 2023, and a second with a subset of participants from August 2023 to March 2024. Deidentified survey data will be made available in an open access registry once results have been published.

Below we describe how we operationalized each guiding principle through survey content, instrument design, and administration (Table 1), providing representative rather than comprehensive examples.

3.1. Operationalization of our approach

3.1.1. Evidence-based
3.1.1.1. Survey content. We conducted a broad review of the literature regarding LC definitions (eg, WHO [54]), pathophysiology, symptomatology (eg, WHO Post-COVID-19

case report form [54]), impacts (eg, quality of life), and recommended management/health-care services. Aiming to fill knowledge gaps concerning areas considered research priorities among LC patients [55], we also identified areas where peer-reviewed studies were still lacking. For example, patient groups had highlighted changes in symptom severity associated with menses, but this had been largely unacknowledged in published LC research. We also asked about PEM in lay terms (referring to "Worsening of symptoms after previously tolerated physical or mental effort/activity") given high frequency of PEM among LC sufferers and evidence of low diagnosis and awareness of PEM in Brazil.

3.1.1.2. Instrument design. We reviewed validated instruments that, while not LC-specific, assessed symptom severity (eg, DePaul Questionnaire for Post-Exertional Malaise) and effect of LC on functioning (eg, Washington Group Short Set on Functioning) and quality of life (eg, EuroQoL) [56–58]. We considered the intersection of LC symptoms literature and specific instrument items to prevent a biased characterization of LC. For example, to avoiding skewing estimates of depression prevalence among people living with LC we adopted a scale without fatigue as an item assessing depression.

3.1.1.3. Survey administration. We used telephone interviews to achieve the desired sample within the project's timeframe. Telephone interviews are useful for screening/assessing clinical conditions [59,60], generating valid data, contributing to high response rates and participation by the many SUS users who are functionally illiterate [61]. Having a researcher administer the survey (rather than participant self-completion) potentially builds trust in the study team encouraging longitudinal retention, and limiting measurement error by providing opportunities to clarify instrument items.

3.1.2. Inclusive, intersectional, and patient-centered understanding of chronic illness and research participation
3.1.2.1. Survey content. Team members living with LC provided critical insight into topics that needed to be included to capture patients' lived experience accurately and holistically, for example, questions about LC impacts on work and income—key social determinants of health and health-care access. We excluded certain questions that, while of theoretical interest for researchers, could have made participants feel uncomfortable, judged, or stigmatized, for example, we removed questions about alcohol and substance use, originally included for comparability purposes based on widely used lists of health measures. Patient-researchers' personal experiences of protracted journeys to identifying their own LC and of health-care providers discounting LC symptoms, prompted inclusion of the question: "Do you think you have post-COVID syndrome?"

3.1.2.2. Instrument design. One very challenging facet of LC research is accurately capturing the episodic nature of

Table 1. Operationalization of 3 guiding principles across survey content, instrument design, and administration

Survey process element → survey guiding principles ↓	Survey content and topics (i.e., what we asked about)	Survey instrument design (e.g., question format, wording, response options, item order)	Survey administration (e.g., delivery mode, accommodations)

Evidence-based	• Searched the literature to identify questions and instruments that would: (1) elicit LC cases (e.g., Post-COVID-19 CRF); (2) assess impact of LC on functionality (WG-SS), quality of life (EuroQol) or specific body system (mMRC); (3) assess impact of LC on labor; and (4) identify health services needed for patients with LC.
	• Identified gaps in literature to more fully assess in this survey.
	
	  o Included questions to assess changes in symptom severity associated with menses.
	  o Included PEM among surveyed symptoms due to the high frequency of PEM among people with LC.
		• Chose a tool without items that may overlap with LC symptoms and skew prevalence estimates (e.g., adopted PHQ-2 for depression screening instead of PHQ-9).
		• Reviewed instrument versions validated in Portuguese (Brazil) — EuroQol 5D-5 L, Washington Group Short Set on Functioning (WG-SS).
		• Followed best practices for asking about gender identity.
			• Used telephone interviews (adequate for gathering clinical, functional, and social data) that allowed quickly replacing unsuccessful contacts, and supporting questionnaire completion in successful contacts.
			• Interviewer-administered survey allowed for 1) development of participant-researcher trust (decreases social desirability bias and improves longitudinal retention); 2) clarification on any question (reduces measurement error); 3) reduce literacy and cognitive symptom barriers to participation.
			• Included participants with proxy respondents to provide a more complete picture of post-COVID symptoms/conditions in survivors of COVID-19 hospitalization.

