What does meaningful engagement in Lyme disease research mean to you?

Survey report

Summary: The "What does meaningful engagement in Lyme disease research mean to you?" survey was created by Canadian Lyme Consortium; a partnership between researchers, medical professionals and the Lyme disease patient and advocate communities (www.ClymeC.ca). One of the goals of the CLC is to establish a national and patient-centered Lyme disease research network, including a national database and biobank. In order to ethically practice patient-centered Lyme disease Research, meaningful patient engagement is vital. The survey was created so as to ask people in the Lyme communities how meaningful patient engagement in Lyme disease research should be accomplished. The survey was available, in both official languages, from June 4- July 1, 2018 and attracted a strong response with 721 respondents from across Canada. This indicates strong interest in, and support of research within the Lyme patient community.

Demographics: The majority of survey respondents identified themselves as Lyme disease patients or family or friends for Lyme disease patients with some medical professionals also participating.

Requirements for ethical research on Lyme disease: There was strong endorsement of all components of ethical research; >60% of all respondents endorsed meeting with patient groups throughout a study, complete disclosure of research plans and processes, patient input into research before implementation, patient-identified research priorities and returning both summarized findings and personal study results. The latter two considerations were deemed part of meaningful engagement by greater than 75% of respondents, with continued patient contact also being supported by nearly 80% of respondents. 77 respondents provided their own selection for important components of meaningful engagement. The two most common components were to allow patients to play a more active role in decision making in Lyme research and for research to involve improved diagnostic tools for Lyme disease in Canada.

Other suggested components of meaningful research were treating patients with respect, inclusion of all patient groups and good communication with patients.

Biobank and patient registry: Participants were asked about two specific research projects, a biobank and a patient database/registry. 93% of respondents supported a biobank and 88% supported a patient database/registry. Comments indicated that the supported function of the biobank was research on improved diagnostics and treatment. Support for a registry was contingent on prioritizing privacy and protection of personal information and samples with a large number of individuals expressing concern about mistreatment arising from linking of medical records would allow those in healthcare to find out about a patient's medical history of Lyme disease.

More than 55% of the respondents indicated that a biobank would need to conform to the following conditions for their support: donors could ask to have samples destroyed, duration of tissue retention would need to be explained in advance, donor identify would be protected, donors would be notified of any findings, donors would be notified of material incidental findings, donors would need to know what research was being conducted, where and by whom, the samples would not be sold and consent would need to be explicit. The three most highly endorsed conditions were rejection of commercial exploitation of samples, notification of unexpected/incidental findings and notification of personal research findings. Conditions of least concern were the more technical issues of tissue storage duration and location of research (but the identity of researchers and nature of researchers were considered important). Additional comments reinforced the need for communication and information protection.

Participants were asked about acceptable registry consent and security procedure. Just over 50% of the 365 respondents felt that consent should always be required, regardless of the anonymity of the data. The most frequently described safeguard, is that the security, protection and privacy of the data and personal information should be a top priority. This involves regulating who can access the information, preventing potential hacking of the patient registry and restricting access to employers, doctors, insurance companies, etc. The majority of

respondents supported either an opt-in only or a combination of opt-in and opt-out model for the registry with only 12% supporting an exclusive opt out model.

Conclusion: Some reoccurring themes arising from this survey is that the protection and security of Lyme patients' personal information and data is of upmost importance. The fear that medical records or data could be accessed by different groups, in particular medical professionals and insurance companies, was a strongly stated and reoccurring them apparent throughout the survey. Another reoccurring theme is that a substantial proportion of Lyme patients want to play an active role in Lyme disease research, be engaged throughout the process and want to be informed of both overall study results and their personal results. Patient support of both a biobank and a patient registry was very strong; indicating a desire to aid in research efforts, particularly those supporting improved diagnostics and treatment. However, in both cases, the need to complete transparency, patient control, privacy and all other attributes high ethical standards was deemed important by most respondents. However, there was evidence of trust in that there was less indication of the need to be engaged in the technical aspects of these projects (where samples would be kept, duration of storage, etc). Finally, many respondents indicated that patients should be involved in setting up the patient registry, either singly or in partnership with other groups.

In summary, this survey indicates strong support of Lyme disease research, including a biobank and patient registry, but only in conjunction of active patient engagement and the highest ethical standards.

