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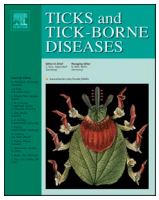

Original article

Self-reported tick exposure as an indicator of Lyme disease risk in an endemic region of Quebec, Canada

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A R T I C L E I N F O

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A B S T R A C T

Background: Lyme disease (LD) and other tick-borne diseases are emerging across Canada. Spatial and temporal LD risk is typically estimated using acarological surveillance and reported human cases, the former not considering human behavior leading to tick exposure and the latter occurring after infection.
Objectives: The primary objective was to explore, at the census subdivision level (CSD), the associations of self-reported tick exposure, alternative risk indicators (predicted tick density, eTick submissions, public health risk level), and ecological variables (*Ixodes scapularis* habitat suitability index and cumulative degree days > 0 °C) with incidence proportion of LD. A secondary objective was to explore which of these predictor variables were associated with self-reported tick exposure at the CSD level.
Methods: Self-reported tick exposure was measured in a cross-sectional populational health survey conducted in 2018, among 10,790 respondents living in 116 CSDs of the Estrie region, Quebec, Canada. The number of reported LD cases per CSD in 2018 was obtained from the public health department. Generalized linear mixed-effets models accounting for spatial autocorrelation were built to fulfill the objectives.
Results: Self-reported tick exposure ranged from 0.0 % to 61.5 % (median 8.9 %) and reported LD incidence rates ranged from 0 to 324 cases per 100,000 person-years, per CSD. A positive association was found between self-reported tick exposure and LD incidence proportion ($\beta = 0.08$, CI = 0.04,0.11, $p < 0.0001$). The best-fit model included public health risk level (AIC: 144.2), followed by predicted tick density, ecological variables, self-reported tick exposure and eTick submissions (AIC: 158.4, 158.4, 160.4 and 170.1 respectively). Predicted tick density was the only significant predictor of self-reported tick exposure ($\beta = 0.83$, CI = 0.16,1.50, $p = 0.02$).
Discussion: This proof-of-concept study explores self-reported tick exposure as a potential indicator of LD risk using populational survey data. This approach may offer a low-cost and simple tool for evaluating LD risk and deserves further evaluation.

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1. Introduction

Lyme disease (LD) is an emerging bacterial disease in Canada caused by *Borrelia burgdorferi* sensu stricto, which is transmitted through the bite of an infected tick (Ogden et al., 2015). It is the most frequently reported vector-borne disease in temperate countries (Mead, 2015; Schwartz et al., 2017). In Canada, the number of reported cases has increased from 144 cases in 2009, when the disease became notifiable, to 2851 cases in 2021 (Public Health Agency of Canada, 2022). The species of tick vector varies geographically, with *Ixodes scapularis* being the principal vector in eastern and central North America and *Ixodes pacificus* the main vector west of the Rocky Mountains (Ogden et al., 2009).

Lyme disease is nationally notifiable in Canada and under passive surveillance whereby physicians, nurse practitioners and laboratories are required to report confirmed LD cases through the public health reporting system. There is also acarological surveillance, which comprises passive and active surveillance methods. Active tick surveillance is performed to identify where tick populations are establishing and to confirm the presence of ticks infected with *B. burgdorferi* within an endemic area (Government of Canada, 2007; Public Health Ontario, 2015; Salomon et al., 2020). Resulting entomological measures have widely been used as proxies for LD risk, although they may not translate so well at fine spatial scales (Eisen and Eisen, 2016; Mather et al., 1996; Pepin et al., 2012; Stafford et al., 1998). Passive tick surveillance includes tick submissions sent by participating health-care professionals, veterinarians and/or the public to regional health units or provincial laboratories, and has been shown to provide an early indication of LD risk at the regional level (Gasmi et al., 2019; Koffi et al., 2012; Ogden et al., 2010; Ripoche et al., 2018). Furthermore, several provinces are now promoting eTick, an online citizen science platform to which users can submit tick photos for identification, as their primary or sole method of passive surveillance (https://www.etick.ca/en). These surveillance methods have their limitations (Ripoche et al., 2018). Active tick surveillance requires significant resources and is not feasible at a large scale in every region of Canada. Passive tick surveillance is strongly associated with human population density, lacks specificity in identifying established tick populations (Koffi et al., 2012), and may be limited by laboratory resource capacity, as evidenced by the reduction of such activities in some endemic regions (Guillot et al., 2022). Finally, the surveillance of human LD cases does not provide an 'early warning' signal of emerging risk, may be affected by under-reporting, and does not provide the same level of temporal or spatial detail as active tick surveillance (Gasmi et al., 2017a, 2017b; Naleway, 2002; Ogden et al., 2019; Ripoche et al., 2018).

Both human behavior and acarological risk affect the probability of being bitten by a tick, and more recently, the concepts of behaviors, vulnerability and coping capacity are being integrated into risk analysis (Bouchard et al., 2022; Fischhoff et al., 2019; Vanwambeke and Schimit, 2021). In comparison to active tick surveillance methods, passive surveillance of ticks found on people, or their immediate environment, takes into account these elements. Self-reported tick exposure, whereby tick exposure is reported 'passively' but not followed up with submission of the tick for identification may offer another means of evaluating LD risk, particularly in contexts where active or passive acarological surveillance activities are reduced. Relatively recent studies have evaluated and demonstrated the utility of self-reported tick exposure as an indicator for LD, either at the individual or household level, in self-reported LD patients, or through a citizen science project (Hook et al., 2021; Maxwell et al., 2021; Porter et al., 2019). Other research suggests that reported tick exposure by an individual does not reflect true tick exposure (Schmid et al., 1985; Schwartz and Goldstein, 1990). However, to our knowledge, self-reported tick exposure measured across a general population has not been evaluated as an indicator for LD at the regional level. The primary objective of this cross-sectional study was therefore to evaluate self-reported tick exposure across the LD endemic Estrie region of Quebec, Canada, as an indicator for LD risk at the census subdivision (CSD) level. We examined the associations of LD incidence proportion with this potential indicator, as well as other alternative risk indicators derived from active and passive surveillance, public health risk level and ecological drivers of *I. scapularis*. A secondary objective was to explore if the alternative risk indicators and ecological drivers were associated with self-reported tick exposure at the CSD level.

2. Materials and methods

2.1. Study design

The key variable of interest, self-reported tick exposure, was measured in a cross-sectional populational health survey conducted in 2018 by the public health department of Estrie (*Direction de Santé Publique (DSP) de l'Estrie*), funded by *le Centre intégré universitaire de santé et de services sociaux de l'Estrie - Centre hospitalier universitaire de Sherbrooke*. Other variables were obtained from secondary data sources, described under 'Other data sources', below. Only CSDs represented by at least one respondent of the populational health survey were included in this study, resulting in data for 116 out of 121 CSDs in Estrie. All variables were computed and analysed at the CSD level.

