Participatory Action Research on Canadians with Alternatively Diagnosed Lyme Disease: Feasibility Study

Feasibility Report

July 2023

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"If we're going to fight a disease, let's fight one of the most terrible diseases of all, indifference." - Robin Williams as Patch Adams

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ADLD PROJECT

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Outline

This feasibility study provides background information on the problem of Lyme Disease (LD) underestimations in Canada, as well as the Federal Framework recommendation to collect human health data for Canadian residents "who do not meet the case definition for probable or confirmed LD, but who experience various symptoms consistent with LD or similar ailments." This is followed by a summary of how the Public Health Agency of Canada (PHAC) assessed the agency's progress in implementing the recommendation over a four-year period beginning in 2017 and ending in 2021.[1] The study then provides a brief description of the mandate issued by PHAC to the consultant team by the end of 2021. It describes the methodology followed in assessing the feasibility of PHAC's proposed initiative, which is to use a Participatory Action Research (PAR) approach to improve knowledge and inform future public health actions for Canadians diagnosed with Lyme disease (LD) via alternative means. The overall conclusions of the report are based on interviews with PHAC'S external partners and discussions of feasibility and success factors. They are organized by three sets of interview questions: project goals, rules of engagement, and data collection methods. The conclusion summarizes the study and offers suggestions on how to proceed with the project's next phase.

[1] Government of Canada, *Federal Framework on Lyme Disease, Report to Parliament*, Ottawa, May 2017.

Background

LD underestimations

Lyme disease (LD) is a vector-borne infectious disease that is endemic or emerging in many parts of Canada, with 17,080 reported cases across the country between 2009 and 2022[2] and a current estimate of 300,000 cases per year in the United States alone, according to the Centers for Disease Control and Prevention (CDC)[3]. The illness is caused by the *Borrelia burgdorferi* bacteria. It is primarily transmitted through the bite of infected blacklegged ticks (also known as deer ticks) and western blacklegged ticks, which are now becoming more common due to a variety of factors such as climate change and the migration or dispersal of host animals (e.g., deer, mice, and birds). The number of LD cases has increased significantly since PHAC started getting surveillance data in December 2009. Anaplasmosis and babesiosis are two other tick borne diseases that are emerging and likely to become more common in the future.

The prevention and control of LD is a federal responsibility. The Federal Framework on Lyme Disease Act[4], passed by the Government of Canada in 2014, requires the federal government to develop a framework and action plan that includes the following pillars:

1. **Surveillance**: The establishment of a national medical surveillance program to use data collected by the Public Health Agency of Canada to properly track incidence rates and the associated economic costs of Lyme disease.
2. **Education and Awareness**: The creation and distribution of standardized educational materials related to Lyme disease, for use by any public health care provider within Canada, designed to
3. **Increase national awareness** about the disease and enhance its prevention, identification, treatment and management.

[2] Government of Canada, Lyme disease: Surveillance, www.canada.ca/en/public health/services/diseases/lyme-disease/surveillance-lyme-disease.html
[3] Centers for Disease Control and Prevention, Lyme Disease, Data and Surveillance, www.cdc.gov/lyme/datasurveillance/index.html
[4] Government of Canada, *The Federal Framework on Lyme Disease Act*, Ottawa, December 2014.

4. **Guidelines and Best Practices**: The establishment of guidelines regarding the prevention, identification, treatment and management of Lyme disease, and the sharing of best practices throughout Canada.

PHAC's Lyme Disease in Canada — A *Federal Framework*[5], published in May 2017, recommends actions to increase LD awareness among Canadians and front-line health professionals, as well as to promote consistent diagnostics across the country. These actions are grouped under two pillars: education and awareness, and guidelines and best practices. Other recommendations are part of the "health surveillance" pillar. They include supporting consistent national reporting, tracking new human infections of LD, and monitoring geographic risk areas in Canada. Specific actions aim to

- Integrate and disseminate innovative methods and best practices for health surveillance among an expanded group of partners;
- Perform an analysis of the costs associated with Lyme disease; and
- Develop a national tick-borne surveillance system that includes Lyme disease and other possible co-infections.

This feasibility study addresses a fourth recommendation that falls under the same heading (Item 1.2), which is to collect "health surveillance data" from Canadian residents "who do not meet the case definition for probable or confirmed LD, but who experience various symptoms consistent with LD or similar ailments" (hereafter referred to as "ADLD patients"). Despite the negative diagnosis they receive, a significant number of Canadians report or continue to experience symptoms consistent with the disease. Because they do not meet the case definition criteria, many seek diagnosis outside of Canada or through other alternative methods. There are currently no mechanisms in Canada to assess the number, demographics, disease history (i.e., tick exposure, laboratory test results, medical trajectory, reported symptoms, and their evolution over time), and life circumstances of ADLD patients. Real-world human health data are required to better

ADLD SURVEILLANCE GAP

Real-world human health data are required to better reflect the number, circumstances, and experiences of Canadians diagnosed with LD using alternative methods.

[5] Government of Canada, *Lyme Disease in Canada, A Federal Framework*, Ottawa, May 2017.

reflect the number, circumstances, and experiences of Canadians diagnosed with LD using alternative methods.

Long-standing concerns about the accuracy of Lyme disease surveillance in Canada prompted the initiative. These concerns are shared by well-informed patient advocates, researchers, health care professionals, and subject matter experts. There is strong evidence that current public health surveillance undercounts the number of new cases (incidence) and even more so the number of existing cases (prevalence) of LD across the country. Only 10% of cases were reported in the United States between 2008 and 2013, according to estimates. From 1997 to 2008, only 65% of cases were reported in British Columbia.[6] In comparison to the southern Québec Montérégie region (7.3 cases per 100,000 in 2017), the stated LD incidence rate for neighboring US states is up to fifteen times higher.[7]

The existing scientific literature suggests several possible explanations for these significant discrepancies and related issues with official LD data reporting systems. They are described below.