Survey structure: The Survey was offered in French and English and included nine questions; the first 7 questions were multiple choice or Yes/No responses and the last two were open ended. All of the questions, excluding question 2, had a comment section or an "other" option which allowed respondents to specify or explain their answers further. No questions were mandatory and respondents withdraw at any time without submitting their survey. The link to the survey was sent to provincial leaders of Lyme advocacy groups in Canada on June 4th 2018. They distributed the link via their respective social media networks for public access. The survey was closed on July 1st 2018. The total number of respondents was 721. The results from the

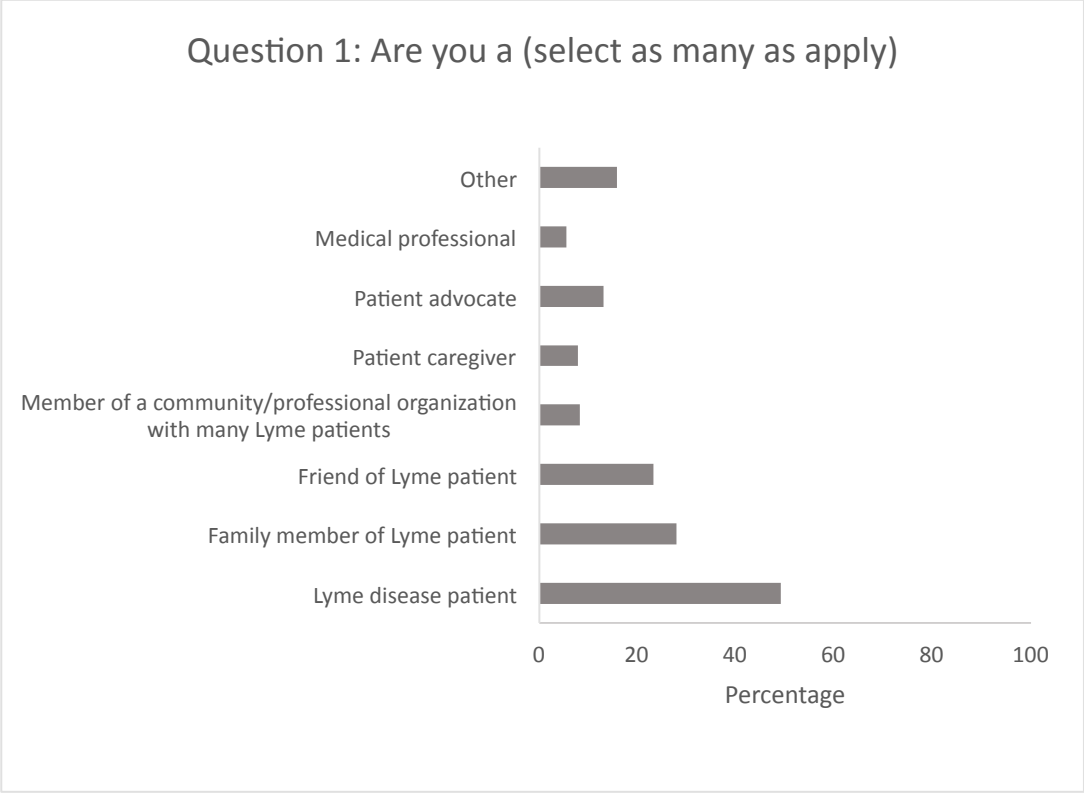
English and French survey were combined and analyzed together. The survey was created and analyzed with the use of SurveyMonkey and some of its accompanying tools; the comment sections and open ended questions were analyzed using a Text Analysis tool offered by SurveyMonkey.

RESULTS

The results of each question were analyzed and graphed. For each question there is an accompanying paragraph that highlights the main results. The number of respondents is shown above each question as an N value.

