

Assessment

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Developing recommendations for the diagnosis and treatment of Lyme disease: the role of the patient's perspective in a controversial environment

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Abstract

Context. The Ministry of Health in Québec requested the National Institute of Excellence in Health and Social Services to produce clinical and implementation recommendations for the prophylaxis, diagnosis, and treatment of Lyme disease.

Objectives. (i) Describe the process of trialing different modalities of patient engagement as a means to integrate a diversity of patient perspectives and (ii) Describe the learning process of INESSS regarding the integration of the patient perspective.

Methodology. All documents were analyzed, and a survey with all advisory committee members and semi-structured interviews with stakeholders were conducted. Each interview was transcribed verbatim and imported into QDA miner software for the purposes of analysis. Data analysis was carried out concurrently with data collection to allow for an iterative approach between data collection and analysis.

Results. Five methods to integrate the perspectives of patients were used: (i) interviews with patients, (ii) inclusion of patient partners within the advisory committee, (iii) literature review, (iv) focus groups with one patient association, and (v) feedback from patient associations on recommendations intended for decision makers and other targeted stakeholders. The patient partners influenced decisions by sharing their experiential knowledge. The patient interviews and the literature review added an in-depth perspective on the disease and experience with the healthcare system. The patient association members shared their perspectives and helped disseminate the recommendation to sustain a practice change.

Conclusion. The combination of methods to collect and integrate patients' knowledge and patient associations' perspectives helped develop a comprehensive understanding of a controversial object of evaluation.

Background

Lyme disease is a growing public health problem in Quebec, where reported cases doubled between 2013 and 2017 and reached 500 in 2019 (1). In autumn 2017, the Quebec Ministry of Health and Social Services mandated the Institut National d'Excellence en Santé et Service Sociaux (INESSS) to develop recommendations, for decision makers and other stakeholders, regarding the diagnosis, treatment, and follow-up of Lyme disease. In winter 2018, two petitions, with 9,000 signatures, were tabled in the National Assembly requesting that Lyme disease be recognized as an urgent public health problem, all stages of the disease be clinically diagnosed, doctors providing alternative care be legally protected, and that a committee, including a patient association, oversee the implementation of a Lyme disease action plan (2). Subsequently, the parliamentary committee highlighted the complexity and difficulty of the diagnosis, the lack of consensus regarding treatment (3) and the need for research and to improve patient care. Thus, INESSS sought to develop recommendations and knowledge-transfer products concerning post-exposure prophylaxis for the prevention of Lyme disease, as well as its recognition, diagnosis, and treatment. Aware of the different schools of thought regarding Lyme disease, the INESSS project team collaborated with stakeholders while preserving their independence and managing conflicts of interest and roles. Given the controversy, INESSS decided that it was important to include patients among these stakeholders and that this mandate was ideal to test different modalities of patient engagement. Thus, they turned to the Centre of Excellence on Patient and Public Partnership (CEPPP) (4) and hired a patient coordinator with partnership expertise and two experts in participation with patients, users, caregivers, and citizens to help enrich the health technology assessment

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(HTA) process with patients' voices, perspectives, and experiences. Five complementary methods were used: (i) two patient partners (PPs) diagnosed with Lyme disease were included on the Lyme disease project advisory committee whose role was to participate in all phases of the HTA; (ii) a systematic literature review specific to Lyme disease patient perspectives; (iii) interviews with Lyme disease patients; (iv) a focus group with representatives of a patient association to account for the advocacy perspective; and (v) patient association feedback regarding the recommendations developed.

The objectives of this study were to describe (i) the process of trialing different modalities of patient engagement to integrate a diversity of patient perspectives and (ii) the learning process of the Lyme project team regarding the integration of patient perspectives. This study received ethical approval from the University of Montréal Hospital Research Center (CRCHUM) Research Ethics Committee (CER-19.350).

