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ORIGINAL ARTICLE

Prevalence of Autoimmune Inflammatory Myopathy in the First Nations Population of Alberta, Canada

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Objective. **To estimate the population-based prevalence of autoimmune inflammatory myopathy (AIM) in Alberta, Canada, with a specific focus on rates in the First Nations population.**

Methods. **Physician billing claims and hospitalization data for the province of Alberta (1994–2007) were used to estimate the probability of having AIM (i.e., polymyositis or dermatomyositis) based on 3 case definitions. A latent class Bayesian hierarchical regression model was employed to account for the imperfect sensitivity and specificity of billing and hospitalization data in case ascertainment. We accounted for demographic factors of sex, age group, and location of residence (urban or rural) in estimating the prevalence rates within the First Nations and non–First Nations populations.**

Results. **The overall prevalence of AIM was 25.0 per 100,000 persons (95% credible interval [95% CrI] 13.4–49.0) in the First Nations population and 33.8 (95% CrI 28.9–39.6) in the non–First Nations population. For both groups, prevalence was increased in women relative to men, rural women relative to urban women, and in those age >45 years.**

Conclusion. **Unlike other rheumatic diseases such as rheumatoid arthritis, systemic lupus erythematosus, and systemic sclerosis, we did not detect an increased prevalence of AIM in Alberta's First Nations population relative to the non–First Nations population. Potential limitations include coding errors, underidentification of First Nations members, and recognized differences in access to care for the First Nations population.**

INTRODUCTION

Polymyositis (PM) and dermatomyositis (DM; currently referred to as autoimmune inflammatory myopathy [AIM]) are autoimmune conditions characterized by inflammation of proximal muscles or their vascular supply, in association with systemic symptoms and sometimes other organ manifestations (rash, arthritis, and lung disease) (1). AIM is often considered one of the "systemic autoimmune rheumatic diseases" (SARDs), which also include systemic lupus erythematosus (SLE), systemic sclerosis (SSc; scleroderma), and Sjögren's syndrome.

An increased prevalence of SARDs (2,3) and inflammatory arthritis (4) has been reported in North America's First Nations population relative to non–First Nations populations. We have recently identified an increased prevalence of SLE and SSc in the First Nations population of Alberta, Canada (5), which includes multiple tribal ancestries such as Blackfoot, Chipewyan, Cree, Dene, Saulteaux, and Sioux Nations. The prevalence of AIM in North America's First Nations population is largely unexplored (3); thus,

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Significance & Innovations

- This is the first population-based estimate of autoimmune inflammatory myopathy (AIM) in the Native North American population.
- Unlike other rheumatic diseases such as rheumatoid arthritis, lupus, and systemic sclerosis, we did not identify an increased prevalence of AIM in this population.
- AIM prevalence was increased in females, rural residents, and individuals age >45 years.

our objective was to use provincial population-based administrative data, which contain an individual's diagnosis based on physician billing claims and hospitalization data, to estimate and compare the prevalence of AIM in Alberta, Canada.

Of importance in using administrative data for this purpose are the systemic differences in access to health care for First Nations patients (6–9). We have previously demonstrated that patient age, sex, socioeconomic status, level of education, and location of residence affect prevalence estimates obtained from administrative data (5,10–13). In most prior prevalence estimates, the effects of race, specifically First Nations status, have not been considered. The effect of demographic factors on disease diagnosis and thus estimated prevalence may be particularly amplified in the First Nations population, and analyses considering not just First Nations status, but also age, sex, and location of residence (urban/rural), are warranted.

MATERIALS AND METHODS

We have previously described our methods in detail (5). To summarize, Alberta Health and Wellness maintains administrative databases for the Alberta Health Care Insurance Plan (AHCIP). All Alberta residents, including First Nations Albertans, receive health care through AHCIP, a universal publicly funded system. This includes physician billing claims and hospitalizations, covering approximately 3.7 million Alberta residents. The number of individuals registered with AHCIP only differs from census data by 0.1% (14). Approximately 6% of the Alberta population is of Aboriginal ancestry (First Nations, Inuit, and Métis) by self-identified census data (15), with 3.7% of the 2007 Alberta population being identified as First Nations according to AHCIP (described below).

We ascertained cases of AIM over a 14-year time period (1994–2007). The prevalence estimate was based on all cases ascertained who were registered with the AHCIP at any time during the year 2007, with the denominator being the midyear provincial population registered with the AHCIP in that year.

Case ascertainment was based on the probability of having a diagnosis of AIM (PM or DM) using physician billing claims coded according to the Ninth International Classification of Diseases, Clinical Modification (ICD-9-CM) system (code 710.4 for PM and code 710.3 for DM), or hospitalization data (16 discharge diagnoses fields with ICD-9-CM prior to April 2002 and 25 discharge diagnoses fields with the International Classification of Diseases, Tenth Revision, Canadian Adaptation, code M33, after April 2002). We used 3 case definitions to estimate the probability of an individual having AIM, based on ≥1 billing code by a rheumatologist, ≥2 billing codes by any physician (≥8 weeks apart but within 2 years), or 1 hospitalization discharge diagnosis.