Question 1 and 2 - Demographics

N=717

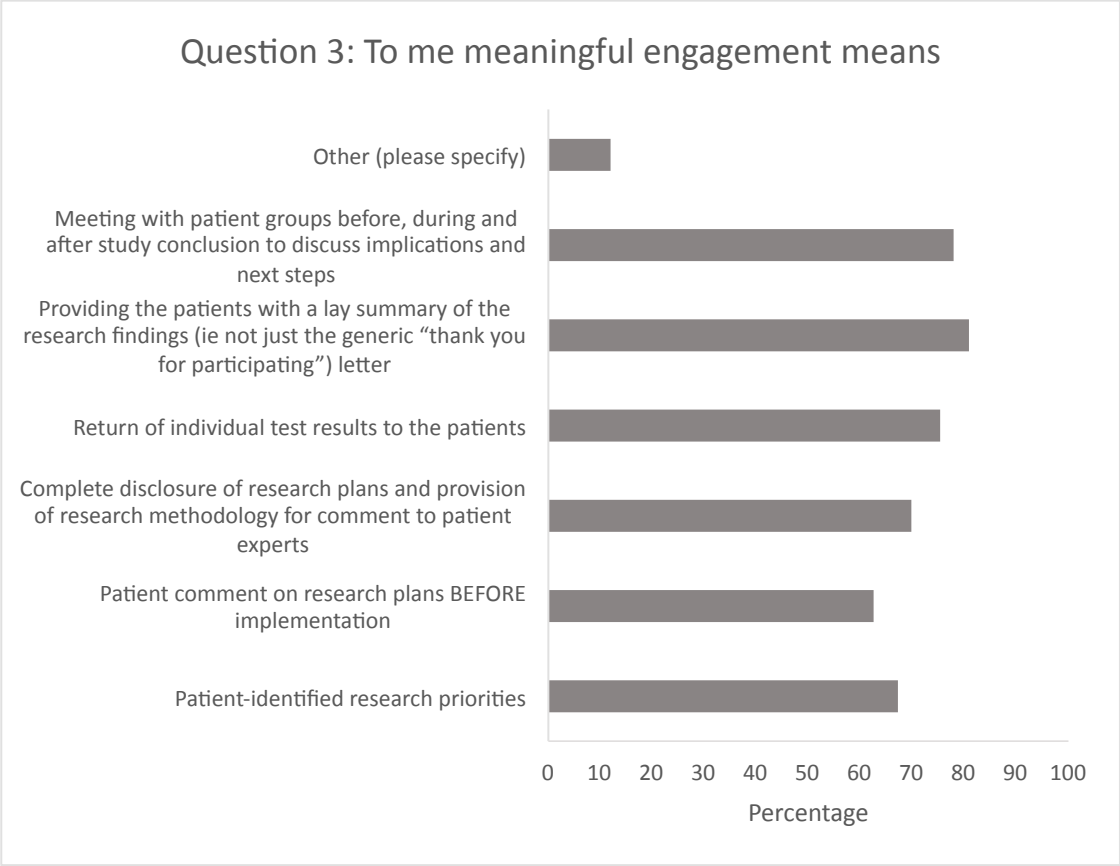


717 respondents answered Question 1. The majority of the respondents are Lyme disease patients (49%) and/or family (27%) or friends (23%) of a Lyme disease patient. 113 respondents chose “other” for Question 1. 28% of these respondents are concerned or

interested citizens and 22% are people with undiagnosed cases of Lyme disease and/or individuals who have been bitten by a tick in the past. 44 respondents selected “medical professional” in question 1. Of those 36% were nurses, medical doctors made up 9% and veterinarians make up another 9% of the Medical Professionals. Dentists, Medical Radiological technologists, medical lab technicians, pharmacy technicians and veterinary technicians each represent 2% of the medical professionals.

Question 3: What does meaningful engagement mean?

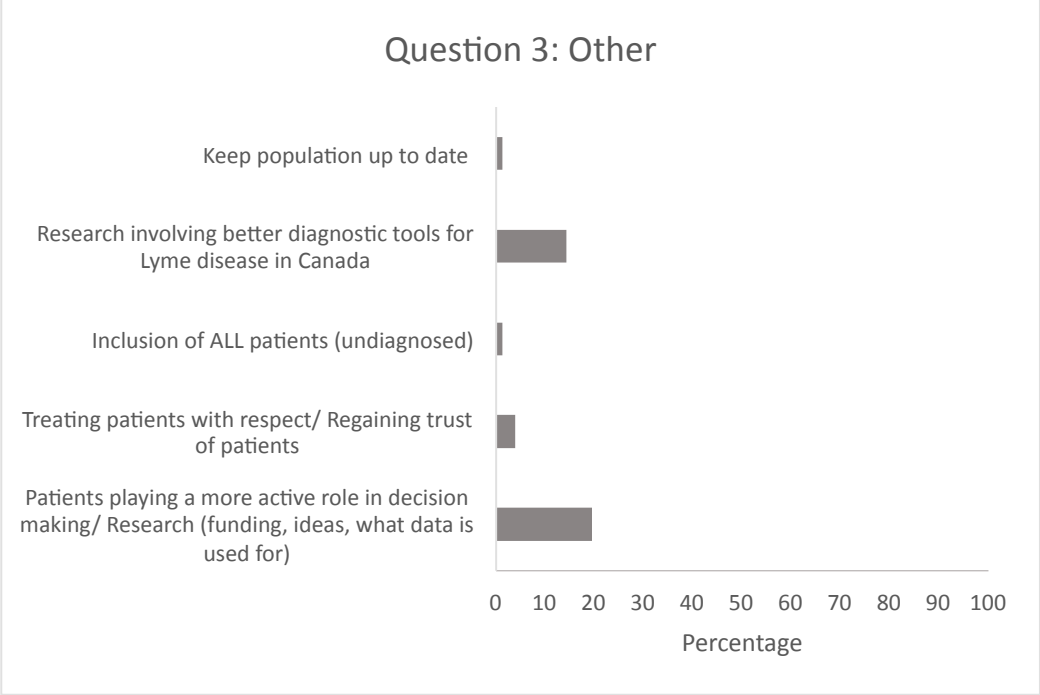
N=643



All of the identified definitions of meaningful engagement were endorsed by over 60% of the 643 respondents. The expectation of a summary of the research findings being provided to

study participants was the most highly endorsed component of acceptable research practices (80%), with 75% of respondents also expressing a desire for the return of individual test results. The expectation of contact throughout a study – before, during and after, also being strongly supported (78%).