2.2. Setting

The region of Estrie is an administrative region in southeastern Quebec, Canada (Fig. 1), with a population of ~ 499,155 inhabitants in 2021 (Institut de la statistique du Québec, 2023) and an area of 10,197 km^2. The Estrie region has a well-established *I. scapularis* tick population which is endemic for *B. burgdorferi* sensu stricto (Gouvernement du Québec, 2021). The number of reported LD cases with probable acquisition in Estrie accounted for 69 % of the total number of LD cases of probable acquisition in Quebec in 2021, and its neighbouring region, Montérégie, accounted for a further 19 % (Ministère de la Santé et des Services Sociaux du Québec, 2023). The incidence rate of LD in Estrie was estimated to be 77.1 cases per 100,000 person-years in 2021 (CIUSSS de l'Estrie - CHUS, 2022), with the number of incident human LD cases likely acquired in the Estrie region increasing from 28 in 2014 to 451 in 2021 (Ministère de la Santé et des Services Sociaux du Québec, 2021). The region is divided into nine health subregions (*Réseaux locaux de services*, RLS), which are further divided into 121 census subdivisions (CSDs). CSDs are defined as "*…municipalities (as determined by provincial/territorial legislation) or areas treated as municipal equivalents for statistical purposes (*e.g.*, Indian reserves, Indian settlements and unorganized territories)*" (Statistics Canada, 2016). In 2019, administrative changes to the health subregions meant that the CSD of Bromont went from being part of the Haute-Yamaska subregion (512) to being part of the Pommeraie subregion (511). For local stakeholders, it is important to note that in this study, Bromont is considered to be part of the Haute-Yamaska health subregion as the data were obtained in 2018.

2.3. Variables, data sources and measurement

Below, we provide a description of the data sources and measurement of each variable used in this study. The computation of the variables is described in Table 1.

2.3.1. Self-reported tick exposure

Participants (≥ 18 years old) of the 2018 Estrie population health survey were sampled randomly, stratified by population density of each health subregion (RLS) from June to November 2018. In addition to other health themes, participants were asked whether they had found a tick on themselves or someone from their family in the last 12 months (see Q3 of the questionnaire available in Supplementary material 1). The detailed methodology, and results of these survey data related to the individual behavioral and environmental risk factors for tick exposure

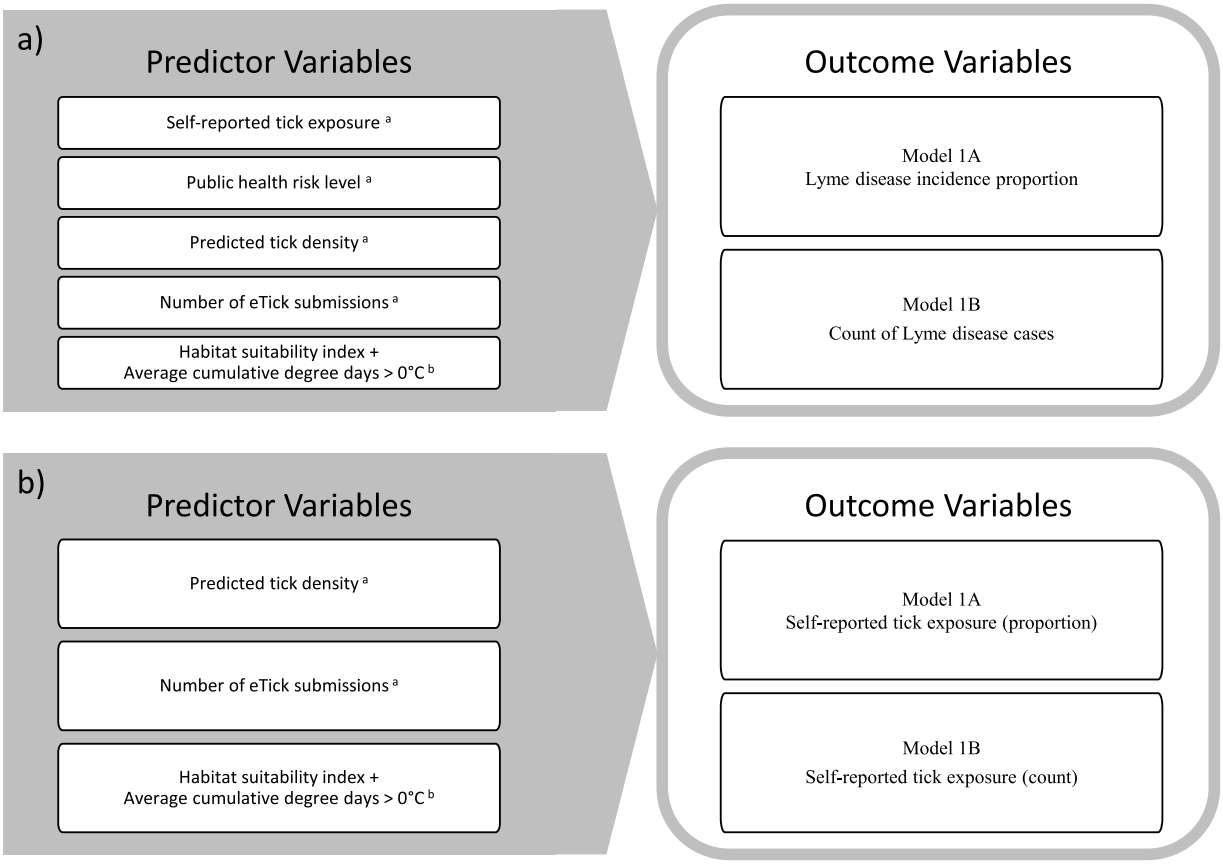

Fig. 1. Predictor variables included in a) Model 1 groups (outcome variable = Lyme disease incidence proportion [Model 1A] or count of reported Lyme disease cases [Model 1B]), and b) Model 2 groups (outcome variable = proportion of respondents self-reporting tick exposure [Model 2A] or count of respondents reporting tick exposure [Model 2B].
[a] Predictor included in a univariable model.
[b] Habitat suitability index and average cumulative degree days were combined in a multivariable model.

and the adoption of protective behaviors against tick bites, have been published elsewhere (Aenishaenslin et al., 2022). The postal code of each participant was used to determine the CSD of residence.

2.3.2. Lyme disease case count and incidence

The human population of each CSD was obtained from Statistics Canada (2016) census data (Statistics Canada, 2017). The total number of reported human cases from each CSD in Estrie in 2018 were obtained from the public health department of Estrie (*Direction de Santé Publique (DSP) de l'Estrie*). The incidence rate of LD per 100,000 person-years for each CSD was estimated using the total number of reported cases in 2018 and the population size as measured in the 2016 census and is used only to provide descriptive statistics results. Incidence proportion and total number of reported cases per CSD were used as outcome variables in the statistical models, as described in Table 1.