Inclusive, intersectional, and patient-centered understanding of chronic illness and research participation	• Avoided survey questions that may inadvertently make participants uncomfortable or feel stigmatized (e.g., drug and alcohol consumption, erectile dysfunction).
	• Assessed important social determinants of health, such as work and financial security, and how this may have changed due to LC.
	• Included open-ended questions to register new diagnoses not listed.
	• Included ''Do you think you have post-COVID syndrome?'' to allow participants to self-identify.
		• Reviewed the order of questions to foster participant's engagement.
		
		  o Started with questions focusing directly on LC as this was the topic motivating participation.
		  o Moved more sensitive topics (e.g., income) to the end when interviewer and participants had established rapport.
		
		• Substituted clinical terms of symptoms/comorbidities for patient-friendly language (e.g., ''hair loss'' instead of ''alopecia'').
		• Symptom frequency assessed to more closely reflect the relapsing-remitting nature of LC.
		
		  o Assessment of symptoms changed from ''1) yes, but not anymore; 2) yes, still present; 3) yes, intermittent; no'' *to* 1) Have never experienced it since getting sick; 2) Used to experience it but not anymore; 3) It still happens some of the time; 4) It still happens most or all of the time.
		  o Assessed variety of sleep difficulties instead of only increase or decrease sleep
			• Options to participate in the interview via telephone, video platform, or in-person to accommodate participants energy, symptoms, and technology access.
			• Participants free to skip any question.
			• Participants encouraged to take breaks or ask to complete the survey at a later time to help avoid symptom exacerbation or fatigue.
			• Decision-tree used to help reduce the number of questions asked based upon earlier responses (e.g., only ask about health-care access if participants endorsed this type of service was needed, otherwise skipped).

(Continued)

Table 1. Continued

Survey process element → survey guiding principles ↓	Survey content and topics (i.e., what we asked about)	Survey instrument design (e.g., question format, wording, response options, item order)	Survey administration (e.g., delivery mode, accommodations)

		• Refined assessment of health-care service needs and utilization
		
		  o Made service assessment more inclusive by adding pharmacy services, alternative medicine, and home health care.
		
		• Symptom questions organized according to how patients may conceptualize their body or similar symptoms to improve recall instead of organized according to clinical assessment categories.
		• Balanced survey comprehensiveness with parsimony so as to not fatigue participants or exacerbate symptoms.
		
		  o Elimination of redundancy (e.g., overlap between EuroQol and WG-SS led to removal of WG-SS).
		  o Assessment of specific specialty care and clinical tests simplified by assessing specialists all together and labs and scans generally since patients may not remember clinical details.
		  o Health-care out-of-pocket expenses focused on general dimensions: medicines, health-care professional visits, and exams.	

Context-sensitive understanding of factors shaping health-care access	• Expanded assessment of social determinants of health, specifically:
	
	  o Fine-tuned positions to differentiate between e.g., paid and unpaid work, full/part-time/seasonal employment.
	  o Assessed changes to employment due to LC.
	  o Assessed changes in income and household composition and who is supported by the income.
	
	• Included viral comorbidities of high prevalence in Brazil (e.g., Zika, Chikungunya, Dengue, and Yellow fever).
		• Provided a ''prefer not to answer'' option for vaccination status question due to sociopolitical climate.
		• Utilized Brazilian National Institute of Geography and Statistics to capture sociodemographic data.
		• Questions about health-care needs, access and use adjusted to Brazilian health-care system context.
		• Fine-tuned wording and item ordering after piloting among SUS patients.
			• Due to limited public awareness of long COVID and barriers to care, interviewer provided participants with long COVID resources based upon participant's expressed need.
			• Option to have proxy of participant (e.g., family member) participate in survey due to high COVID morbidity and mortality rate in Brazil.