We used the Alberta Health and Wellness AHCIP file to determine patient sex, age (dichotomized as less than or greater than age 45 years), location of residence by postal code (rural or urban defined on census metropolitan area classifications) (16), and First Nations status using validated methodology developed by Alberta Health and Wellness for health services research and adopted by the Alberta research community (8,17–19). This methodology uses the provincial health premium payment history to identify individuals whose premiums were paid by the First Nations and Inuit Health Branch (Health Canada) at any time point since 1994. All others were classified as non–First Nations.

Our estimates were derived using a Bayesian latent class hierarchical regression model (11,13,20). This modeling approach accounts for the imperfect sensitivity and specificity of billing and hospitalization data in case ascertainment, as well as the influence of demographic factors on prevalence. Latent class models are applicable in instances where there is no readily available gold standard for determining cases, such as in administrative data sets, as they incorporate information from multiple case definitions, each of which provides information on a patient's probability of having the disease in question. Our model accounted for possible correlation between the case definitions based on physician billing claims (rheumatologist billing and any physician billing), as these are derived from the same source. The sum of the individual-level probabilities across all subjects provides the numerator for the prevalence point estimate. Bayesian methods produce 95% credible intervals (95% CrIs), representing values between which there is a 95% probability of containing the parameter of interest, given the prior information and the data at hand. Throughout our analyses, noninformative prior distributions were used, except for the specificity, which is known to be very high. We therefore used a beta (248.3, 1.65) prior density for the specificity, which covers the range from 0.98 to 1.0. These informative prior densities were based on the results generated in our previous analyses (5). Statistical analyses were programmed in WinBUGS, version 1.4.3 (MRC Biostatistics Unit). Ethics approval was provided by the McGill University Institutional Ethics Review Board.

RESULTS

Using a latent class Bayesian hierarchical regression model, the overall prevalence of AIM in Alberta's First Nations population was estimated to be 25.0 cases per 100,000 persons (95% CrI 13.4–49.0), compared to the

Table 1. Autoimmune inflammatory myopathy prevalence estimates for Alberta First Nations and non–First Nations populations, per 100,000 people*

	Females		Males	
	First Nations	Non–First Nations	First Nations	Non–First Nations
Age <45 years				
Rural dwellers	27.7 (10.0–67.2)	19.2 (13.3–27.1)	2.1 (0.1–13.4)	5.7 (3.0–9.6)
Urban dwellers	4.5 (0.1–35.8)	13.4 (9.6–18.7)	4.8 (0.5–23.7)	4.2 (2.4–7.2)
Age >45 years				
Rural dwellers	137.3 (55.9–345.6)	124.5 (97.5–161.9)	12.7 (1.1–54.7)	58.7 (43.5–78.9)
Urban dwellers	87.3 (4.8–415.9)	86.7 (70.6–107.6)	17.4 (0.2–153.9)	37.6 (28.5–49.5)

* Values are the prevalence estimate (95% credible interval).

estimated prevalence of 33.8 cases per 100,000 persons in the non–First Nations population (95% CrI 28.9–39.6). Consistent with other SARDs, women were more frequently affected, with 42.6 cases per 100,000 First Nations females (95% CrI 21.8–87.7) and 47.7 cases per 100,000 non–First Nations females (95% CrI 40.2–57.1), versus 6.5 cases per 100,000 First Nations males (95% CrI 1.6–18.9) and 19.6 cases per 100,000 non–First Nations males (95% CrI 15.5–24.9).

Except for rural First Nations women, all prevalence point estimates were higher in the non–First Nations population, although the credible intervals were wide and overlapping (Table 1). Therefore, we cannot rule out even rather large differences in prevalence rates comparing the 2 groups. Also of note is the higher prevalence of AIM in rural women of both ethnicities.

DISCUSSION

We provide contemporary population-based estimates of AIM for Alberta, Canada. Our prevalence estimates are consistent with those reported from the province of Quebec using the same methodology, with a 2003 provincial prevalence of PM and DM of 21.5 per 100,000 persons (95% CrI 19.4–23.9) (11). Our estimate is also consistent with population-based data from Rochester, Minnesota, which identified an age- and sex-adjusted prevalence of DM of 21.42 per 100,000 persons (95% confidence interval [95% CI] 13.07–29.77) (21), and of PM of 3.45 per 100,000 persons (95% CI 0.00–7.35) (22). Our estimates may be slightly higher, given that Alberta physician billing claims allow up to 3 diagnostic codes, which may increase the likelihood of identifying a chronic disease with administrative data. In contrast to the Quebec estimates, we identified the highest prevalence of AIM in older rural females, but not older urban females. The reason for this discrepancy is unknown, but it is possible that there are differences between provinces in how well our operational definition (based on postal codes) captures rural versus urban residence. Differences in environmental exposures between provinces and/or rural versus urban dwellers may also explain this (23). However, the finding that AIM predominantly affects older rural-dwelling females may have implications for service delivery, as AIM patients require longitudinal observation by specialists in rheumatology or neurology, who are typically located in urban centers.