Methods

Data Collection

A mixed methods case study (5) was conducted. Data were collected between June 2019 and May 2020. First, all documents produced during the project (e.g., meeting minutes, documents prepared for meetings, the focus group summary, the analyses of the patient interviews, and the recommendations) were collected to help understand how the patient voice was integrated to co-construct the recommendations. Second, semistructured interviews, between 30 and 90 min in duration, were conducted by three coauthors (MPP, AL, and LL) trained in evaluation research. Two interviews with the PPs and six with the INESSS Lyme project team (ILP) were conducted at the end of the project. An additional fourteen interviews with the two PPs ($n = 2$) and the Lyme disease experts (ELD), including healthcare professionals (HCPs) ($n = 4$) and a parasitologist ($n = 1$) on the advisory committee, as well as with ILP ($n = 5$), a member of the INESSS methodology team ($n = 1$) (IMT), and one representative of a Lyme Patient Association (LPA) ($n = 1$) were conducted. The interviews focused on how patient perspectives were integrated in the HTA and the perceived added value of these perspectives and were recorded (Supplementary Appendix 1 and 2). Third, a survey regarding the perceived role of the PPs was sent to all advisory committee members and the LPA representative ($n = 21$) to assess their stage of readiness to work with PPs and their perceived contributions of the PPs during the project (Supplementary Appendix 3).

Data Analysis

Documents were analyzed to reconstruct the chronology of the HTA process and to identify and understand how the various patient perspectives data were integrated in the recommendations. Interviews were transcribed verbatim by MPP, reviewed by LL, the patient coordinator, and analyzed using QDA miner software (6). Data analysis was conducted concurrently with data collection to allow for an iterative approach. Five analytic stages were used (7). First, two coauthors (MPP and AL) read and reread all documents and interviews to familiarize themselves with the data. Second, they identified, independently of each other, the thematic framework that fit the data. Third, they met to discuss the framework and their respective themes and determine which thematic

would guide their data analysis. Fourth, using this thematic framework, three coauthors (MPP, AL, and ODP) coded two interviews, independently of one another, and then discussed their coding and compared data across patterns they identified. Subsequently, all interviews were coded by one coauthor (MPP) and the coding was checked by another (AL or ODP). For the survey data, descriptive statistics were calculated. Finally, the qualitative results of the framework analysis and the quantitative results from the survey were integrated; all coauthors helped map and interpret the overall results.

Results

Methodological Choices and Associated Challenges

Given that the Lyme project team members had no previous patient engagement experience, they initially found it challenging to incorporate the patient perspective in their evaluation. The Lyme project team, the CEPPP, and the INESSS methodology experts had several discussions regarding the added value of having PPs on the advisory committee. With a prudent mindset and the reassurance that they would be supported by the CEPPP, they recruited two PPs with a diagnosis of Lyme disease on the advisory committee, thereby ensuring that the recommendations would include patients' perspectives (and not only those of health professionals): "I expect patients to give us their opinion when we ask for it to help us improve our work" (ILP2). Additionally, a monitoring committee of representatives of professional orders, medical federations, the Québec National Institute of Public Health, and the Ministry of Health and Social Services was kept abreast of the project's progress.

The selection criteria for the PPs of the advisory committee were determined jointly by the INESSS project team and the CEPPP. Through discussions, it became clear that it was important to recruit patients with a confirmed Lyme disease diagnosis to avoid controversy regarding their participation. Thus, recruited PPs had to have consulted physician for systemic, neurological, musculoskeletal or cardiac symptoms, or atypical erythema migrans, have been diagnosed with Lyme disease based on their case history and/or positive serology according to a recognized diagnostic test, and be fully or partially recovered (i.e., experiencing persistent symptoms). Moreover, it was decided that the PPs' physicians could not be on the advisory committee to help ensure all committee members could express themselves freely. Together, the CEPPP and the clinicians already recruited to the advisory committee identified seven potential PPs. Patient associations were not used to recruit PPs, given that it was important to all actors that the PPs not be involved in or associated with any of the controversy surrounding Lyme disease. The seven candidates were interviewed by the CEPPP expert and the INESSS patient coordinator. Based on their diversity of experiences, their collaboration skills, and their ability to be objective despite their own diagnosis, two PPs were recruited: "The CEPPP, at the beginning, helped a lot in choosing the patients. It reassured us that they were there" (ILP1). The CEPPP provided a one-day training session on patient partnership concepts and the PPs' potential contribution to the mandate prepare the PPs and the Lyme project team for their collaboration and to ensure that they had a shared understanding of the PPs' role:

"Having received training in this field made it easier for me to get involved" (PP1).

“The training session together was crucial. Everyone spoke up. The atmosphere was very positive. It allowed us to break the ice. It showed the people at INESSS that the patients had a head on their shoulders and that they had to be open-minded” (PP2).