Of the 77 respondents who chose "other" in question 3, the two most common comments were to allow patients to play a more active role in decision making in Lyme research (19%) and for research to involve improved diagnostic tools for Lyme disease in Canada (14%). Other suggested components of meaningful research were treating patients with respect, inclusion of all patient groups and good communication with patients.

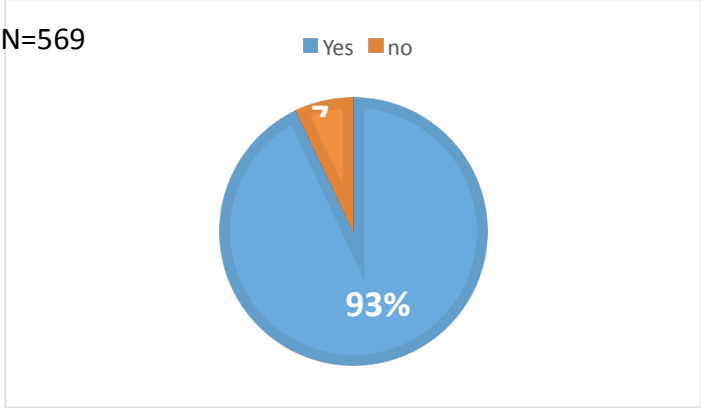


Question 4. Biobank. Participants were asked about the usefulness of a biobank. A biobank was defined as: "One of the priorities identified by the federal government for Lyme disease research is a "biobank" – a collection of blood, urine or other body fluids, or biopsied or autopsied tissues from Lyme disease patients." There were 569 responses to this question with

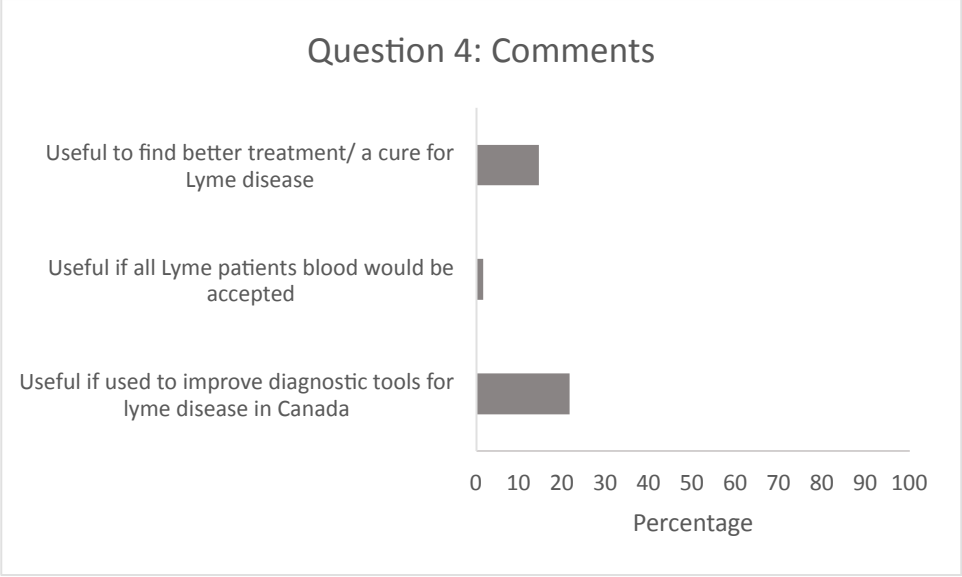
93% of people saying that they felt a biobank is useful for research and only 7% feel it would not be useful. In the comment section of this question, 21% of the comments indicate the biobank would be useful if the research improved diagnostic tools for Lyme disease in Canada, 14% of the comments indicate the biobank would be useful if it is used to research better treatments for Lyme disease patients and eventually find a cure.