2.3.3. Public health risk level for Lyme disease

In Quebec, tick surveillance is coordinated by the *Institut National de Santé Publique du Québec* (INSPQ) and results are used to classify and monitor disease acquisition risk levels across Quebec's municipalities to aid authorities in risk management (Institut national de santé publique du Québec, 2021). Three levels of LD risk by CSD, as measured by the INSPQ, were used in this study as a comparable indicator of LD risk. These levels were defined using thresholds for the number of locally acquired human cases, the number of *I. scapularis* specimens submitted through passive tick surveillance activities, and presence of different *I. scapularis* life stages observed during active tick surveillance activities (https://www.inspq.qc.ca/zoonoses/maladie-de-lyme). To be defined as a risk level 1, at least 2 cases of locally acquired LD must have been recorded in the past five years, or between 11 and 22 specimens of *I. scapularis* of human origin be obtained through passive surveillance, or at least one specimen of *I. scapularis* (larva, nymph, adult) be obtained through active surveillance. To be defined as a risk level 2, at least 3 cases of locally acquired LD must have been recorded in the past five years, or 23 specimens of *I. scapularis* of human origin be obtained through passive surveillance, or the three life stages (any life stage) be collected through active surveillance with at least one nymph tested positive for *B. burgdorferi*. CSDs for which data were absent or did not support the allocation of risk level 1 or 2 were defined as risk level 0. It is important to note there is still a risk of acquiring LD in a risk level 0 region. While it seems counterintuitive to consider public health risk level as a predictor for incidence proportion, given that it is formulated in part by LD cases, we considered this 'predictor' as an indicator to which other indicator variables could be compared.

2.3.4. Predicted tick density and eTick submission data

Predicted tick density for 2018 was estimated using active field surveillance data for *I. scapularis* collected between 2007 and 2017 (Bouchard et al., 2018). These surveillance activities comprised a standardised drag sampling method, the protocol and site selection which are described in full elsewhere (Bouchard et al., 2011, 2015, 2018; Ogden et al., 2010). Briefly, the annual increase in tick numbers was modelled to produce standardised predicted tick density values for 113 of the 116 CSDs included in this study. The three CSDs without predicted tick density estimates were Milan, Newport and St-Benoît-du-Lac and were not included in the analyses using this variable.

Table 1
A summarised description of the covariates used in the models for this study and how they were computed.

Objective 1 predictor variables	
Self-reported tick exposure	This was computed, at the CSD level, as the percentage of participants reporting a tick exposure (i.e., at least one tick bite and/or tick found on body) on themselves or a family member within the previous 12 months.
Predicted tick density	A standardised value, modelled from active field surveillance data collected between 2007 and 2017. Refer to Bouchard et al. (2018) for full details on methods(Bouchard et al., 2018).
eTick submissions per CSD	The number of positively identified *I. scapularis* tick submissions from people, pets, and the environment, per CSD, between 2018 and 2020.
Public health risk level	Public health risk levels are defined by the INSPQ and categorised as 0, 1 or 2.(Institut national de santé publique du Québec, 2021) A risk level of 0 represents a constant low level of risk posed by adventitious ticks, a risk level of 1 represents an increasing level of risk, while a risk level of 2 represents a significant risk found in endemic areas.
Number of cumulative degree days >0 °C	This value represents the averaged sum of each monthly value from 2014 to 2018.
Habitat suitability index	The probability that a CSD is suitable for *I. scapularis* (i.e., offering optimal conditions for the presence of *I. scapularis*), based on deciduous and mixed forest land cover.
Objective 1 outcome variable	
Lyme disease risk	The incidence proportion of LD in each CSD for 2018 was estimated by dividing the total number of reported human cases in 2018 per CSD by the population of that CSD in 2016, assuming a stable population [a]. A count variable for the number of reported human cases of LD was also modelled (Model 1B group).
Objective 2 predictor variables	
Predicted tick density	*As described above*
eTick submissions per CSD	*As described above*
Habitat suitability index	*As described above*
Number of cumulative degree days >0 °C	*As described above*
Objective 2 outcome variable	
Self-reported tick exposure	This was computed as the proportion (i.e., a decimal value) of participants reporting tick exposure on themselves or a family member within the previous 12 months. A count variable representing the number of respondents reporting tick exposure was also modelled (Model 2B group).

[a] We did not consider changes in population over time and could not take into account individuals who may have received more than one diagnosis in that year, which are limitations.

CSD: Census subdivision.

INSPQ: Public health institute of Quebec (*Institut National de Santé Publique du Québec*).

LD: Lyme disease.

The full details of this modeling are described by Bouchard et al. (2018, 2022).

As an alternative indicator of risk based on tick surveillance, data were obtained from eTick, a citizen science platform (https://www.etick.ca/). Images of ticks collected from individuals, animals, or the environment are submitted by the public for identification by trained personnel, and the tick species is recorded with other information including the probable locality of acquisition and date of collection. For this study, we used the number of *I. scapularis* eTick submissions per CSD, submitted between 2018 and 2020. This time frame was chosen as there were insufficient data from the first year alone to explore eTick submissions as a signal for LD risk, given eTick was launched in 2017. The proportion of *I. scapularis* submissions from animals between 2018 and 2020 was approximately 70 % (personal communications, Jade Savage, 2023).

2.3.5. Ecological drivers

A habitat suitability index (HSI) for *I. scapularis* was included as one of two ecological drivers of *I. scapularis*. This index follows the form of a logistic function that includes a parameter corresponding to the proportion of forest cover. The proportion of deciduous and mixed forests for each CSD was calculated using 2015 land cover data obtained from the North American Land Change Monitoring System (data available at http://www.cec.org/north-american-land-change-monitoring-system/). The index value lies between 0 and 1 and can be interpreted as the probability that a CSD is suitable for *I. scapularis* (i.e., offering optimal conditions for the presence of *I. scapularis*), depending on the threshold of forest proportion chosen. The value of zero indicates an unsuitable CSD, whereas higher values towards one indicate highly suitable CSDs for *I. scapularis*. Preliminary results showed that the best models of LD risk, based on Akaike information criterion (AIC), included a threshold of 15 % and so we present results using this threshold. The full methodological details of how this variable was modelled are available in a prior publication (Tardy et al., 2023).

The second ecological variable used in this study was the number of cumulative degree-days above 0 °C, which can indicate the suitability of conditions for certain plants and pests. Specifically for *I. scapularis*, values over the threshold of 2800–3100 have been proposed as favouring tick establishment (Ogden et al., 2005). Data were obtained from Climate Data (https://climatedata.ca/), with the value representing the averaged sum of each monthly value from 2014 to 2018, per CSD.

2.4. Statistical analyses

The data were compiled from the previously described sources. Analysis was limited to the 116 CSDs (out of 121 CSDs in Estrie) for which data were collected in the 2018 Estrie health survey.