“During the training session, we got to know the patients. It allowed us to break the ice, to be a little less afraid of embarking on this adventure, despite having no experience. We felt supported and well supervised with rigorous methodologies by the CEPPP. We could trust them” (ILP1).

This training session was also a space where everyone could express their expectations:

“As a patient partner, I hope to be able to give my opinion on the strategies implemented at the main stages of the project as well as on some of the results. I would like my input to be sought in contexts where team members feel it would be useful. Thus, I prefer to intervene in a targeted and relevant manner rather than in the project as a whole. It is also easier and more realistic for me to contribute to the project in this way” (PP1).

“I expect patients to share their experiential knowledge, to provide information that is complementary to the scientific evidence, to identify contextual issues, to nuance recommendations if necessary, and to make the information more accessible to other patients or citizens” (ILP1).

“As for me, a patient partner, I am convinced that our presence will result in publications that will have more relevant content, not only in terms of the means used to gather information, but also in terms of the language, the method and the subjects used to disseminate the documentation” (PP2).

To collect additional patient perspective data, interviews with patients of two Lyme disease profiles were conducted. The first profile ($n = 2$) consisted of patients who had consulted for a typical erythema migrans, and the second profile ($n = 6$) was defined as patients who had consulted for systemic, neurological, musculoskeletal or cardiac symptoms, or for atypical erythema migrans. For the project team member: “It seemed essential to us to gather the perspectives of 8 to 10 people other than the patient partners in order to produce the recommendations” (ILP2). However, the PPs felt that they could have participated in the interviews: “To conduct the interviews, it would have been interesting to have a patient as well to form a tandem with [INESSS team member]” (PP2). In addition, the Lyme project team members did not know how to integrate these with the results of the review of the scientific literature: “We didn’t know how, but we knew we wanted to interview patients and the patient association. We wanted the patient perspective, but to what extent?” (ILP1).

The Lyme project team also held a focus group with five people from a Lyme disease patient association where the PPs were present. Although the team members were eager to include the association’s perspective, they were concerned the association representatives might take an activism stance given the political context previously described. The PPs helped highlight the complementary nature of the association’s contributions:

“We took part in the meeting between INESSS and the patient association. The members of the project team had some concerns. We were able to reassure the association members that the INESSS team would consider all their recommendations with a positive attitude. The project team really appreciated our participation” (PP2).

Finally, the project team conducted a literature review on the perspectives of Lyme disease patients. They worked with the PPs to define the eligibility criteria of the studies to include in the review, the aspects to evaluate, and to refine the quality-of-life

conceptual framework used. Once the review was written, the team sought the PPs’ comment and suggestions (1;8;9).

Learning in Action

Several things were clarified during the first advisory committee with the PPs:

“The two patients presented their care pathways to the committee members and there was immediate acceptance. This was when we established the definitions. The discussion was more scientific. There were suggestions such as talking about the disease ‘carrier’ tick, not the infected tick” (ILP1).

The Lyme project team presented the preliminary results of the literature reviews on various aspects of Lyme disease, but chose not to simplify these documents for the PPs. This was a challenge for both the project team and the PPs. On the one hand, aware of the project’s tight timeframe, the project team did not want to devote a lot of time to drafting preliminary results. On the other hand, the complexity of the content and the technical jargon limited the PPs’ potential to contribute. After discussions with the CEPPP expert and the INESSS patient coordinator, the PPs and project team agreed to take the time to go over the documents together, as needed, prior to the advisory committee meetings. This helped increase PPs’ participation and avoid ensure the PPs did not feel excluded:

“We wrote one document, it would have been too long to write one specifically for the PPs, so we told them which pages to read. This is a must because otherwise it’s difficult for patients to prioritize” (ILP5).

“The scientific advisors always answered our questions. This made it possible to fully understand everything that was presented to us. They were always willing to elaborate on the subject. If I was uncomfortable with certain topics, I could have a conversation with the scientific advisors who were willing to discuss them with me. When I sent in suggestions, they would respond very quickly” (PP1).

During the meetings, patients sometimes had to assert themselves to get their ideas across:

“At one of the meetings, we talked about giving the banding pattern (raw result of serological analysis) to patients who request them. Some ELDs were against it and the patient partners were in favor. It was not unanimous. Patients were able to explain the need to be transparent and that knowing the banding pattern could help patients to accept their condition” (ILP5).