Question 4. I feel that a biobank is useful for research:

N=569

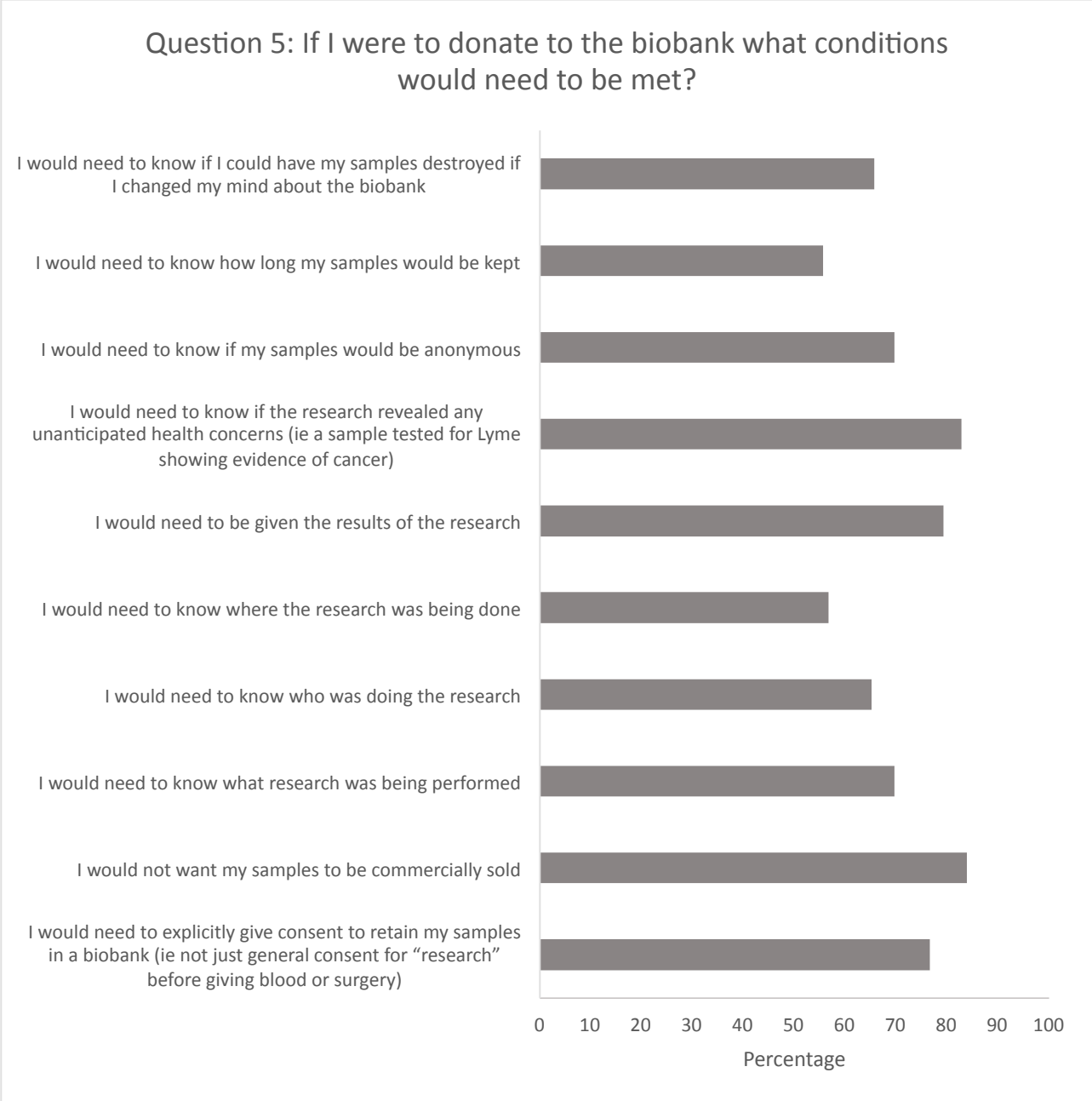


Comments, N=325



Question 5: Conditions required for endorsement of a biobank

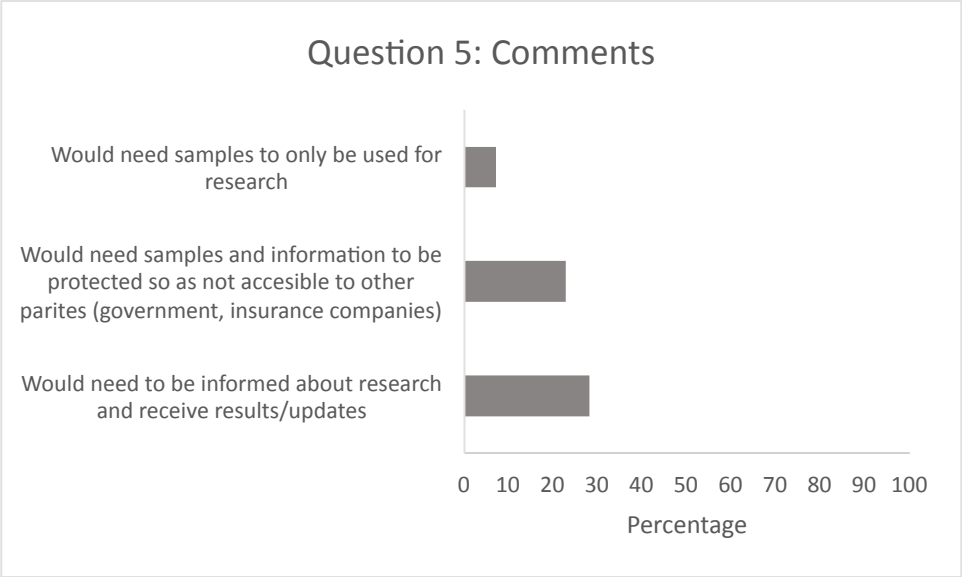
N=559



All of the listed conditions needed to donate to a biobank were supported by at least 55% of the 559 respondents. The three most highly endorsed conditions were rejection of commercial exploitation of samples (84%), notification of unexpected/incidental findings (83%)

and notification of personal research findings (79%). Additional comments reinforced the need for communication, information protection and use of samples restricted to research; 28% of those who wrote a comment say they would need to be informed about the research and receive updates, 22% say that their information would need to be protected (not accessible to the government, insurance companies, or unauthorized personnel) and 7% specifying that their samples would need to be restricted to research uses.

Comments, N=57