(Continued)

Table 1. Continued

Survey process element → survey guiding principles ↓	Survey content and topics (i.e., what we asked about)	Survey instrument design (e.g., question format, wording, response options, item order)	Survey administration (e.g., delivery mode, accommodations)
	• Included a variable to capturing the need/use of social assistance. • Added assessment of barriers to accessing services to inform service provision instead of only assessing whether or not they received service. o Broader assessment of access barriers, including barriers that could be overlooked in the context of a free public health-care system, such as financial costs like transportation or missing work.		

LC, long COVID; PEM, postexertional malaise.

CRF, case report form; mMRC, Modified Medical Research Council; PHQ, patient health questionnaire; WG-SS, Washington Group Short Set.

many symptoms. Building from the DePaul questionnaire [56] format, and iterating among our team, we incorporated the following response options: 1) Have never experienced it since getting sick; 2) Used to experience it but not anymore; 3) It still happens some of the time; 4) It still happens most or all of the time.

3.1.2.3. Survey administration. Research participation can be physically taxing for patients with energy-limiting conditions, risking substantial selection bias or harm to participants. To mitigate this, in addition to repeated deliberations to balance comprehensiveness with parsimony to avoid triggering excess fatigue or PEM, patient-researchers' experiences informed several adaptions in survey administration. For example, we added wording at the start to encourage participants to take breaks, ask to complete the survey at a later time or to complete it over multiple occasions, and administrators were trained to intermittently inquire whether participants felt able to continue.

3.1.3. Context-sensitive understanding of factors shaping health-care access
3.1.3.1. Survey content. Many Brazilians of working age (especially those receiving SUS care) are informally employed, doing seasonal work or working multiple jobs. To appropriately capture LC impacts on work in this context we incorporated response options such as "ad hoc/seasonal hours" and "decreased *number* of jobs." In assessing comorbidities, we included endemic infectious diseases like tuberculosis and sequelae from viral infections (eg, chikungunya, dengue, yellow fever, Zika) which can lead to chronic conditions similar to LC [62]. To ensure context-specific, patient-centered understanding of needs, we assessed both services accessed by participants, and

reasons why participants may not have accessed needed care. We included an open-ended option and questions about specific barriers, for example, feeling emotionally uncomfortable, financial costs like transportation or missing work—access barriers which could be easily overlooked in the context of a "free" public health-care system.

3.1.3.2. Instrument design. Since vaccination status was a sensitive topic given the sociopolitical climate in Brazil during an election, we included a "prefer not to answer" option for this question. We also piloted the survey to get feedback from SUS users about question clarity, which was especially pertinent since the survey had been developed through back-and-forth translation.

3.1.3.3. Survey administration. Due to social and structural disparities SUS users face, and limited LC public awareness, many participants expressed challenges accessing resources they needed to manage LC symptoms, including little awareness of existing specialist LC services. Survey administrators dedicated time at the end of surveys to provide participants with information to help access these services (not during, to avoid priming participants' responses). From our perspective, not only was this ethically important, it also strengthened the study since it bolstered rapport and trust with the research team, potentially enhancing retention.