The similar prevalence rate for AIM in the First Nations population compared to the non–First Nations population was an unexpected finding, given higher prevalence estimates for other SARDs in the First Nations populations (2,3,5). One explanation for this possible finding is using administrative data coding for case ascertainment without a clinical "gold standard," as no AIM cohorts exist in our province and permission to access medical charts is not easily granted. Thus, our estimates are based on the assigned physician billing and/or hospital discharge diagnoses. Our modeling approach combined the results of 3 case definitions to provide the best estimate of prevalence based on the available data, but there is still a potential for imperfect adjustment.

Our methodology to determine First Nations status using health care premium payer information is consistent with that used for disease surveillance and health services research in our province for other conditions, such as chronic kidney disease (8) and multiple sclerosis (24). Our estimates thus are generalizable to the population of First Nations individuals with official Treaty Status as defined by the Indian Act of Canada (25); those who are non–Treaty Status or of Métis ethnicity have been classified as non–First Nations.

In AIM, we expect that the disease expression is sufficiently severe to require medical care, but it is possible that providers unfamiliar with AIM might assign a diagnostic code for a more commonly occurring SARD, such as SLE. These cases might not be verified by a specialist for First Nations patients, as systematic differences in health care utilization for First Nations patients are recognized (6–9). This would result in an underestimation of AIM cases in the First Nations cohort. As well, our estimates only consider those residents of Alberta who use the health care system; however, previous work using the same data sources shows higher per capita physician claims, emergency room visits, and hospital stays for First Nations compared to non–First Nations residents, including for musculoskeletal conditions (18). Our current analysis was based on our prior experience with SLE and SSc where the same data sets, identification techniques, and analysis methods were used (5). In those analyses, a higher prevalence of SLE and SSc was demonstrated in First Nations subjects. Thus, it seems unlikely that underutilization of health care by First Nations people would explain our findings, and we cannot entirely explain why a

true increase in AIM among First Nations people (if it existed) would not be discovered by the same methods.

To date, there are almost no published studies of AIM in North America's First Nations populations (3); this study represents the first large population-based study. Boyer et al described rheumatic disease in the southeast coastal Indians of Alaska, finding 2 cases of physician-diagnosed PM in a population of 9,810 (26). The Nuu-Chah-Nulth population had 6 cases of overlap syndrome involving PM in a population of 2,300 (27). These studies suggest a relatively high occurrence in those populations; however, no reports of PM or DM occurred in the Inuit (28) or Inupiat Eskimos (29). The only other description of AIM in indigenous populations was a comparative study of the clinical phenotype and genotypes between Mesoamerican Mestizos and North American Caucasians, which found that Mesoamerican Mestizos more frequently had severe muscle involvement (with more distal weakness, muscle atrophy, and falls) and skin involvement, but with less inflammatory arthritis and interstitial lung disease (30).

Indigenous Australians and New Zealanders did not have an increased prevalence of AIM; no cases of PM or DM were found during a World Health Organization/International League of Associations for Rheumatology Community Oriented Program for the Control of Rheumatic Diseases study of rheumatic disease in Australian Aborigines (31). A hospital admission– and death certificate–based estimate of systemic connective tissue diseases in New Zealand in the 1960s identified 2 cases of DM in a Maori population of 201,000 as compared to 83 cases in 2,476,000 Europeans (32). Both of these populations have been found to have higher than expected rates of other SARDs (33–35) similar to North American indigenous people. Thus, our results mirror those seen in other indigenous populations of the Pacific Rim, and our study does highlight the need to study each SARD separately, rather than assuming that there is an excess burden of all SARDs in First Nations people.

In summary, we did not demonstrate any differences in estimates of the prevalence of AIM between First Nations and non–First Nations populations in Alberta, although we cannot rule out higher or even lower rates. Older women living in a rural location had the highest prevalence of disease regardless of ethnicity.

AUTHOR CONTRIBUTIONS

All authors were involved in drafting the article or revising it critically for important intellectual content, and all authors approved the final version to be submitted for publication. Dr. Barnabe had full access to all of the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis.

Study conception and design. Barnabe, Joseph, Svenson, Bernatsky.

Acquisition of data. Barnabe, Fritzler, Svenson, Bernatsky.

Analysis and interpretation of data. Barnabe, Joseph, Bélisle, Labreque, Barr, Svenson, Peschken, Hemmelgarn, Bernatsky.

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