“One of the patient partners wanted to talk about the post-treatment symptoms of the disease. The patient was challenged by the ELDs who questioned what she was saying. Sometimes clinicians find it interesting to have patient partners on a committee, but I’m not sure they give equal weight to what they say” (ILP5).

The project team found it relevant that PPs were present at all meetings to hear all discussions and intervene when relevant:

“I was glad they were present at all the meetings. For example, during the meeting on neuroborreliosis, patients were able to discuss their own clinical manifestations. For the diagnostic value of the laboratory analysis, their interventions were less relevant, but the meetings allowed for providing examples” (ILP1).

Overall, the Lyme project team found that the interviews and focus groups data complemented the literature reviews: “The interviews with patients of two different Lyme disease profiles and the focus group with the patient association made it possible

to gather complementary information that was interesting and essential for this complex project” (ILP4). They also learned from and how to work with PPs. It was a continuing learning opportunity for the project team: “Definitely, this is the project I’ve learned the most from in my career” (ILP1).

Based on the document analysis, over the course of the project, the Lyme project team and the INESSS methodology team clarified the ethical basis for a responsible management of conflicts of interest and roles. Specifically, they determined that the advisory committee required members (professionals as well as patients) to act as partners in the project and not advocates for their respective causes. Moreover, there were discussions at the INESSS to integrate patient associations in monitoring committees on the same footing as professional associations. However, it was decided to avoid this with controversial evaluation topics, given the many factors that could be at stake.

Specific Contributions of Patient Engagement According to Different Stakeholders

Although the PPs did not know whether their participation influenced the orientation of the project, they were able to get their points of view across:

“When they do the tests, they can do a western blot—also called a strip sample—and depending on the number of bands you can tell if you have Lyme disease. The question that came up during the committee was whether the banding profile should be made available to clinicians and patients. We (both patients) felt that it was essential to make them available to patients in a transparent way, so that we don’t feel that something is being hidden from us. For us, this sharing contributes to building trust. The health professionals on the committee weren’t sure about giving access to this profile to clinicians. But when they were presented with the idea that it might be useful for research, they agreed” (PP1).

“I think we were able to give the INESSS project team and the committee members a form of authorization to take a more humanistic look at this pathology. Because these people often hide behind scientific rigor” (PP2).

The health professionals mentioned that the PPs’ participation was particularly important, given the need to consider the patient care pathway, which is not always optimal, in the recommendations:

“Having seen patients in great distress, patients who were shown the door, it’s important they share their experience of diagnosis” (ELD1).

“I think that the most eye-opening part for us is to see to what extent these patients can be suffering due to the symptoms, and the number of medical visits they go to with the hope of finding solutions” (ELD5).

“I believe that the participation of patients is interesting to learn about their personal experience (for example, experience of being cared for by the healthcare system) and their overall perspective of a disease they have suffered from. There is a lot of information circulating, particularly about Lyme disease, and it is interesting to see how patients perceive the various health care/care/etc. providers in this context” (ELD3).

Both the PPs and the health professionals found that the most significant ways the PPs contributed was by sharing their experiences and participating in the development of the knowledge-transfer tools:

“Their contribution was, above all, during the tools development process in order to avoid falling into the trap of professionals who always want to say too much. They thus made it possible to produce tools that are popularized, accessible and understandable” (ELD4).

“Thanks to their testimonials, they contributed their experience of being cared for by the healthcare system. I believe that this kind of information is difficult to find in the literature, and so, in this context, patient participation benefits the committee” (ELD1).

“They brought the human dimension, their journey, the delays incurred, the moral suffering. They told us how difficult it was to get a diagnosis and how their diagnosis was given to them” (ELD3).

“By sharing my experience, I was able to contribute a more human perspective of the disease in an environment that focusses on research evidence. When developing the tools, I was able to contribute to each meeting and make the tools more adapted to patients’ needs” (PP2).

“I feel that I provided a more human dimension to the recommendations, there was insufficient evidence to formalize the recommendations. As patients, we were able to raise issues that no one else could have afforded to raise” (PP1).

The survey results showed that 39% of all advisory committee members thought the PPs provided a unique perspective, 44% had no opinion regarding the PPs’ contribution, and 17% said they had not contributed. Moreover, 60% of respondents said PPs had influenced the course of the mandate, and 57% of the HCPs intended to change their care practices as a result of their experience.