3.2. Fielding the survey

In 10 months, we reached 1223 of the 2978 patients we attempted to contact. Invalid telephone numbers accounted for most failures to reach a patient. Of those reached, 344 discontinued communication during the recruitment process (eg, stopped replying to texts); 228 declined to participate. A
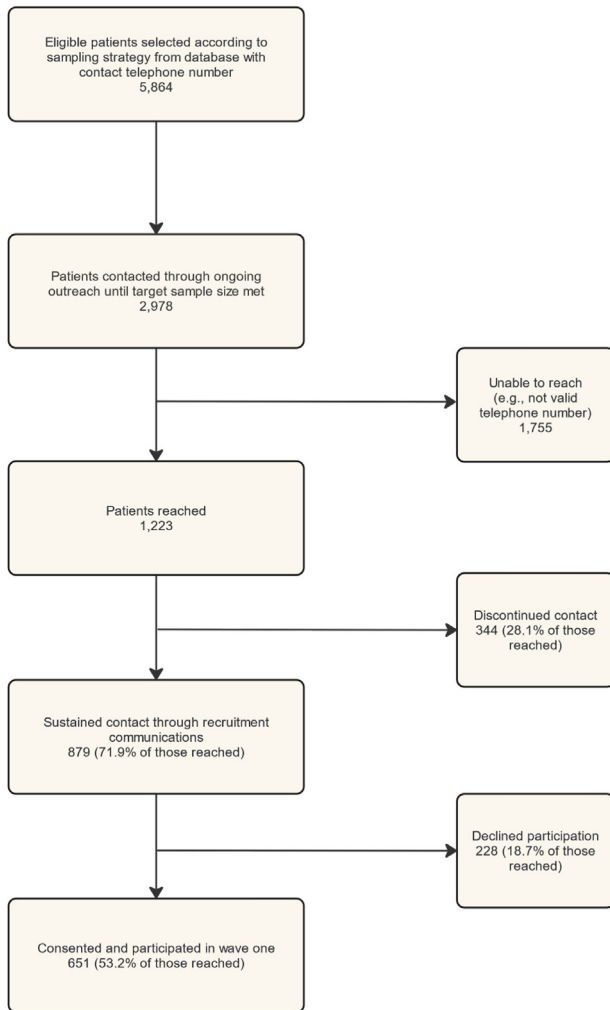
Figure 1. Flow chart of recruitment of participants—Wave 1. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

total of 651 (53.2%) patients or their proxy completed the survey in the first wave (Fig 1); average duration 33 minutes. In the second wave, we conducted 340 interviews out of 447 invited participants (retention rate 76.1%) (Fig 2). This sample met the sampling strategy requirements to enable population estimates; we discuss below our success in terms of relevant dimensions of diversity in the final sample of patients who completed the survey (Table 2).

4. Discussion

We described a patient-engaged approach to developing a survey that enabled participation by diverse LC patients in Brazil. Our approach achieved success on multiple dimensions.

4.1. Feasible

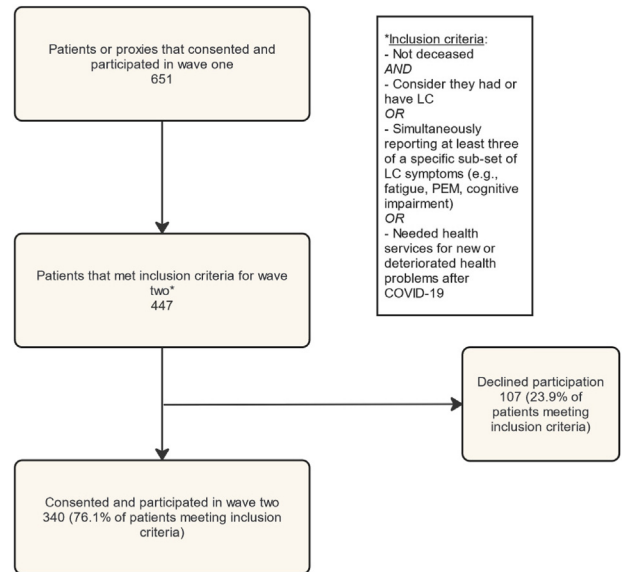Response rates compare favorably with surveys performed in the UK and US, and in Brazil where response

Figure 2. Flow chart of recruitment of participants—Wave 2. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

rates range from 22% to 81% [30,31,34]. Our high retention rate (76.1%) underscores the success of our approach to developing survey content, instrument design, and administration, particularly given the survey's relatively long duration. Though some participants perceived the questionnaire as long, many expressed satisfaction in contributing to research to help SUS users while others welcomed the opportunity for a sustained conversation with researchers.