To fulfill the two objectives, four different groups of models were built (illustrated in Fig. 1), and AIC values were used to compare the fit of the predictor variables to the outcome. For objective 1, univariable models were built to compare the fit between self-reported tick exposure, as well as alternative risk indicators (public health risk level, predicted tick density, eTick submissions), and two LD frequency outcome variables; the incidence proportion of LD (Model 1A group) and the total count of reported LD cases (Model 1B). In addition, multivariable models were built using two ecological variables (habitat suitability index for *I. scapularis* and average cumulative degree days > 0 °C) as predictor variables for LD incidence proportion (Model 1A group) and count of reported cases (Model 1B group). To compare the fit between alternative risk indicators and ecological covariates with self-reported tick exposure (objective 2), univariable and multivariable models were built using two outcome variables: the proportion (Model 2A group) and count (Model 2B group) of self-reported tick exposure per CSD. Predicted tick density and eTick submissions were included as predictors in univariable models, and habitat suitability index for

I. scapularis and average cumulative degree days > 0 °C were included as predictors in the multivariable models. As public health risk level includes count of LD cases (i.e., a downstream event to tick exposure), this variable was not tested as a predictor for self-reported tick exposure in Objective 2.

Models 1A and 2A groups (having a proportion outcome variable) were tested using a logit link function and assuming a binomial outcome distribution, including population and total number of survey respondents as weights in the respective models. Models 1B and 2B groups (having a count outcome variable) were built with a log link function assuming either a negative binomial or Poisson distribution depending on overdispersion, with the log of the total population and log of the total number of survey respondents included as offsets in the respective models.

The analyses were performed using generalised linear mixed models (GLMMs) and generalised additive models. GLMMs provided a better quality of fit and were therefore chosen to present results. The Moran's I test was performed and identified the presence of spatial autocorrelation in the residuals of the regression models, which determined that a spatial covariance structure should be included in the regression models. AIC and DHARMa diagnostic plots and tests were also used to determine the inclusion of a spatial covariance structure. Furthermore, GLMMs also included the nine health subregions (RLS) as a random effect, to account for spatial autocorrelation at the sub-regional level. Standardised longitude and latitude centroids of each CSD were used for application of methods to account for spatial autocorrelation. ArcGIS software (version 10.6.1) was used to display results using maps.

Problems with convergence of models were addressed using variable centering, standardization and/or by changing the optimization algorithms, i.e. using the Broyden-Fletcher-Goldfarb-Shanno algorithm instead of the default optimiser (nlminb). Other changes considered in cases of poor convergence, or according to AIC values and DHARMa tests, included the use of Generalised Poisson or, Conway-Maxwell-Poisson as appropriate. All analyses and data visualization were done using the R statistical software version 4.1.1 (R Core Team, 2021), using following packages: glmmTMB (Brooks et al., 2017), mgcv (Wood, 2011), DHARMa (Hartig, 2021), performance (Lüdecke et al., 2021), MASS (Venables and Ripley, 2002), pscl (Zeileis et al., 2008), broom.mixed (Bolker and Robinson, 2021), and tidyverse (Wickam et al., 2019).

3. Results

3.1. Participants

In total, 10,790 participants responded to the survey, from 116/121 CSDs. The five CSDs with no data were Bedford, Hatley, Stanstead, St-Robert-Bellarmin, and Valcourt. Descriptive statistics for participant demographics can be found in a prior publication of behavioral risk factors associated with tick exposure (Aenishaenslin et al., 2022; Bouchard et al., 2022).

3.2. Descriptive statistics by public health risk level and health subregion

The percentage of residents from each CSD who responded to the 2018 health survey ranged from 0.1 to 8.7 % (Fig. 2). Descriptive statistics for CSDs by public health risk level and health subregion are provided in Table 2 and Table 3, respectively. CSDs assigned a public

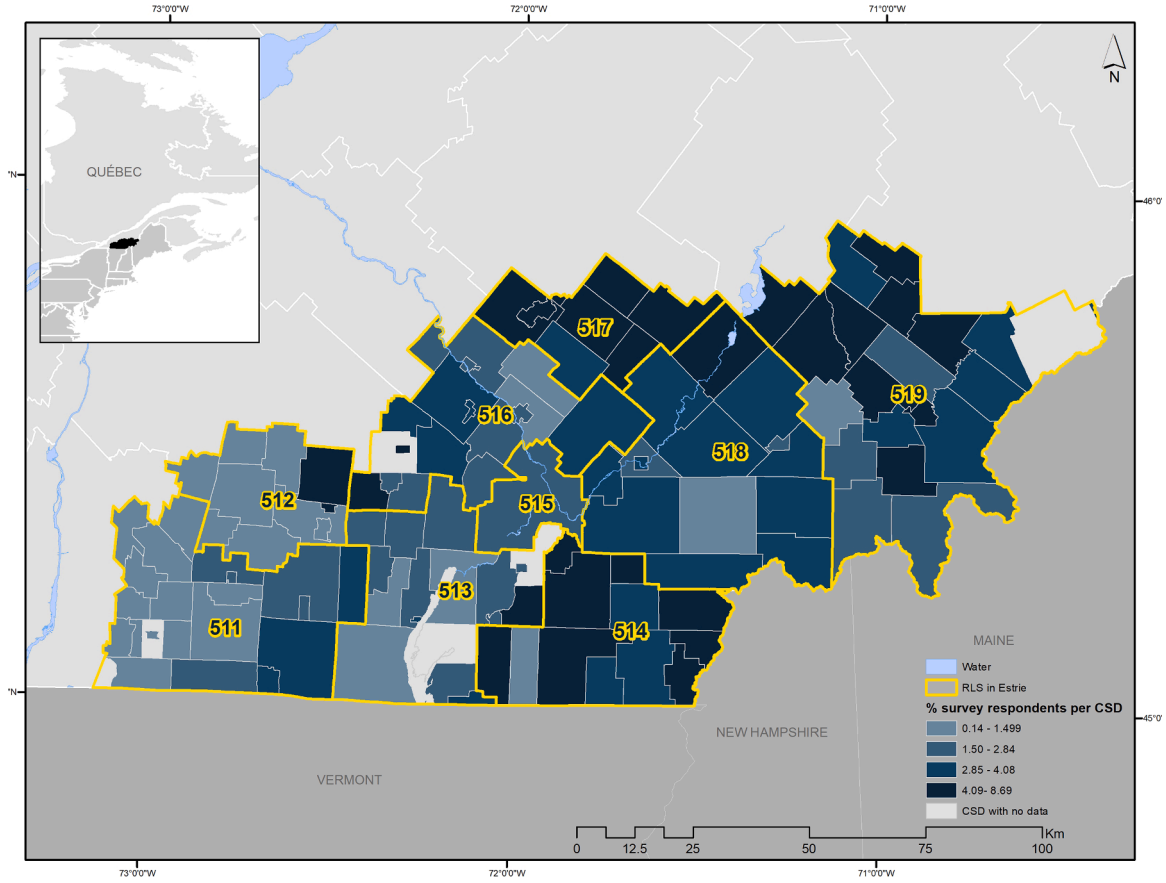

Fig. 2. Percentage of the population responding to the 2018 Estrie populational health survey, by census subdivision.
The population values for each census subdivision were obtained from 2016 census data. Quantile breaks were used to classify the data. RLS = health subregion (*réseaux locaux de services, n* = 9). CSD = census subdivision (data available for 116/121 CSDs). RLS names: Pommeraie (511), Haute-Yamaska (512), Memphrémagog (513), Coaticook (514), Sherbrooke (515), Val-Saint-François (516), Asbestos (517), Haut-Saint-François (518), Granit (519).