1. Case definition. Even though LD has been nationally reportable in Canada since 2009, provinces and territories only report cases that meet the national case definition to PHAC. Reportable cases are based on guidelines provided by the Infectious Diseases Society of America (IDSA), which differ significantly from those developed by the International Lyme and Associated Disease Society (ILADS) and used by some Canadian front-line health professionals. The IDSA case definition is supported by medical colleges and the Association of Medical Microbiology and Infectious Diseases Canada (AMMI Canada), and the Canadian medical community is expected to follow it. Using these guidelines, a case of LD is confirmed through laboratory detection of B. *burgdorferi* bacteria, by PCR or an immunoglobulin response after exposure in a high-risk area (identified through passive and active tick surveillance). Alternatively, confirmation can be obtained through clinical evidence combined with two things: a history of residence in or visit to an LD risk area, and laboratory evidence

LD UNDERESTIMATION

The existing scientific literature suggests several possible reasons for these important discrepancies and related issues with official LD data reporting systems.

[6] Ontario Agency for Health Protection and Promotion (Public Health Ontario). Lyme disease human surveillance in Ontario: a systematic review. Toronto, ON: Queen's Printer for Ontario; 2016.
[7] INESSS, Maladie de Lyme — Stades localisé et disséminés, Situation actuelle et accompagnement vers le changement, Gouvernement du Québec, mai 2019.

of infection in the form of a positive serologic test using the two-tiered approach (screening ELISA followed by an immunoblot assay). The case is labelled “probable” when the evidence is limited to either positive laboratory results or clinical observations of an erythema migrans (EM) rash.[8]

Despite these common standards, the criteria for classifying and counting LD cases for public health surveillance differ by country. In the United States, the CDC monitors “suspected” cases of LD based on clinical observations of an erythema migrans, regardless of other clinical observations, evidence of tick exposure, or laboratory test results. To count the number of LD patients in Canada, public health officials use only "confirmed" and "probable" cases.

2. Surveillance vs diagnosis. Another source of underreporting is the distinction made between guidelines for surveillance reporting and those used for clinical diagnosis and administrative purposes. In the United States, commercial insurance records show significant differences between the number of LD cases that are reported and the number of people who are being treated for it. Since the same phenomenon is observed In Canada, PHAC has decided to assist in the expansion of a pilot study of administrative health records in Manitoba, in the hopes of better capturing cases missed by the Canadian Notifiable Disease Surveillance System.

3. Administrative burden and professional risk. Many physicians fail to report LD cases due to their limited knowledge of the illness and the burden of properly completing the appropriate forms and obtaining laboratory tests. As a result, official LD statistics do not include many cases of early (localized) infection and diagnoses made without serological confirmation. Physicians are also hesitant to diagnose and treat Lyme disease due to the penalties and professional consequences of not conforming with the reference guidelines.

4. Laboratory testing. More importantly, the underestimation of LD cases comes from the fact that the illness is hard to detect and that standard diagnostic methods have their limits and flaws. Laboratory testing is dependent on the immune system’s response, which is weak in both the early and advanced infection phases. Borrelia’s ability to evade detection by the immune system results in false negatives on the conventional ELISA and Western blot laboratory tests.

[8] Centres for Disease Control and Prevention, National Notifiable Diseases Surveillance, ndc.services.cdc.gov/case-definitions/lyme-disease-2022/

5. Complex infection. The fact that LD is a multisystemic bacterial infection with multiple strains and diverse manifestations further complicates detection. It can mimic a wide range of illnesses and produce symptoms that are frequently misdiagnosed as fibromyalgia, chronic fatigue, and depression, among others. More confusion sets in when the disease persists even after treatment and causes primary infections together with a slew of opportunistic co-infections (bacterial, viral, fungal, parasitic). Secondary infections then take advantage of a weakened immune system, attack the central nervous system, and cause multiple symptoms that combine in different ways for each person. No two patients are alike, and most doctors have difficulty diagnosing the disease. Some may also be reluctant to diagnose patients with CLD (Chronic Lyme Disease) and accept long-term responsibility for their treatment. CLD symptoms appear during the disease's disseminated and late stages, as part of what the CDC refers to as the post-treatment Lyme disease syndrome. ILADS defines CLD as "a multisystem illness with a wide range of symptoms that are either continuously or intermittently present for a minimum of six months. CLD is the result of an active and ongoing infection by any of several pathogenic members of the *Borrelia burgdorferi* sensu lato complex."[9]

A COMPLEX BACTERIAL INFECTION

The fact that LD is a complex bacterial infection with multiple strains and diverse manifestations further complicates detection.

6. Tick vs LD surveillance. Because of the complexity and ambiguity that pervade virtually all discussions of LD, many researchers have abandoned the idea of a comprehensive tick-borne disease surveillance system and opted instead to strengthen tick surveillance activities across the country. Research conducted by the Canadian Lyme Disease Research Network (CLyDNR), funded by PHAC and the CIHR, focuses on closing critical knowledge gaps through scientific research that may improve LD prevention, diagnosis, and treatment in Canada. However, surveillance studies

[9] See https://www.ilads.org/evidence-based-definition-of-chronic-lyme-disease-published-in-antibiotics journal/

of ADLD incidence and prevalence involving the ongoing, systematic collection, analysis, and interpretation of health-related data are not part of the network's stated mission or mandate.