4.2. Accessible

Our approach facilitated participation by people experiencing high symptom burden, including symptoms impeding survey completion, for example, 60% reported fatigue and 54% reported cognitive impairment. Some of these participants described realizing they might be experiencing LC symptoms only after taking the survey, potentially due to limited attention to LC in Brazil.

4.3. Inclusive

In line with our principle of context sensitivity and since there is no standardized criterion for assessing diversity in the Brazilian context, we assessed diversity in dimensions relevant to the context and purpose of our study. Thus we prioritized diversity in terms of symptoms (as described above) and demographics reflecting the population of SUS users in Rio. We succeeded in recruiting a diverse sample (Table 2) on these terms. The demographic composition of SUS users contrasts with private health insurance users, where white ethnicity and advanced education are over-represented [20]. Our sample included 5.1% with no schooling and 45% with middle school education or less, suggesting successful inclusion of people who at risk of

Table 2. Demographic characteristics of participants

Variable	Sample (n = 651)	
	n	%
Age at hospital admission		
18 to 39	73	11.2
40 to 49	104	16.0
50 to 59	154	23.7
60 to 69	146	22.4
70 to 79	107	16.4
80+	67	10.3
Gender		
Cisgender woman	319	49.0
Cisgender man	329	50.5
Transgender woman, transgender man, or nonbinary people	0	0.0
Other gender identities not listed	1	0.2
Preferred not to answer	2	0.3
Race/color		
White	248	38.1
Black	98	15.0
Mixed race	291	44.7
Asian	4	0.6
Indigenous	4	0.6
Preferred not to answer	6	0.9
Education		
No school	33	5.1
Middle school uncompleted	165	25.3
Middle school	128	19.7
Graduated High School or equivalent	251	38.6
Bachelor's degree	53	8.1
Postgraduate degree	7	1.1
Unknown	12	1.8
Occupation (by the time of interview)[a]		
UNPAID Domestic/caregiving worker	37	6.7
PAID Domestic/caregiving worker	14	2.5
Private sector employee	86	15.5
Public sector employee	16	2.9
Self-employee	114	20.5
Informal worker	11	2.0
Student	4	1.0
Retired/Receiving pension	198	35.6
Unemployed	76	13.7
Income (by the time of interview) (Brazilian real monthly per capita)[a]		
<200	15	2.7
200 to 637	130	23.4
638 to 999	79	14.2
1000 to 1499	109	19.6
1500 to 2000	57	10.3
2000 to 2999	53	9.5
≥3000	26	4.7
Preferred not to answer	87	15.7

[a] Only to those who were alive by the time of interview (556).

exclusion due to literacy issues had we used self-administered electronic questionnaires [61]. We also reached a geographically distributed population, including areas of high social vulnerability and violence within Rio. Diversity in our sample compares favorably with other Brazilian LC surveys [26,27,33], in terms of inclusion of Black and mixed race participants and those with lower education and income, although not with regard to gender diversity.

4.4. Opportunities

Fielding the survey highlighted potential improvements. Following successive iterations and deliberations, we ultimately missed an opportunity to directly ask about severity of symptoms "when at their worst" given participants may complete a survey when feeling relatively well. The questionnaire also lacked an option to report using the same type of health service *both* through SUS *and* the private sector, a particularly common pattern for pharmacy services. Income information remained sensitive, even though placed at the end, with 15.7% preferring not to answer. While telephone-based administration offered valuable advantages, frequent changes of mobile numbers and obsolescence of landlines necessitated many contact attempts and significant loss of potential participants. Finally, while our process made strides toward enabling transformative partnerships [42], we acknowledge we could have included additional perspectives not on our team. Analysis of the full survey results will provide important opportunities to examine the impact of greater equity, diversity and inclusion on the scientific contributions from those data.