Table 2
Descriptive statistics of the regional demographic data, Lyme disease incidence rate, and predictor variables by public health risk level in Estrie, Quebec for this study. Values were obtained by calculating the average value of CSDs included in the dataset ($n = 116/121$).

	Public health risk level for 2018		
	0	1	2
No. CSDs	79	23	14
Total population (2016)	105,628	204,899	156,640
Population density (/km^2, 2016)	36.72	113.09	94.17
Total no. LD cases (2018)	3	6	80
LD incidence rate/100,000 person-years (2018)	1.97	20.59	104.51
Total no. survey respondents (2018)	3523	5056	2211
Self-reported tick exposure (%, 2018)	8.68	16.35	22.34
Mean predicted tick density (2018) [a]	−0.39	−0.13	0.19
Total no. eTick submissions (2018–2020)	83	127	280
Mean no. eTick submissions per CSD	1.05	5.52	20.00
Mean habitat suitability index (10 % cover, 2015)	0.94	0.75	0.97
Mean habitat suitability index (15 % cover, 2015)	0.89	0.66	0.94
Mean cumulative degree days >0 °C (2014–2018)	3075.91	3351.77	3389.10

[a] Predicted tick density: standardised values.
INSPQ = National public health institute of Quebec (Institut national de santé publique du Québec).
CSD = Census subdivision.

health risk level of 2 ($n = 14/116$) had, on average, the highest incidence of LD cases (105 per 100,000 person-years), the highest percentage of survey respondents reporting tick exposure (22.3 %), the highest number of eTick submissions and mean number of eTick submissions per CSD between 2018 and 2020 (280 and 20, respectively) as well as the highest values for predicted tick density, habitat suitability index and average cumulative degree days > 0 °C (0.19, 0.94, and 3389.10, respectively). CSDs assigned public health risk levels of 0 and 1, on average, reported lower incidences of LD (1.97 per 100,000 person-years and 20.59 per 100,000 person-years respectively). The percentage of survey respondents reporting tick exposure was also lower, with 8.7 % reporting tick exposure in CSDs assigned a public health risk level of 0 and 16.4 % in CSDs assigned a public health risk level of 1.

The health subregions of Pommeraie and Haute-Yamaska had the highest incidence of LD of all health subregions in Estrie in 2018 (66.4 per 100,000 person-years and 43.9 per 100,000 person-years, respectively). Respondents from these regions reported the highest percent of tick exposure within Estrie (22.7 % and 17.4 %, respectively) and this is where the predicted tick densities were highest (0.30 and 0.02 respectively), as was the cumulative degree days above 0 °C (3464.2 and 3385.8). In contrast, there were no reported cases of LD in Coaticook, although 9.2 % of survey respondents reported tick exposure. More

Table 3
Descriptive statistics of the regional demographic data, Lyme disease incidence rate, and predictor variables by health subregion (*réseaux locaux de services*, RLS) in Estrie, Quebec for this study. Values were obtained by calculating the average value of CSDs included in the dataset ($n = 116/121$).

	RLS (health subregion)								
	Pommeraie (511)	Haute-Yamaska (512)	Memphrémagog (513)	Coaticook (514)	Sherbrooke (515)	Val-Saint-François (516)	Asbestos (517)	Haut-Saint-François (518)	Granit (519)
No. CSDs	21	10	15	12	1	17	7	14	19
Total population (2016)	52,687	100,237	47,273	18,497	161,323	29,642	14,286	22,335	20,887
Population density (/km^2, 2016)	66.44	117.48	58.73	12.70	456.03	87.32	39.85	44.31	19.85
Total no. LD cases (2018)	35	44	3	0	2	1	2	1	1
LD incidence rate/ 100,000 person-years (2018) *	66.43	43.90	6.35	0	1.24	3.37	14.00	4.48	4.79
Total no. survey respondents (2018)	809	1151	822	803	3971	813	803	810	808
Self-reported tick exposure (%, 2018)	22.73	17.38	11.99	9.18	4.86	6.28	6.62	8.53	8.21
Mean predicted tick density [a]	0.30	0.02	−0.43	−0.46	−0.31	−0.44	−0.35	−0.48	−0.46
Mean habitat suitability index (2015)	0.61	0.90	0.94	0.87	0.81	0.90	0.83	0.95	0.92
Mean cumulative degree days >0 °C (2014–2018)	3464.2	3385.8	3209.8	3074.0	3236.6	3206.4	3117.0	3025.2	2840.8
Total no. eTick submissions (2018–2020)	231	57	69	5	94	14	4	9	7
Mean no. eTick submissions per CSD	11.00	5.70	4.60	0.42	94**	0.82	0.57	0.64	0.37

[a] Predicted tick density: standardised values, data obtained for 113/116 CSDs.
* Equals the total number of reported LD cases in the RLS divided by the total population of the RLS x 100,000.
** Sherbrooke health subregion includes only one CSD.
RLS = health subregion (réseaux locaux de services, $n = 9$).
CSD = census subdivision.