Regarding the characteristics of the PPs, all of the Lyme disease experts said that the PPs had been well selected. However, initially some had concerns regarding how well they would collaborate:

“At first, when I heard that patients were going to be on the committee, I was very reluctant. In the past, I participated in consultations organized by the Public Health Agency of Canada regarding the federal Lyme disease framework. Several types of patients were present at these consultations. When patients from all categories are included, we find ourselves in a situation where we no longer know how to differentiate between true cases of Lyme disease versus false ones. The information about the experience of patients who really have Lyme disease, according to the scientific community, is diluted and it becomes difficult to obtain the information sought” (ELD1).

“I expected it to be rock and roll, but they were collaborative” (ELD3).

“The people on the committee were not activists, neither the patients nor the professionals. They were very rigorous and objective and were able to look at what the science was saying whether they [patients] liked it or not” (ELD4).

Overall, the healthcare professionals and the INESSS Lyme project team found that the PPs’ participation on the advisory committee helped ensure that the evaluation process was democratic, legitimate, and transparent, and that the knowledge-transfer tools developed were ones healthcare professionals could use with their patients:

“It helps to have a democratic and transparent scientific process and to make sure the recommendations are accepted by everyone” (ILP3).

“At the populational level, I think that this could increase the credibility and trust of the population towards the INESSS’s report and its recommendations” (ELD5).

“It was a good idea to include patients in a controversial project like Lyme. It helped to make the approach even more credible. It got rid of a possible suspicion of a conspiracy theory in society at large that can arise when small, closed and inaccessible groups make decisions. The patients were the guardians of a democratic process that ensured transparency to limit potential bias on the part of the committee” (ELD2).

One ELD expressed being reluctant to speak at times during meetings given the presence of the PPs:

“I have to admit that the presence of patients on the committee may have hindered some of the discussion points in the meetings that I might have brought up regarding laboratory diagnosis had they [PPs] not been

present. I sometimes contacted the INESSS representatives outside the meetings to improve my explanations. I want to avoid appearing rigid regarding the patients' points of view and condescending to some of their opinions that go against current scientific evidence" (ELD1).

Including the patient association in the process led to their decision to help disseminate the recommendations and knowledge-transfer tools:

"Submitting the recommendations and the tools to the patient association helped us in disseminating the tools and the report. It was a good idea also to do a focus group, it helped us create a relationship with those associations" (IMT1).

"We tried to contribute to the dissemination of the report and recommendations by making them known to our members who in turn shared them with their health care professionals. Without us, I'm not sure the [health] professionals would have learned about these tools" (LPA).

Discussion

Below, we discuss how the different modalities of patient engagement helped INESSS include diverse and complementary patient perspectives. Then, we describe what the Lyme project and INESSS methodology teams learned regarding the integration of patient perspectives in the evaluation process such that fair and reasonable recommendations were developed.

In the politically controversial context regarding the recognition diagnosis and treatment of Lyme disease, INESSS was mandated to produce clinical and implementation recommendations and knowledge-transfer tools. Given a desire to include patient perspectives in their evaluations, INESSS decided that this was an opportunity to trial various modalities of patient engagement. This study shows that despite the context and their initial resistance, the project team was able to adapt, learn, and successfully use various means to collect and integrate patient perspectives in all deliverables (1;8–11). INESSS' willingness to incorporate patients' voices and experience in its work, to collaborate with the CEPPP, and to hire a patient coordinator (12;13) allowed for this unique patient engagement project to be possible.

This window of opportunity allowed the project team to combine multiple patient engagement modalities, from including PPs on the advisory committee and patient association feedback regarding the recommendations developed (14). Moreover, the interviews with patients of two Lyme disease profiles and with a Lyme patient association and the feedback from the PPs on the patient experience literature review increased the place that patient voices, perspectives, and experiences held in this evaluation (15–18). In fact, this study illustrates that it is possible for an HTA organization to engage patients in a combination of ways and enrich their evaluations (15).