The disease's elusive and resilient nature, as well as detection failures, make it difficult for Canada to provide the comprehensive and personalized treatments that patients require. This has led many undetected, misdiagnosed, and mistreated patients with a highly debilitating and perplexing illness to seek a clearer diagnosis through other means, such as self-diagnosis or testing in Lyme specialty laboratories in the United States. Others do their own research and seek advice from health-care practitioners who use the ILADS guidelines or the Horowitz Lyme-MSIDS Questionnaire to estimate the likelihood of having LD. Many patients with ADLD are stigmatized and forced to move from one specialist to another, especially in the late stages of the Lyme disease. Abandoned by the Canadian health system, they spend large sums on private care from doctors outside of Canada (mostly in the United States), Lyme-literate specialists and physicians, physical therapists, naturopaths, and other complementary and alternative health practitioners.

MEDICAL WANDERING AND STIGMATIZATION

Many patients with ADLD are stigmatized and forced to move from one specialist to another, especially in the late stages of the Lyme disease. Abandoned by the Canadian health system, they spend large sums on private care from doctors outside of Canada (mostly in the United States), Lyme-literate specialists and physicians, physical therapists, naturopaths, and other complementary and alternative health practitioners.

Federal framework

The PHAC Office of Audit and Evaluation evaluated the effectiveness of LD Framework activities from May 2017 to March 2021. The report was published in January 2022, and a report to Parliament was issued a few months later (May 2022). Overall, PHAC's progress towards meeting its obligations under the Framework was deemed uneven and slow in some critical areas. This is due to several factors, the most significant of which are a lack of dedicated funding, issues with scientific uncertainty, and the disruption caused by PHAC's concurrent work on the COVID-19 response.

The need for data on Canadians with ADLD remains largely unmet. The difficulty that subject experts face in agreeing on the best way to measure these cases, as well as the lack of baseline rules to follow, are major impediments to progress in this area. According to the evaluation report, PHAC investigated the possibility of establishing a voluntary LD patient registry. However, the idea was abandoned because of concerns about data security and privacy, as well as disagreements on the best way to collect and manage patient data.

CHALLENGES IN DEVELOPING PARTNERSHIPS

PHAC faces even greater difficulties in engaging and forming formal partnerships with people with lived experience, their representatives, as well as patient-aligned researchers and physicians. This is especially true for activities other than education and public awareness campaigns.

The Framework evaluation also addressed issues of external engagement and collaboration. In Canada, several federal, provincial, territorial, and international regulatory agencies must work together to gather LD health surveillance information. Coordinating efforts across multiple jurisdictions is subject to several limitations and long delays. There is also disagreement about the role of the agency in providing leadership and guiding collective actions. However, PHAC faces even greater difficulties in engaging and developing formal partnerships with people with lived experience, their representatives, as well as patient-aligned researchers and physicians. This is especially true for activities other than education and public awareness campaigns. Despite direct contact with these groups, collaborative efforts have been limited to information sharing, inviting people with lived experience to tell their stories, soliciting input and feedback, facilitating roundtable discussions, and hosting multistakeholder consultation events.

The Framework evaluation report discusses issues of stakeholder engagement at some length. Key findings highlight PHAC's challenges in effectively engaging with LD patients, patient groups, their advocates, and some LD specialists. The same concerns were expressed even more strongly in many interviews conducted as part of this feasibility assessment. They are addressed in the report's final section, which outlines the feasibility conditions for launching the ADLD project as well as the factors critical to its successful development and implementation.

Feasibility study mandate

Post-framework activity. PHAC's activities for the Framework and Action Plan ended in 2022. The Policy Integration and Zoonoses Division of CFEZID (Centre for Food-borne, Environmental, and Zoonotic Infectious Diseases) continues to work on the Framework goals, led by the Domestic Vector-borne Diseases Technical Team. Their work is done in collaboration with the National Microbiology Lab (NML) and the Infectious Disease and Climate Change Fund (IDCCF).

Terms of reference. PHAC sought an external contractor in the winter of 2019 to investigate the feasibility of using Participatory Action Research (PAR) to collect information about Canadians with ADLD. Its request for proposals was inspired by the Framework's recommendation to explore "innovative methods and best practices for human surveillance among a wider group of partners." The decision to hire an independent and experienced PAR consultant team stemmed from the realization that more efforts were needed to build trust and bring about greater respect between LD patient communities and the Public Health Agency of Canada.

Following delays caused primarily by the COVID-19 pandemic, a contract with the following title was signed in November 2021: A *Participatory Action Research study to improve knowledge and inform future public health actions for Canadian residents diagnosed with Lyme disease (LD) through alternative means but who do not meet the LD national surveillance case definition*. The contract specifies that the goal of the initiative is not to review the national case definition or the transmissibility of LD in Canada. Nor does it aim to settle disagreements over LD treatment protocols.

PROJECT TITLE

A Participatory Action Research study to improve knowledge and inform future public health actions for Canadian residents diagnosed with Lyme disease (LD) through alternative means but who do not meet the LD national surveillance case definition.

The consultant team's mandate begins with Phase 1 of the project, which focuses on determining the feasibility of using PAR methods to collect meaningful data on Canadians diagnosed with LD through alternative means, including non-reference laboratory testing in Canada or abroad. Long-term goals include learning more about people with ADLD who fall outside the national LD surveillance system, with a view to guiding future public health, education, and awareness efforts in Canada. PAR is a novel multi stakeholder approach to human health research where all interested parties can work together in designing, overseeing, and conducting the necessary data collection activities in ways that are relevant to them.

To achieve the desired goals, the overall project will proceed through four phases.

Phase 1. This study meets the requirements of Phase 1. It provides a feasibility assessment of

- The basic goals and limitations of the project;
- The potential interest and involvement of PHAC's external partners (people with lived experience, patient groups, researchers, health care professionals, medical authorities, and public health agencies across Canada) in designing and conducting project activities;
- Possible steps and mechanisms for stakeholder engagement;
- Some further planning of Phase 2.