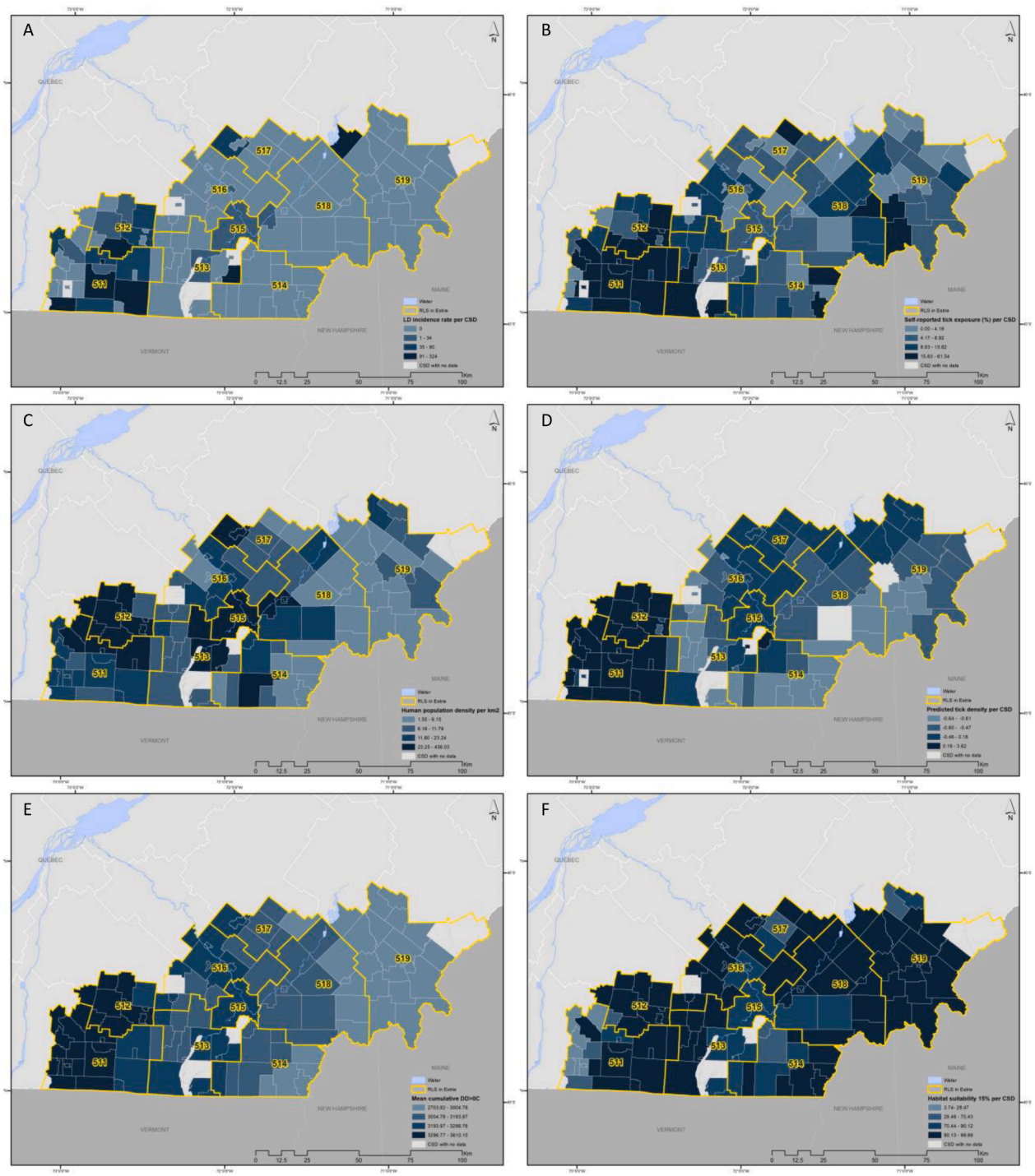

Fig. 3. Choropleth maps showing regional variation across Estrie, by census subdivision, in (A) Lyme disease incidence rate/100,000 person-years in 2018, computed using case data obtained from the public health department of Estrie and 2016 Statistics Canada census data (B) self-reported tick exposure in the previous 12 months, measured as a percentage from a 2018 populational health survey, (C) human population density/km^2, computed from 2016 census data, (D) standardised values of predicted tick density for 2018 previously modelled using active tick surveillance data from 2007 to 2017, (E) mean number of cumulative degree days above 0 °C from 2014 to 2018, (F) habitat suitability index for *Ixodes scapularis* using a 15 % threshold for deciduous tree surface cover, previously modelled using 2015 data.

Quantile breaks were used to classify the data. RLS = health subregion (*réseaux locaux de services,* $n = 9$). CSD = census subdivision (data available for 116/121 CSDs). RLS names: Pommeraie (511), Haute-Yamaska (512), Memphrémagog (513), Coaticook (514), Sherbrooke (515), Val-Saint-François (516), Asbestos (517), Haut-Saint-François (518), Granit (519).

eTick submissions came from Pommeraie than any other region between 2018 and 2020, and these submissions accounted for 47 % of all submissions from Estrie in that time ($n = 231$ out of 490 total). Sherbrooke, the most densely populated health subregion in Estrie, had the second lowest incidence of LD (1.24 per 100,000 person-years), the lowest rate of reported tick exposure (4.9 %), yet the second highest number of *I. scapularis* eTick submissions, accounting for 19 % of all Estrie submissions ($n = 94$ out of 490 total).

3.3. Spatial distribution patterns of predictor variables

Lyme disease incident cases in 2018 were mostly confined to the western health subregions of Pommeraie, Haute-Yamaska, Memphrémagog and Sherbrooke (RLS 511, 512, 513 and 515 respectively) (Fig. 3A). Most of the eastern census subdivisions of Estrie reported no LD cases, with a few exceptions. Although Pommeraie and Haute-Yamaska reported the highest tick exposure, there were also CSDs within central and eastern health subregions reporting a high prevalence of tick exposure (Fig. 3B). Predicted tick density was high across the health subregions of Pommeraie and Haute-Yamaska, moderate in the northern and eastern regions, and low in the southern regions (Fig. 3D). The number of cumulative degree days above 0 °C was visibly higher in Pommeraie and Haute-Yamaska and decreased in an eastwardly direction (Fig. 3E). The habitat suitability index for *I. scapularis* was high across most of Estrie, with noticeably lower values in seven western CSDs of Pommeraie (Fig. 3F). Although Pommeraie reported the overall highest LD incidence rate of all health subregions, six of the seven CSDs with a lower habitat suitability index corresponded with CSDs reporting lower LD incidence compared to other CSDs in Pommeraie. Larger versions of the maps from Fig. 3 are available in Supplementary material S4.

3.4. Statistical analyses

The results and interpretation of the models for LD incidence proportion (Model 1A group), and count of reported cases (Model 1B group) were similar. Therefore, we present here and in Table 4 the results using incidence proportion data and provide count data results in Supplementary Material S2, Table 1.

Self-reported tick exposure was positively and significantly associated with LD incidence ($\beta = 0.08$, 95 % CI: 0.02,0.1) (Table 4). Public health risk level 2 (compared to level 0), predicted tick density and self-reported tick exposure were also positively and significantly associated with LD incidence proportion (Table 4), but the number of eTick submissions per CSD was not. The multivariable model including 15 % habitat suitability index and cumulative degree days above 0 °C showed both factors to be positively and significantly associated with LD incidence proportion (Table 4).

In our second objective models, predicted tick density was the only significant predictor of self-reported tick exposure (model 2A group; $\beta = 0.83$, 95 % CI: 0.16,1.50, $p = 0.02$, AIC = 522.6), although the 15 % habitat suitability index approached significance in the multivariable model ($\beta = 0.68$, 95 % CI: −0.01,1.40, $p = 0.05$, AIC = 526.6). Number of eTick submissions and cumulative degree days above 0 °C were not significantly associated with self-reported tick exposure. The full results for these models can be found in Supplementary material S2: Tables 2 and 3.

As the sampling of survey respondents was proportional to the population density of the health subregion, some CSDs were represented by a small number of respondents. Of the 116 CSDs included in the study, 32 had fewer than 15 respondents. A sensitivity analysis to investigate the effect of removing these CSDs from the dataset in our primary model revealed no difference in overall conclusion, although the AIC values suggested a better fit. To provide more conservative results and interpretation, all CSDs were included in the final analyses. The results of this sensitivity analysis are presented in Supplementary material S3.