Question 6: Patient Registry: Participants were asked about support for a patient registry.

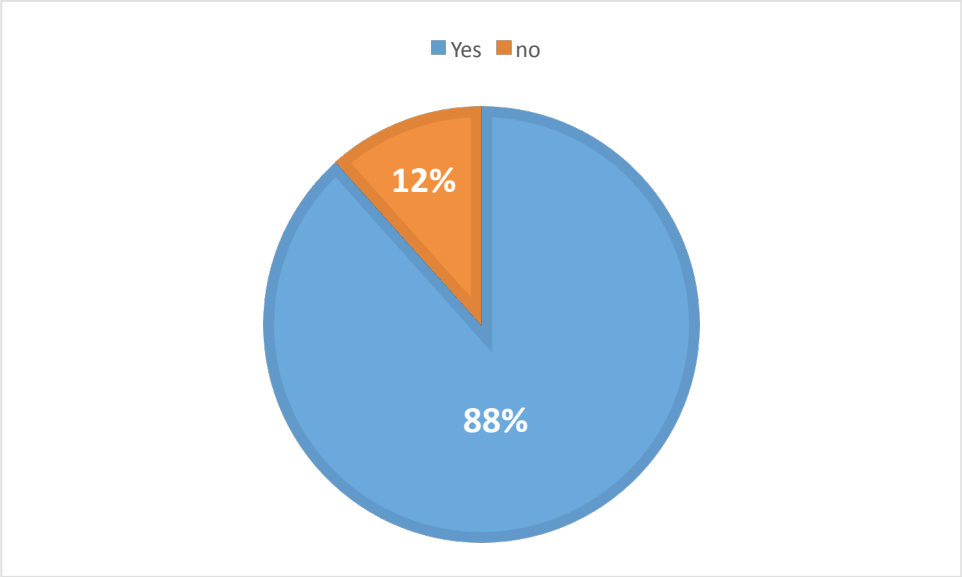
A patient registry was defined as: "A patient registry is a database with information about patients. This could be simply a list of emails so people can be updated about Lyme-related information or it could be complex medical information such as obtained by linking medical records – for example linking your family doctor's files on you with National Microbiology Laboratory (NML) test results and pharmacy prescriptions. Patient registries can also be patient-initiated – an example of this is MyLymeData in the US."

88% of the 509 respondents indcated that a patient registry would be useful for research. The comment section included 143 comments and indicated that 34% support a registry only if

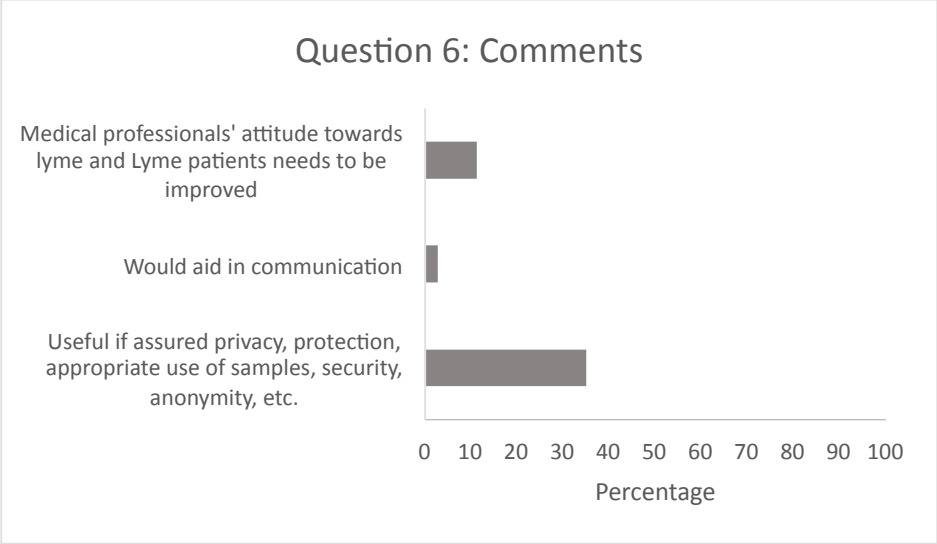
privacy and protection of personal information and samples are a priority. 11% of comments expressed concern about mistreatment arising from linking of medical records and/or revealing a patient's medical history of Lyme disease.