MULTI-STAKEHOLDER ENGAGEMENT

PAR is a novel multi-stakeholder approach to human health research where all interested parties can work together in designing, overseeing, and conducting the necessary data collection activities in ways that are relevant to them.

Following the completion of the feasibility assessment, additional assistance from the consultant team may be required in the process of designing and carrying out the project in a participatory mode.

Phase 2. If the project is deemed feasible and there is a shared desire to move forwards, Phase 2 activities will develop planning and build consensus on the exact nature of the engagement process, the information that should be sought, the best methods for data collection, and possible innovations based on PAR principles, those of partnering in research to improve knowledge that guides public health action. Issues to be addressed collaboratively will include:

- The structure, composition, and procedures of the committee(s) responsible for designing and conducting project activities;
- The key question(s) pertaining to ADLD and the data required to answer them;
- The data collection process and rules of informed consent, confidentiality, and security;
- The analysis protocol and rules of data access;
- A critical path and timeline for the overall project;
- The cost and resources needed to achieve project goals;
- Some further planning of Phase 3.

Phases 3 and 4. If Phase 2 is successful, the project team will move on to implementing the project plans (Phase 3). This will be followed by a final evaluation and knowledge translation of project findings (Phase 4).

Methodology

Invitations and interviews

1. The project's first phase is a feasibility study, which began with a review of published reports, papers, and reference websites about LD surveillance in Canada and other affected countries. Key documents consulted include, but were not limited to the following:
    - Cartter ML, Lynfield R, Feldman KA, Hook SA, Hinckley AF. Lyme disease surveillance in the United States: Looking for ways to cut the Gordian knot. Zoonoses Public Health. 2018 Mar;65(2):227-229. doi: 10.1111/zph.12448. PMID: 29431297.
    - Community Based Participatory approach is used by Lymedisease.org, a non-profit advocacy association based in USA that hosts on its web site an interface named “MyLymeData”, a new survey tool that tracks patient progress over time (patients diagnosed with Lyme disease in USA initially and extension to other countries is planned). It allows patients to register and pool diagnosis and treatment experiences. A summary of the MyLymeData is available at https://www.lymedisease.org/mylymedata/. https://www.lymedisease.org/assets/about-my lyme-data.pdf
    - Boudreau, Corinne R, Vett K Lloyd, and Odette N Gould, Motivations and Experiences of Canadians Seeking Treatment for Lyme Disease Outside of the Conventional Canadian, Health Care System, Journal of Patient Experience, 2018, Vol. 5(2) 120-126
    - Dubié, Jeanine et al, D’INFORMATION DÉPOSÉ en application de l’article 145 du Règlement par la Commission des affaires sociales en conclusion des travaux de la mission sur la maladie de Lyme : améliorer la prise en charge des patients, Paris, 7 juillet 2021
    - Fournier, Lucie et al, Épidémiologie de la borréliose de lyme en médecine générale, France métropolitaine, 2009-2016, BEH 19 juin 2018
    - Henry B, Roth D, Reilly R, MacDougall L, Mak S, Li M, Muhamad M. How big is the Lyme problem? Using novel methods to estimate the true number of Lyme disease cases in

British Columbia residents from 1997 to 2008. Vector Borne Zoonotic Dis. 2011 Jul;11(7):863-8. doi: 10.1089/vbz.2010.0142. Epub 2011 Mar 17. PMID: 21413887.

- INESSS, Du diagnostic au traitement de la maladie de Lyme aux stades localisé et disséminés Rapport en soutien aux outils d'aide à la décision clinique sur le diagnostic et le traitement, Gouvernement du Québec, Mai 2019
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- Ogden NH, Bouchard C, Badcock J, Drebot MA, Elias SP, Hatchette TF, Koffi JK, Leighton PA, Lindsay LR, Lubelczyk CB, Peregrine AS, Smith RP, Webster D. What is the real number of Lyme disease cases in Canada? BMC Public Health. 2019 Jun 28;19(1):849. doi: 10.1186/s12889-019- 7219-x. PMID: 31253135; PMCID: PMC6599318.
- Rutz H, Hogan B, Hook S, Hinckley A, Feldman K. Impacts of misclassification on Lyme disease surveillance. Zoonoses Public Health. 2019 Feb;66(1):174-178. doi: 10.1111/zph.12525. Epub 2018 Sep 21. PMID: 30242983.
- Septfons A, Goronflot T, Jaulhac B, Roussel V, De Martino S, Guerreiro S, Launay T, Fournier L, De Valk H, Figoni J, Blanchon T, Couturier E. Epidemiology of Lyme borreliosis through two surveillance systems: the national Sentinelles GP network and the national hospital discharge database, France, 2005 to 2016. Euro Surveill. 2019;24(11)
- Shing E, Wang J, Khoo E, Evans GA, Moore S, Nelder MP, Patel SN, Russell C, Sider D, Sander B. Estimating direct healthcare costs attributable to laboratory-confirmed Lyme

disease in Ontario, Canada: A population-based matched cohort study using health administrative data. Zoonoses Public Health. 2019 Jun;66(4):428-435. doi: 10.1111/zph.12560. Epub 2019 Jan 21. PMID: 30665259.

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- White J, Noonan-Toly C, Lukacik G, Thomas N, Hinckley A, Hook S, Backenson PB. Lyme Disease Surveillance in New York State: an Assessment of Case Underreporting. Zoonoses Public Health. 2018 Mar;65(2):238-246. doi: 10.1111/zph.12307. Epub 2016 Sep 10. PMID: 27612955.