4. Discussion

4.1. Self-reported tick exposure and other predictors of regional Lyme disease incidence proportion

Our results suggest that every one percent increase in self-reported tick exposure is associated with an increase of eight percent in the regional incidence proportion of Lyme disease. While it is not appropriate to compare this strength of association with that of other risk indicators (given the very different scales), it is possible to compare relative fit of the models by examining the AIC values. The fit of the model including self-reported tick exposure was slightly lower but comparable to that of the models including predicted tick density and ecological predictors of habitat suitability, suggesting its potential utility as a surveillance tool. The best fit was found with public health risk level as a predictor variable, which is not surprising, given that public health risk levels are defined using reported human cases in addition to passive and active tick surveillance. The lowest fit was observed when eTick data was modelled as a predictor.

Other studies have also found positive associations between self-reported tick exposure and LD risk in other contexts. Self-reported human-tick encounters were associated with an increased risk of tick-borne disease at the individual and household level within LD endemic states in the United States (Hook et al., 2021). Similarly, another study found that tick bite encounters from self-reported LD patients geographically aligned with serologically confirmed LD patients and canine-positive testing for *Borrelia burgdorferi* sensu stricto in Texas (Maxwell et al., 2021). Porter et al. (2019) found that submissions of *I. scapularis* by citizen scientists at the county level were well correlated with CDC confirmed cases when compared within the state (Porter et al., 2019). Differences in study design and target populations make it

Table 4
Results of univariable and multivariable binomial regression models exploring associations between different risk indicators and Lyme disease incidence proportion at the census sub-division level.

Group 1A models: Outcome variable = Lyme disease incidence proportion							
		ß	Standard error	Confidence interval (95 %)		p-value	AIC [b]
	CSD [a] level univariable predictors						
New risk indicator	Self-reported tick exposure	0.08	0.02	0.04	0.11	<0.0001	160.4
Alternative risk indicators	Public health risk level						144.2
	Ref [e]	–	–	–	–	–	
	1	1.06	0.89	−0.62	2.74	0.216	
	2	3.55	0.73	2.11	5.00	<0.0001	
	Predicted tick density [c]	0.95	0.22	0.51	1.39	<0.0001	158.4
	No. eTick submissions	0.01	0.01	−0.01	0.02	0.411	170.1
	CSD level multivariable predictors						
Ecological risk indicators	Habitat suitability index of 15 %	3.42	1.35	0.77	6.07	0.0114	158.4
	Average cumulative degree days >0 °C [d]	1.71	0.45	0.83	2.60	0.0001	

[a] CSD: Census subdivision.
[b] AIC: Akaike information criterion.
[c] Predicted tick density: standardised values.
[d] Average cumulative degree days above 0 °C: standardised values.
[e] Reference category: Public health risk level 0.

difficult to draw direct comparisons between the results of these studies and ours. The previous studies evaluated associations between tick encounters in various demographic groups and LD risk groups, recruiting participants in a targeted manner, such as selecting households with specific property characteristics or individuals with LD experience (Hook et al., 2021; Maxwell et al., 2021). In the present study, self-reported tick exposure data was collected as part of a populational health survey of more than 10,000 people, with proportional sampling across CSDs. This method of data collection can help avoid the spatial and temporal biases often present when using reports of tick exposure (Porter et al., 2019, 2021; Zhang and Zhu, 2018) and better represents the general population. However, using this approach likely results in the inclusion of many participants who may not be aware of ticks or tick exposure, leading to a comparable decrease in the strength of association between self-reported tick exposure and LD incidence. Another consideration when comparing results is the spatial scale used for analysis. In the present study, analyses were performed at the level of the Canadian census subdivision, whereas the aforementioned studies used the US county level, representing a larger spatial area. It is reasonable to assume that there is significant movement of individuals across CSDs in day-to-day life, so that an individual may be at increased risk of tick exposure and LD infection in a different CSD to the one in which they live, receive healthcare and ultimately receive a LD diagnosis.

4.2. Considerations when evaluating predictors of regional Lyme disease incidence proportion

The fact that the association between self-reported tick exposure and LD incidence proportion in our study was not stronger can be explained by three key elements. First, the fact that approximately 40–50 % of people diagnosed with LD do not recall being bitten by a tick prior to illness (Eisen and Eisen, 2016) has most likely led to an underestimation of the true tick exposure. Furthermore, previous research suggests that the majority of reported tick exposures are adult ticks, with nymphal stage ticks more likely to be missed (CDC, 2020; Eisen and Eisen, 2021). Second, the 'gap' between self-reported tick exposure and LD incidence could be explained by varying levels of adoption of pre- and post-exposure preventive behaviors in high incidence CSD, such as seeking prophylactic antibiotics, which has been available in the region since 2017, or rapidly removing ticks after a bite. Previous research has demonstrated differences in preventive behaviors in Estrie subregions and between other regions in Quebec and Canada (Aenishaenslin et al., 2017, 2022; Bouchard et al., 2018, 2022). However, previous analysis of the same populational health survey data found that the adoption of preventive behaviours in the high-incidence health subregions of Pommeraie and Haute-Yamaska were heterogeneous and that neither global preventive behavior nor adoption of tick checks were significant predictors of LD cases or reported tick exposure at the CSD level (Bouchard et al., 2022). Finally, tick exposure was reported in many eastern CSDs where 0 cases of LD had been reported, which in itself can be explained by a lower prevalence of infection in more easterly tick populations at that time (CIUSSS de l'Estrie - CHUS, 2019), and/or several of the eastern CSDs having a low population density, resulting in a bias towards high reporting rates, although the latter should have been mitigated by using weights in the model.

This study was performed in a region of Quebec considered endemic for LD, and we cannot assume that our findings would have been the same in another region of similar, lower, or higher endemicity. We offer some suggestions for how this indicator could be improved in future studies. First, as previously noted, the use of the census subdivision may have negatively impacted the strength of association and testing a larger spatial unit may be more practical and provide different results. Second, with respect to the measurement of self-reported tick exposure, including the count of exposures in future surveys (rather than presence vs absence of exposure) will better reflect the intensity of tick exposure in the region. Furthermore, we note that tick exposure data was collected at the household level, whereas LD incidence is recorded using individual case data, which could have attenuated the association between self-reported tick exposure and LD incidence. Future surveys could determine the counts by individual within a household to harmonize the level of exposure and outcome. In addition, exposure to a tick does not imply the same level of risk as a tick bite and so these two could be differentiated. Thirdly, it is unlikely that all 10,790 survey respondents were able to correctly identify and remember a tick exposure, leading to recall and misclassification bias of the self-reported tick exposure variable. Unpublished data from eTick suggests that 4 %, 8.1 % and 10.6 % of submissions from Quebec in the years 2018, 2019 and 2020, respectively, were not ticks (personal communications, Jade Savage, 2023). Future studies could take into account potential misclassification bias, either by testing the respondents' ability to identify a tick in the questionnaire, or through mathematical or statistical methods that deal with the bias (Althubaiti, 2016; Greenland, 2005; MacLehose et al., 2009). Finally, there is potential for misclassification bias of LD diagnoses, likely resulting in an underestimation of LD incidence. In Canada, an estimated one third of cases are reported in regions of LD emergence (Ogden et al., 2019). This misclassification bias is likely to be differential and related to regional LD incidence, making it difficult to estimate the impact on associations found in this study.