I feel that a registry is useful for research:

N=509



Comments, N=143

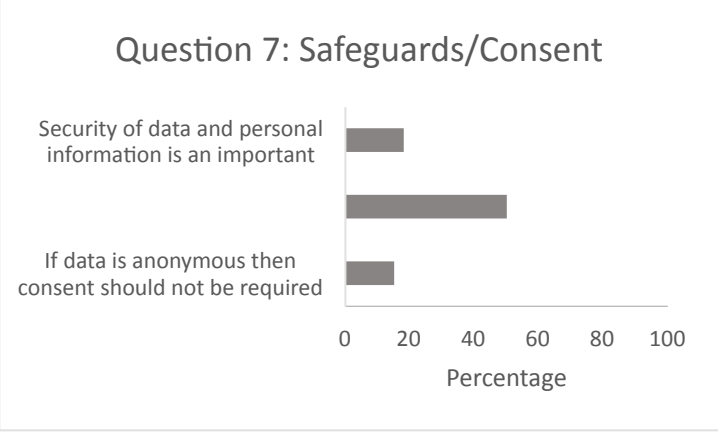


Question 7: Consent: Participants were asked about acceptable registry consent procedure. The questions were "What safeguards would be necessary for a registry? Should consent be required to enter patient information into a registry? If only anonymized information (names have been removed from the information) is used in a registry is consent required?"

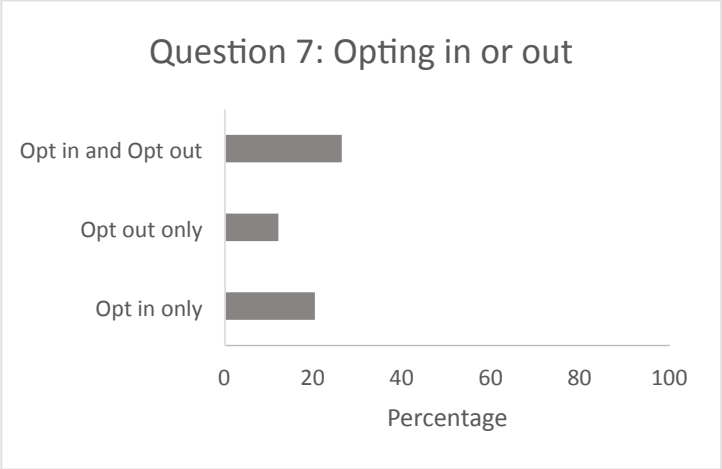
The first part of the question asks what safeguards would need to be in place for a patient registry and when consent should, or should not, be required. Just over 50% of the 365 respondents felt that consent should always be required, regardless of the anonymity of the data. 15% of respondents feel that if the data is anonymized, then consent should not be required as this could hinder research. The most frequently described safeguard, (18%) is that the security/protection/privacy of the data and personal information should be a top priority. This involves regulating who can access the information, preventing potential hacking of the patient registry and restricting access to employers, doctors, insurance companies, etc.

In the follow up question, whether the registries should be Opt in (you have to agree to have your data included) or opt out (you have to ask for your data to be removed) or a combination of both (where you have to agree to have your data included, but can have it removed upon request), of the 365 respondents, 26% indicated that a combination of opt in and opt out system would be preferred. 20% of respondents indicate that the registry should be opt in only and 12% indicate the registry should be opt out only.