2. The consultant team then prepared announcements and invitations to participate, which were distributed via email to the PHAC's *Lyme and other tick-borne diseases email subscription list*. The PHAC subscription list now has about 875 members, the majority of whom are public health professionals, researchers, physicians, and people who have lived experience.
3. More detailed messages were sent out to participants who expressed an interest in the project, along with information about the note-taking process and a privacy notice approved by the Privacy Management Division.
4. Following the initial announcements, PHAC sent out a second round of invitations to 15 key stakeholders who were not reached through the subscription list but might want to share their thoughts about the project and possibly contribute to its future development.
    - Seven accepted to participate.
    - Another 6 did not respond (3 health professionals supporting patient groups, 2 researchers, and 1 official medical association)

- 1 declined the invitation but referred to someone else. Self-identified participants reached through the subscription list encouraged an additional number of interested parties to participate.

5. The consultant team also communicated directly with a few additional specialists and subject experts, using publicly available contact information.
6. The interview guide was distributed in advance and revolved around the following questions:
    - What do you think of the proposed initiative to gather meaningful information on Canadian residents with ADLD who fall outside the national LD surveillance system?
    - How do you feel about the idea of using a collaborative approach for this initiative?
    - Do you have any suggestion on ways to set up a collaborative approach for this initiative?
    - In your view, what would be the most effective ways of gathering meaningful information for Canadian residents with ADLD?

    Rather than asking participants to respond to a pre-determined plan of action, the questions were kept broad so that interviewees could express general support or raise objections and concerns about the initiative.

    The interviews allowed for questions directed at the consultant team as well as two-way discussions about possible rules of engagement and data collection methods.

7. This feasibility study also draws on follow-up discussions with several patient advocacy and support groups who needed time to discuss the proposed initiative among themselves and with the LD community before responding to the interview questions. The questions proposed by the consultant team became the subject of multilateral discussions, which helped clarify the key conditions and success factors for meeting project goals.

Scope and reach

A total of 39 interviews were conducted via videoconference in either French or English, each lasting 30 to 60 minutes. Throughout the first three months of 2023, larger discussions among LD community members and subject experts working with them shaped some of the ideas expressed in these interviews. All had expert knowledge or high levels of literacy with regards to LD issues. They include 18 people with lived experience, 11 representatives of patient groups, 4 doctors and

clinical microbiologists, 3 researchers, 2 public health officials, and 1 veterinarian. Together, they represent a broad range of non-PHAC stakeholders involved in LD-related activities.

Many of the participants expressed views that reflect the various roles they play and combine in real life. Some LD researchers, for example, are also patients with confirmed or alternatively diagnosed LD. For reasons that remain unknown, 8 patients and 6 medical professionals did not respond to follow-up meeting requests sent out by the consultant team. It should be noted that many individuals on PHAC's *Lyme and other tick-borne diseases email subscription list* did not respond to the general invitation to be interviewed. Some patient group representatives declined direct invitations to participate, preferring to share their perspectives with other patient associations and advocates engaging in the consultation process.

RANGE OF PEOPLE INTERVIEWED

All interviewees had expert or high levels of literacy with regards to LD issues. They include 18 people with lived experience, 11 representatives of patient groups, 4 doctors and clinical microbiologists, 3 researchers, 2 public health officials, and 1 veterinarian. Together, they represent a broad range of stakeholders engaging in LD-related activities.

The consultant team did not conduct interviews with representatives of PHAC and other federal ministries that share responsibility for the prevention and control of Lyme disease. Also, while they expressed an interest in the project, representatives of medical associations such as AMMI (Association of Medical Microbiology and Infectious Disease Canada) and the College of Physicians and Surgeons from each province were unavailable for interviews. Given the exploratory nature of this feasibility study, the consultant team believes nonetheless that the number and range of people reached are sufficient to determine whether the requirements for proceeding to Phase 2 are met.

Feasibility and success factors

This assessment of the proposed ADLD project combines two sets of considerations. The first set addresses the feasibility conditions that must be met before the project can begin. The second set consists of conditions of success, or key elements needed to produce the best results for all parties involved. Further discussions among interested parties will be required to clearly distinguish conditions that are necessary to move forward (e.g., the minimum funding threshold) from the ideal setting to advance project goals.

The report's findings for all conditions are organized around the general questions raised with PHAC's external partners, as discussed below: the initiative's overall goal or focus, the rules of engagement that would make it possible, and the types of methods required to achieve meaningful results.

Project goal and focus

The stated goal of the ADLD project raises three issues: focus, relevance, and real-world impact. The goal of the project is to collect data on Canadians with ADLD, which most interviewees agree is a step in the right direction, albeit not a decisive one. The proposed action recognizes the limitations of the current case definition. It acknowledges the lack of surveillance data in this regard and seeks to assess the scope and complexity of the public health problem at hand. The general question is highly relevant and is less contentious than revising the national LD case definition, coming up with better testing and diagnostic tools, or developing more effective approaches to public education and medical treatment. The project thus begins with a simple fact: many cases of LD are diagnosed using alternative methods that are not recognized by Canada’s public health care system. The situation can be thoroughly investigated without debating the scientific validity of each alternative method. The data that may be collected and analyzed can include all the methods currently in use.

PATIENTS' SUPPORT AND CONCERNS

People with lived experience who were willing to be interviewed were generally supportive of the project. The same can be said for the Canadian Lyme Disease Foundation (CanLyme) and most of the patient-aligned physicians and researchers who were interviewed. However, some patient group representatives from highly affected provinces have found prior interaction with PHAC on the Lyme file to be unproductive and thus declined the invitation to be interviewed or discuss future engagement with PHAC. Other patient groups and advocates are willing to support the proposed action, but they are reserved on the issue.