In our study, the weaker strength of association and lesser model fit between LD incidence and eTick submissions again likely reflects that people are not aware of all tick exposures. Another contributing factor is the varying activity of eTick users across CSDs during the period of data collection; several CSDs did not have any eTick submissions, others had only a few, yet two CSDs had > 90 submissions due to a few regular contributors. This variation was likely due eTick being relatively novel at the time, and this association should be tested again in future studies. There are now several tick-related citizen science projects, such as TickSpotters through TickEncounters in the United States (https://web.uri.edu/tickencounter/) and Tekenradar in the Netherlands (https://www.tekenradar.nl/home). The benefits and limitations of citizen science methodologies have been described extensively elsewhere and we will not attempt to compare existing citizen science platforms with eTick given the novelty of the platform at the time of data collection (Eisen and Eisen, 2021; Hines and Sibbald, 2015; Koffi et al., 2017; Kopsco et al., 2020; Lewis et al., 2018; Nieto et al., 2018).

4.3. Ecological predictors of regional Lyme disease incidence proportion

Both ecological predictors evaluated in our multivariate model were significantly and positively associated with LD incidence. Several studies have demonstrated how the presence and amount of forest may predict LD cases at both the household and regional level (Glass et al., 1995; Kitron and Kazmierczak, 1997) as reviewed by Killilea et al. (2008). As demonstrated in Fig. 3, the lower habitat suitability index in the western CSDs of Pommeraie (RLS 511) corresponds with a lower LD incidence, moderate reported tick exposure, yet high predicted tick density. This may reflect the concept that regions with more fragmented forest habitats can lead to greater entomological risk, yet lower incidence in humans, possibly due to decreased human activities in these habitats (Fischhoff et al., 2019). While the association between climate and tick density/activity is well established (Burtis et al., 2016; Eisen et al., 2016), the association between cumulative degree days above 0 °C and human LD incidence has been explored to a lesser degree. The results of our model indicate that temperature is strongly associated with LD incidence proportion, aligning with previous research (Robinson et al., 2015), and our maps show how the decrease in LD incidence rate from west to east mirrors (albeit less smoothly) the decrease in average number of cumulative degree days above 0 °C.

4.4. Predictors of self-reported tick exposure

From our group 2 models, only predicted tick density was

significantly associated with self-reported tick exposure, with the 15 % habitat suitability index approaching significance. Contrary to its association with LD incidence, we did not find a significant association between average number of cumulative degree days above 0 °C and reported tick exposure. Our maps support this result by demonstrating how tick exposure was reported across several CSDs, whereas LD incidence rate was generally limited to the warmer CSDs in the west of Estrie. Furthermore, while LD cases were reported by probable CSD of acquisition, self-reported tick exposure was not, which may contribute to this difference.

4.5. Practical utility of self-reported tick exposure as a risk indicator

It is beyond the scope of this paper to discuss active and passive surveillance methods which may be more or less useful in terms of aiding risk assessment, and they have been described extensively (Bouchard et al., 2022; Brooks et al., 2022; Holcomb et al., 2023; Ripoche et al., 2018). The potential utility of self-reported tick exposure as an indicator of LD risk, measured through a populational survey, can complement existing surveillance methods in three important ways. First, the survey design with random sampling methodology can differentiate between no risk and absence of information, offering a more representative understanding of the risk distribution in a region than passive surveillance methods. Second, it offers an opportunity to estimate the rate of human-tick encounters, which may better reflect the true level of LD risk compared to ecological indicators, thereby helping to inform control interventions (Eisen and Eisen, 2021; Fischhoff et al., 2019). Indeed, including this indicator in future studies may help us to understand how other ecological indicators are associated with tick exposure, TBD cases and entomological risk, and differences between expected and observed tick exposure. Finally, while surveys can be time-consuming and costly to design and implement, the addition of questions related to tick exposure to planned population health surveys does not require a lot of resources. This approach may represent a low-cost option for evaluating LD risk, keeping in mind that the value of measuring self-reported tick exposure will likely be context-specific and dependant on factors such as pre-existing surveillance measures, resources, and level of endemicity.

4.6. Limitations of this study

This study brought together several datasets aggregated by region and may therefore be subject to the potential biases and pitfalls of such ecological studies. Furthermore, the use of a cross-sectional survey to measure self-reported tick exposure means that temporality of events cannot be established, and the individuals who responded may not have accurately reflected the population in terms of motivation, health status and socio-demographics (Jang and Vorderstrasse, 2019; Keyes et al., 2018; Lallukka et al., 2020). There are two main considerations specific to the data sources which should be taken into account when interpreting these results. First, as previously described, it is possible that respondents encountered ticks in one or more CSDs and not necessarily in the CSD in which they resided or acquired LD. It is unknown whether the large sample size in this survey was enough to overpower this spatial uncertainty. Second, although most of the data sources originate from 2018 or a period leading up to 2018, there were two exceptions: eTick data from April 2018 to July 2020 was included to ensure representation for each CSD, and the 15 % habitat suitability index for *I. scapularis* was derived from 2015 data. The latter data source was deemed to be less critical to the interpretation of results due to the low variance across the Estrie region.

5. Conclusion

This proof-of-concept study provides a first look at how population survey data measuring self-reported tick exposure could be used to evaluate risk for LD at the regional level in an endemic region of Quebec. While self-reported tick exposure demonstrated a positive association with LD incidence proportion and a comparable fit to predicted tick density and recognised ecological variables, we highlight some important considerations and offer suggestions as to how this indicator may be improved in future studies.

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Authors' contributions

CA, CB and NB designed the study. CA, CB, GB and FM participated in the development of the study questionnaire. JS and OT provided eTick data and habitat suitability index data, respectively. MSC, NB and CB performed statistical analyses. CA, CB and NB performed analyses and initial interpretation. GB, HC, PC, PL, FM, LR, JS and OT provided further interpretation. NB wrote the first version of the manuscript, with major contributions from CA and CB. All authors reviewed and approved the final manuscript.

Ethical approval and consent to participate

All methods related to the CIUSSS de l'Éstrie-CHUS 2018 survey were carried out in accordance with Helsinki Declaration. Oral consent was obtained from all study participants because data collection was performed using phone interviews. The study protocol, as well as the procedure for obtaining oral consent, was approved by the Comité d'éthique de la recherche du CIUSS de l'Estrie – CHUS (project #2018-2612).

Declaration of Competing Interest

The authors declare they have nothing to disclose.

Data availability

The data that has been used is confidential.

Acknowledgments and data sharing

We thank *la Direction de santé publique de l'Estrie* for providing access to Lyme disease case data and results of the 2018 general populational health survey, Olivia Tardy et al. for permitting us to utilize the habitat suitability index data, Bouchard et al. for providing tick density predictions, and eTick for providing access to data and all the volunteer participants who gathered data. We also thank the Canadian Lyme Disease Research Network, which has supported NB through her PhD.

The various data that support the findings of this study were used with the permission of the respective authors and organisations. Restrictions may apply to the availability of some of these data.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.ttbdis.2023.102271.

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