N=365



N=365

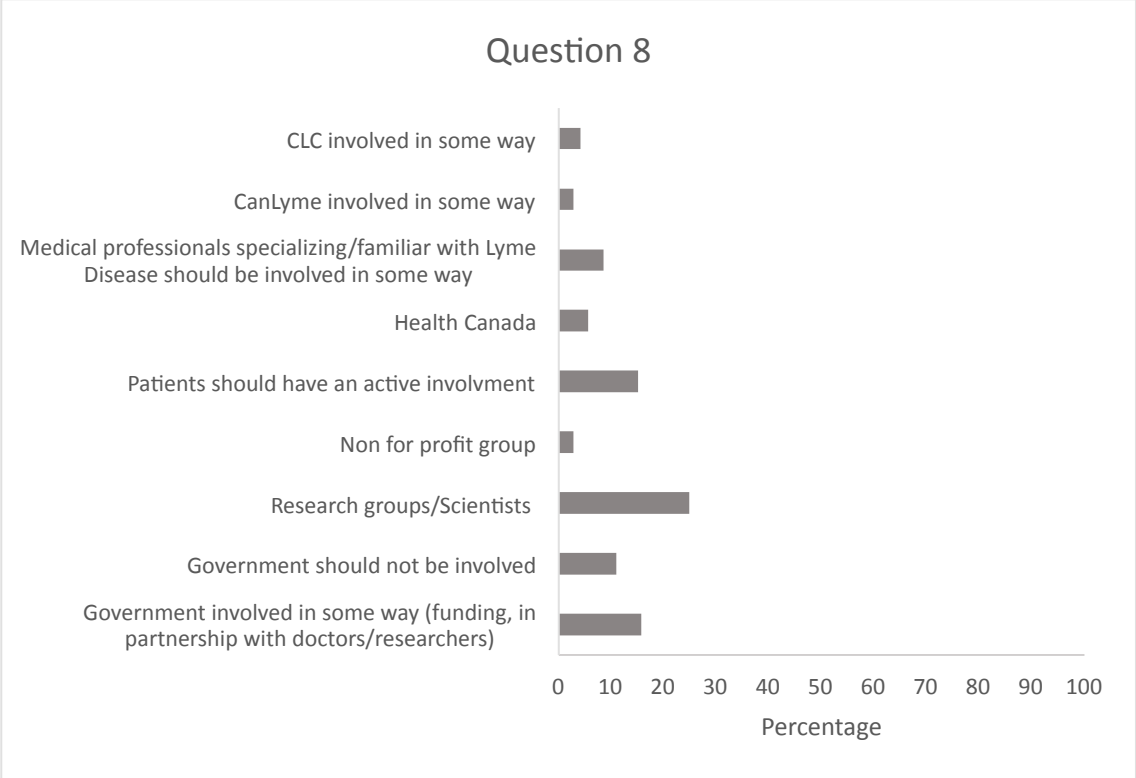


Question 8: Who should be setting up a registry?

Most of the 319 responses indicated a preference for multiple groups working together; for simplicity the responses presented here were analyzed so that the frequency of each group (i.e. number of responses indicating researchers) was counted independently of the other groups indicated in the response (i.e. researchers and medical professionals).

The most frequent response for the group who should be responsible for setting up a registry was “researchers or scientists”, at 25%. Other frequent comments were that patients should have an active role in setting up the registry (15% of responses), the government should be involved in some way (15%) and the government should not be involved in any way (10%). 6% of responses supported the Public Health Agency of Canada (Health Canada) being involved in setting up the registry, which would be associated with the government. 8% of responses indicated that medical professionals should be involved in setting up the registry.

N=319



Global conclusions:

Some reoccurring themes arising from this survey is that the protection and security of Lyme patients' personal information and data is of upmost importance. The fear that medical records or data could be accessed by different groups, in particular medical professionals and insurance companies, was a strongly stated and reoccurring them apparent throughout the survey; most notably in questions 5, 6 and 7. Another reoccurring theme is that a substantial proportion of Lyme patients want to play an active role in Lyme disease research, be engaged throughout the process and want to be informed of both overall study results and their personal results. This is apparent throughout the survey; in questions 3 and 5, all the choices that entail patients receiving more information about research were chosen at high frequencies. Patient support of both a biobank and a patient registry was very strong; indicating a desire to aid in research efforts, particularly those supporting improved diagnostics and treatment. However, in both cases, the need to complete transparency, patient control, privacy and all other attributes of TCPS 2.0 (and proposed 3.0) ethical standards was deemed important by most respondents. However, there was evidence of trust in that there was less indication of

the need to be engaged in the technical aspects of these projects (where samples would be kept, duration of storage, etc). Finally, 15% of respondents think that patients should be involved in setting up the patient registry, either singly or in partnership with other groups.

In summary, this survey indicates strong support of Lyme disease research, including a biobank and patient registry, but only in conjunction of active patient engagement and the highest ethical standards.

Survey Authors: CLC steering group: Vett Lloyd, Ph.D.; Melanie Wills, Ph.D.; Janet Sperling, Ph.D. candidate; Liz Zubek, MD; Jim Wilson, CanLyme; Sue Faber & Jennifer Kravis, LymeHope.
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