People with lived experience who were willing to be interviewed were generally supportive of the project. The same can be said for the Canadian Lyme Disease Foundation (CanLyme) and most of the patient-aligned physicians and researchers who engaged in the consultation. However, some patient group representatives from highly affected provinces have found prior interaction with PHAC on the Lyme file to be unproductive and thus declined the invitation to be interviewed or discuss future engagement with PHAC. Other patient groups and advocates are willing to support the proposed action, but they are reserved on the issue. Many are concerned about how the data will be used and the project's impact on their priority concerns and hopes for improved testing and diagnostic tools, as well as long-overdue improvements in patient care. Conducting yet another study that further delays public health measures has little appeal for people suffering from severe infections. According to some, more surveillance does not always result in better LD responses by PHAC and medical authorities.

Patient groups' concerns point to a critical factor in achieving success: real world data must help raise public, scientific, and political awareness of the risks and suffering associated with LD, resulting in concrete action in a foreseeable future. The project provides a clear focus for collaborative action, but patient

REAL-WORLD DATA FOR ACTION

Real-world data must help raise public, scientific, and political awareness of the risks and suffering associated with LD, resulting in concrete action in a foreseeable future.

groups and advocates want to understand how their input will be used. This real impact factor should be addressed in discussions and directions taken during the design phase (Phase 2). Success and patient-group engagement will depend on efforts made to ensure that ADLD research findings are not sidelined or discredited before or as soon as they are made public.

Engagement processes

PHAC is considering using Participatory Action Research (PAR) to conduct a meaningful study of Canadians with ADLD. This entails a multi-stakeholder approach in which all parties believe the initiative is relevant to them and collaborate as full partners in the project. They are actively involved in designing, supervising, and conducting the necessary work, and their contributions are appropriately acknowledged. The approach differs significantly from traditional public consultation processes in which the public is asked to share their knowledge, ideas, and personal experiences on issues that affect them but is excluded from the subsequent decision making process. PAR develops formal engagement processes that cut across the all-too common divide between those who speak up in the hope of being heard and those who listen knowing that they have the power to make any final decision.

PARTICIPATORY ACTION RESEARCH

PAR develops formal engagement processes that address the all-too-common divide between those who speak up in the hope of being heard and those who listen knowing that they have the power to decide.

About 95% of those interviewed welcome the proposed shift and hope that the initiative will serve to identify common grounds for action rather than maintaining rigid views that block progress. If well conducted, the project could contribute to addressing the parties' long standing lack of trust and dialogue, a situation that predates the Framework and, by all accounts, has worsened in recent years. To proceed to Phase 2, all parties committed to the project must be willing to make room for genuine participation and take the expectations of LD communities across Canada more seriously. Given this requirement, the majority of interviewees expressed a

number of concerns and offered suggestions on engagement rules that could improve the project's chances of success. Stakeholder representation, shared leadership models, safeguards for engagement, flexible and time-efficient methods of participation, adequate funding, and information sharing were all discussed and framed along the lines described below.

Stakeholder representation and shared leadership. The collection of useful ADLD data requires the commitment, support, and buy-in of key stakeholders such as patient groups, Lyme-literate healthcare professionals, researchers, and public health authorities from different levels and branches of government. Researchers who work with patient groups play an important role and should be involved in the development of data collection methods and protocols. This inclusive approach to multistakeholder engagement will play a key role in efforts to translate new knowledge into health care and public actions that better meet the needs of patients.

All interested parties should investigate different models of shared leadership that call for transparent and effective governance and draw on novel forms of public engagement and citizen science. The initiative should include "patient partners" or "people who have personal experience with a health issue and informal carers such as family and friends" (CIHR). However, the engagement process must avoid appointing a token number of ADLD people simply to meet the minimum participation criterion. Formal mechanisms are required to fully integrate groups actively representing ADLD patients.

Safeguards for engagement. Explicit statements and actions are required to make it professionally safe for patient advocates and allied researchers and physicians to participate in this initiative. For example, support and representation from the Office of the Chief Science Advisor could help clarify the different roles and expectations of federal (PHAC) employees and independent researchers in the research process, as well as shielding patient-aligned participants from reputational harm.

Flexible and time-efficient participation. People's involvement in this project should be tailored to their individual circumstances. Some participants may be willing to devote less time to it than others due to health issues, professional obligations, other priorities, or any other reason. It follows that each stakeholder's expectations and the role they can expect to play in project activities must be clearly defined. Another success factor is the efficient use of the time spent on

this project by patients and their advocates. Instead of lengthy sessions of sharing information and personal stories about dealing with LD, the emphasis should be on useful discussions and tasks that must be completed in order to meet project goals.

ADEQUATE FUNDING

In order to succeed, this project cannot place the burden of responsibility on the shoulders of dedicated volunteers, whether they are patient partners, their representatives, or any health care professional working outside of their regular duties. PHAC must step up to the challenge it has set for itself and find solutions to the problems of surveillance underfunding.

Adequate funding. Even though the COVID-19 pandemic has significantly weakened patient groups, they continue to play an important role in spreading knowledge about LD and educating the public and health care professionals about the disease. Much of their work is done by volunteers from all over the country, many of whom live with LD daily. This initiative, which aims to learn more about Canadians with ADLD, could have a significant impact on the public health actions and the scientific research required to address what has become a leading infectious disease. However, in order to succeed, this project cannot place the burden of responsibility on the shoulders of dedicated volunteers, whether they are patient partners, their representatives, or any health care professional working outside of their regular duties. PHAC must step up to the challenge it has set for itself and find solutions to surveillance underfunding. This should include adequate and equitable coverage of the costs of Phase 2 engagement incurred by patients, their representatives, and patient-aligned researchers and physicians.

Information sharing. Many interviewees expect that, in keeping with PAR principles, this feasibility study and the findings of future studies be made available to all participants. Proper safeguards will be required to ensure that the collection of personal and medical data complies with the rules of informed consent, confidentiality, and security from the start. The public and the medical community should also be kept up to date on future information gathering activities, as well as their precise nature and implications for LD prevention and control.