4.5. Broader applicability of our approach for equity, diversity and inclusion in research

We suggest the principles guiding our study set a new standard for inclusivity in survey research, and have broad transferability [36] to research in other contexts, particularly involving patients with other complex conditions or traditionally excluded from research. Most fundamentally, tackling equity, diversity, and inclusion in research means being prepared to change "standard practice" and power inequalities in the research process [63,64]. Our approach of foregrounding patient experience at every decision point in study design can be adopted by researchers in other contexts. We benefitted from diverse collaborators willing to confront and work through differences [43] and grounded our work in a diverse evidence-base; an inclusive, intersectional, and patient-centered understanding of chronic illness and research participation; and sensitivity to contextually specific complexities of ensuring equity in health-care access—all widely applicable principles. Our approach offers a means of responding to the critical calls for much greater community engagement in responses to public health emergencies like the COVID pandemic, premised as our approach is on recognizing the legitimacy and the

value of diverse local knowledge [65,66]. The specific practices we developed to mitigate risks that patients' symptoms would limit their survey participation are widely replicable for research with patients with energy-limiting conditions, episodic conditions, and disabilities. The practices designed to be inclusive to those marginalized through low literacy or competing socioeconomic demands are also widely replicable for research in contexts of marginalization and inequality beyond Brazil.

4.6. Limitations

Our study was not registered, although we sought to publish the protocol. We have not used a reporting guideline as we were unable to identify (via the EQUATOR network) a guideline relevant to a methodological paper reporting a novel, collaborative survey development approach.

5. Conclusion

By centering the patient experience, we developed a survey that enabled participation by diverse respondents in Brazil, in a challenging context of participants experiencing disabling symptoms, and in a country marked by socioeconomic, racial, and health inequalities. Standard survey methodologies are often informed primarily by researcher perspectives rather than patient perspectives. We have argued that centering patients can help to produce more equitable and inclusive research. We encourage survey researchers in partnership with patients to explore using the 3-fold principles of being evidence-based, inclusive, and context-sensitive, to guide their survey design to enhance equity, diversity, and inclusion.

Patient involvement

Leticia Soares and Elisabeth Stelson are members of the Patient-Led Research Collaborative, a group of LC patients and patients with associated illnesses such as Myalgic encephalomyelitis/chronic fatigue syndrome and Postural Orthostatic Tachycardia Syndrome, who are also researchers. The mission of the group is to "facilitate patient-led and patient-involved research into LC and associated conditions while following rigorous research methodology, and to advocate for policies that enable patients, particularly the most marginalized, to access care and live with dignity".

CRediT authorship contribution statement

Bárbara Caldas: Writing − review & editing, Writing − original draft, Methodology, Funding acquisition, Conceptualization. **Margareth Portela:** Writing − review & editing, Writing − original draft, Supervision, Project administration, Methodology, Funding acquisition, Data curation, Conceptualization. **Elisabeth Stelson:** Writing − review & editing, Writing − original draft, Methodology. **Sara Singer:** Writing − review & editing, Methodology, Funding acquisition, Conceptualization. **Thatiana Amaral:** Writing − review & editing, Methodology, Conceptualization. **Cledir Amaral:** Writing − review & editing, Methodology, Conceptualization. **Claudia Escosteguy:** Writing − review & editing, Methodology. **Mônica Martins:** Writing − review & editing, Methodology. **Carla Lourenço Tavares de Andrade:** Writing − review & editing, Methodology. **Letícia Soares:** Writing − review & editing, Methodology. **Flora Cornish:** Writing − review & editing, Methodology, Conceptualization. **Meredith Rosenthal:** Writing − review & editing, Supervision, Methodology, Funding acquisition, Conceptualization. **Emma-Louise Aveling:** Writing − review & editing, Writing − original draft, Supervision, Project administration, Funding acquisition, Conceptualization.

Declaration of Generative AI and AI-assisted technologies in the writing process

The authors declare that they have not used generative AI and AI-assisted technologies in the writing process.

Data availability

Data will be made available in a repository after publication of results.

Declaration of competing interest

Acknowledgments

Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jclinepi.2024.111423.

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