The case study illustrates that including PPs on the advisory committee is a way to gather and integrate knowledge that is not available in the scientific literature (19–21). The PPs' presence served as a continual reminder to the healthcare professionals that, beyond the scientific data, there are complex experiences lived by patients that should be taken into account throughout the scientific recommendation development process. Participants considered the PPs' presence essential to guarantee transparent decision making and a democratic process, and this despite the fact that one ELD was concerned about causing worry for them or arguing over the science with them. Additionally, the PPs' contributions were recognized not only by the Lyme project team and advisory committee members,

but also by the PPs themselves and the patient association, which is often overlooked in the literature (17). The PPs also acted as a bridge between the patient association representatives and the Lyme project team by being able to describe each person's role and the complementary nature of their respective perspectives.

This study illustrates the different points of view about the value of integrating patients in the Lyme project that stakeholders held when the project began. The Lyme project team members wanted to integrate the patient point of view but had no experience in doing so. The healthcare professionals were concerned whether the PPs would be able to be objective regarding the research evidence and about disagreeing with them on the science. The patient association, for its part, wanted to be part of the advisory committee, but INESSS wanted to, first, clarify what would be at stake in terms of their conflicts of interests and roles. This study highlights that, although stakeholders have distinct agendas and convictions, it is possible to create a place for dialogue among them. Moreover, this study shows that dialogue is possible even when there is a lack of trust among the actors, given a highly politicized context such as that of Lyme disease in Quebec (2). A factor that can explain how space was created to ensure fruitful collaboration is the support of CEPPP and the patient coordinator because they helped the INESSS project team decide how to integrate patient perspectives and insisted on the necessity of including PPs on the advisory committee. The support provided by the patient coordinator and the CEPPP gave the Lyme project team confidence in their ability to develop patient engagement skills, while maintaining their usual work processes (e.g., not adapting documents for the PPs). Moreover, given that a rigorous selection process for the PPs was implemented, the health professionals were more ready to recognize the value added by their participation. Because the PPs each had a clear and accepted diagnosis of Lyme disease and were able to understand the research and share their experiences, the other advisory committee members were open to listen to the difficulties that Lyme disease patients face and to maintain a patient-centered focus. These results corroborate findings from a previous study conducted at INESSS that highlighted the importance of selecting PP using disease-related criteria, among others (18).

Additionally, for the patient association, they understood the scientific evaluation methodology and found that the advisory committee had done valuable work. By the end of the Lyme disease evaluation project, the project team and a majority of the advisory committee members felt they had developed new competencies and were proud of the results of their work.

In the light of the findings from this experience and a similar one (17), the following five strategies appear to be critical to the effective integration of patients and patients' perspectives in HTA. First, it is important to define each of the HTA patient engagement modalities and their complementarities. Second, patients on advisory committees must not take an activist stance, but rather, participate as partners. Third, the selection criteria for these PPs must be specific to the needs of each project. Fourth, project teams should be trained in patient engagement methods and provided with ongoing support, from internal and external resources, as needed. Fifth, patient association representatives could be included in strategic committees to bring their point of view. Although it may be difficult to combine all five strategies in every evaluation, it is important to consider their relevance to the complexity of the evaluation.

Strengths and Limitations

Although this case study followed the CORE-Q guideline, it has one main limitation, namely that many of the coauthors work at INESSS, hindering, perhaps, their objectivity (22;23). However, the authors who conducted the interviews and the analysis are not INESSS employees. Moreover, they did not involve the coauthors in the interview guide development, relying instead on their document analysis to articulate relevant questions. Furthermore, all stakeholders agreed to participate. Finally, the interview transcripts suggest the participants spoke freely.

Conclusion

This article illustrates how, despite the challenge of conducting this evaluation in a politically charged context, the multilayered patient engagement strategy put in place by the Lyme project team made it possible to integrate varied and rich sources of patient perspectives in the Lyme disease project.

The PPs on the advisory committee actively participated in the discussions, despite their scientific nature, and were able to influence decisions by sharing their experiential knowledge. It is worth noting that INESSS and the Lyme Disease Patient Association have come to trust each other, and the association had the opportunity to comment on the implementation recommendations and clinical decision tools developed by the project team. Moreover, the association has disseminated these products to patients to help induce and sustain healthcare practice change.

Following the Lyme evaluation project described herein, INESSS followed up with an evaluation specific to the highly debated chronic form of Lyme disease. The lessons learned from first evaluation are being used to integrate patient perspectives in this second project.

Supplementary material. The supplementary material for this article can be found at <https://doi.org/10.1017/S0266462320002123>.

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