BUILDING TRUST

Many parties interviewed are willing to proceed with the joint development of a pan-Canadian project that will close the evidence gap, advance understanding, and raise awareness of the public health issues associated with ADLD. However, some key patient groups and advocates are still unwilling to commit to Phase 2 activities (aimed at planning and building consensus around data collection methods and rules of engagement). These groups are not, for all that, opposed to others (notably CanLyme) moving forward and attempting to find common ground for collaborative action.

Phase 2 of the proposed ADLD project involves working out the rules of engagement and key details of the data collection process. At this point, it is difficult to predict whether the success factors discussed in this study will be in place, allowing for the effective implementation of the original Framework recommendation. Many parties interviewed are willing to proceed with the joint development of a pan-Canadian project that will close the evidence gap, advance understanding, and raise awareness of the public health issues associated with ADLD. However, some key patient groups and advocates are still unwilling to commit to Phase 2 activities (aimed at planning and building consensus around data collection methods and rules of engagement). These groups are not, for all that, opposed to others (notably CanLyme) moving forward and attempting to find common ground for collaborative action. If successful, the initiative may be a unique opportunity for PHAC, researchers, health care professionals and the LD community to build trust and create a space for the kind of dialogue that connects well-structured information gathering with meaningful action. Many feel that passing up this opportunity will further stall efforts to improve Canada's ability to prevent, detect, and treat LD across the country.

The project will require nonetheless the assistance of an experienced third party to facilitate discussions about project roles and design, as well as the active support of high-level outreach personnel and officials within PHAC. Resuming the Annual Multi-Disciplinary Stakeholder Meetings, which were held prior to the COVID-19 pandemic, should also be seriously considered, in due time, with a consensus-building approach rather than traditional public consultations methods. These meetings are not required for project completion, but they do provide a forum for discussing more decisive and higher-impact issues such as better LD testing, diagnostic tools, and

treatment approaches. Improvements in these areas are desperately needed, even if only on a temporary basis as research and science progress.

Data collection methods

In this study, feasibility and the likelihood of success are also assessed based on the data collection methods required to achieve project goals. Collecting sound data and providing an accurate picture of Canadians with ADLD raises methodological issues and challenges that will be addressed in Phase 2. However, none of the interviewees saw any major technical challenges with studies of ADLD cases. Instead, most discussions focused on success factors such as identifying good data sources and ensuring that the collected data have real-life applications. Models that some would like to emulate emphasize the benefits of a "one-health" approach to human and animal health, as well as improved synergies between information gathering and other LD activities, such as education and best health care practices.

Data sources. In Canada, centralized databases for passive surveillance, such as Quebec's *Registre central des maladies à déclaration obligatoire* (MADO), are the primary source of information on patient demographics, clinical data signs and symptoms, diagnostic test results, and medical case history. For reasons already stated, they do not provide an accurate picture of the incidence and prevalence of LD in Canada, let alone its demography and impact on people's lives. A gamut of accessory methods can be used to gather more information on ADLD and help fill the gap. They range from administrative claims and public health laboratory findings to active, population-based studies of LD using appropriate survey sampling techniques. Medical laboratories track diagnostic tests and collect limited demographic data, primarily for operational purposes. Administrative claims provide testing, diagnostic, and treatment data from patients who interact with private or public health service providers. They, too, have limits. Many physicians and billing agencies are unaware of the diagnostic codes for LD, which are imprecise and may even vary from system to system.

Other possible data gathering measures mentioned by some interviewees include adding Lyme related questions to the Canadian Community Health Survey or collecting biobank samples from LD and ADLD patients. Health agencies, physicians, patient groups and researchers also conduct

their own surveys and reach out to concerned communities and the public. The most recent examples include the *Sondage bilingue de l'AQML sur la maladie de Lyme et les co-infections du Québec* (2018) and the Lymescape survey which arose from a partnership between LymeNB and the Lloyd Lab at Mount Allison University (2019). While survey fatigue and low response rates may hinder research efforts of this nature, the patients interviewed as part of this feasibility study appeared to be highly motivated by the prospect of obtaining a comprehensive picture of the affected population across Canada. This is especially true for those suffering from late or chronic LD. Participants agree that whatever methods are used, steps must be taken to share and ensure consistency of data across various regions and jurisdictions, using collection standards for compatibility and clear privacy safeguards.

ADLD PATIENTS WILLING TO CONTRIBUTE

While survey fatigue and low response rates may hinder research efforts of this nature, the patients interviewed as part of this feasibility study appeared to be motivated by the prospect of obtaining a comprehensive picture of the affected population across Canada. This is especially true for those suffering from late or chronic LD.

Real-life applications. The Framework evaluation report (January 2022) raises several concerns about the development of surveillance tools and methods, particularly their limited role in guiding personal behavior or work practices in real time. The citizen science project eTick, a public platform that helps monitor ticks in Canada, should be mentioned here. It is a PHAC funded example of how participatory research can aid in the development of risk maps and studies of populational health issues. However, current maps lack the precision required to guide behavior or inform diagnosis and treatment on an individual level. They do not provide the public and health care professionals with widely accessible and regularly updated information about existing LD risks and their evolution over time. The proposed study should consider alternatives in this respect, with a focus on cases of ADLD, and ensure that the information gathered meets the needs of all stakeholders, not just researchers and public health officials. Special attention should be given to the visual appeal and interactive features of online products intended for regional and community distribution. Warnings should also be issued against data misinterpretation, such as when maps

are used to deny testing or insurance coverage in low-risk areas. The limitations of Lyme disease mapping include the fact that people travel and that their place of residence may differ from where the ADLD patient is believed to have been infected.[10]

RISK MAPS USED APPROPRIATELY

Risk maps should provide the public and health care professionals with widely accessible and regularly updated information about existing LD risks and their evolution over time, along with warnings against data misinterpretation, such as when maps are used to deny testing or insurance coverage in low-risk areas.

Partners in the proposed initiative should draw on international expertise and new technologies in the field of participatory LD surveillance, including measures to ensure adequate privacy safeguards. According to some interviewees, one model to consider is a made-in-Canada version of **MyLymeData**, an online big data project launched by LymeDisease.org in 2015 that now has a patient registry with over 12,000 members. By pooling a large number of diagnoses and treatment experiences, this patient-powered research project helps advance patient-centered research at the same time as it allows patients to learn from each other's experience, access a list of physicians treating LD, and keep track of new developments in the field.

Breaking down silos

Despite their own health issues and great dissatisfaction with Canada's health-care system, many ADLD patients are willing to contribute to research, participate in clinical trials, and provide blood samples. They offer support to others in need and devote time to public education and awareness campaigns. They do all of this out of altruism and with courage. The proposed ADLD initiative should nonetheless explore ways to meet some of their immediate needs. This can be done by offering information that may help survey respondents minimize risk, stay up to date on the latest research and treatment options, make informed choices, navigate the health care system, and explore new treatment avenues.

[10] See https://www.cdc.gov/lyme/datasurveillance/lyme-disease-maps.html.

The approach used should assist patient groups in educating Canadian adults and schoolchildren about LD infection prevention. Rather than operating in a silo mode, the project would benefit from a multisectoral and multidisciplinary approach to public health issues, ensuring synergies with public education, medical practice, and health care. Once formed, the project team should also consider involving veterinarians in developing a multilayered methodology that recognizes the direct connections between tick-infected people and animals (especially dogs). This would be consistent with the One Health principles currently promoted by the World Health Organization and the Food and Agriculture Organization of the United Nations.

DATA MEETING NEEDS

The proposed ADLD initiative should explore ways to meet some of the patients' immediate needs. This can be done by offering information that may help survey respondents minimize risk, stay up to date on the latest research and treatment options, make informed choices, navigate the health care system, and explore new treatment avenues.

Summary and next steps

This report assesses the feasibility of PHAC's idea of using a participatory action research approach to improve knowledge and inform future public health actions for Canadians with ADLD — people diagnosed with Lyme disease (LD) through methods that differ from the national criteria for the case definition. Given this focus, the proposed initiative makes no attempt to address concerns about LD treatment protocols. Nor does it try to resolve current debates about the national case definition or the transmissibility of LD in Canada.

The consultant team conducted 39 semi-structured interviews with Lyme-literate individuals and people with lived experience, covering a sufficiently broad range of stakeholders involved in LD-related activities. The focus of the proposed initiative, the rules of collaborative engagement, and the data gathering methods required to achieve meaningful results were all discussed. These interviews and review of publicly available documents led to the following observations and conclusions:

- The project provides a clear focus for collaborative action based on the principles of Participatory Action Research. It is recommended in the Federal Framework on Lyme Disease and is generally supported by interviewees with lived experience and patient-aligned researchers and physicians who engaged in the consultation.
- Some patient groups and advocates are willing to pursue the idea. They consider that collecting data on Canadians with ADLD is a step in the right direction. However, it is far from decisive when compared to revising the LD case definition, developing better LD testing and diagnostic tools, and making long-overdue improvements in patient care.
- Other patient group representatives remain distrustful of the federal government's overall approach to Lyme-related issues. They turned down the interview invitation, preferring to share their thoughts with the patient advocates who agreed to participate in the discussion, notably CanLyme.
- Given persisting tensions, the project will require the assistance of an experienced third party to facilitate discussions about project roles and design, as well as the active support of high level outreach personnel and advisory officials within PHAC.

- Success and patient-group engagement will depend on obtaining adequate federal government funding for all phases of the project, including the engagement process.
- Various models of shared leadership and partnering should be investigated to support transparent and effective project governance, clear role definitions, and novel forms of public engagement and citizen science.
- The data collection process should provide adequate privacy safeguards from the start. It should be designed in such ways as to raise public, scientific, and political awareness of the risks and suffering associated with LD and effectively guide people's behavior.
- Synergies between ADLD data gathering and other pillars of the Lyme Framework should be explored. A one-health approach to investigating the direct links between tick-infected people and animals is also worth considering.

In terms of next steps, the consultant team suggests that Phase 2 begin with the creation of a multi-stakeholder working group that provides a good representation of all interested parties. It should ideally include at least three patient group representatives, one patient partner, two members of the PHAC's Domestic Vector-borne Diseases Technical Team, one higher official within PHAC, two researchers, two public health officials, and one Lyme literate doctor. Counting on an experienced third party to convene the working group and facilitate consensus building would also be advisable. The working group would be tasked with determining the membership of an **advisory committee** that can provide strategic direction and broader support for the initiative. Once formed, the advisory committee could revisit its own composition and mandate, and then address issues of project leadership, overall project parameters and their financial implications, as well as Phase 2 critical path planning and budgeting. The composition and mandate of the committee responsible for designing and carrying out project activities should also be explored.

PHASE 2 WORKING GROUP

In terms of next steps, the consultant team suggests that Phase 2 begin with the creation of a multi -stakeholder working group that provides a good representation of all interested parties.

Conclusion

This project, entitled *A Participatory Action Research study to improve knowledge and inform future public health actions for Canadian residents diagnosed with Lyme disease (LD) through alternative means but who do not meet the LD national surveillance case definition*, can be successful provided certain conditions are met (as described in the preceding section). The study concludes nonetheless that the project is feasible in principle and could provide significant benefits if implemented using a participatory approach, as proposed.

Participatory Action Research on Canadians with Alternatively Diagnosed Lyme Disease: Feasibility Study

Feasibility Report

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ADLD PROJECT

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