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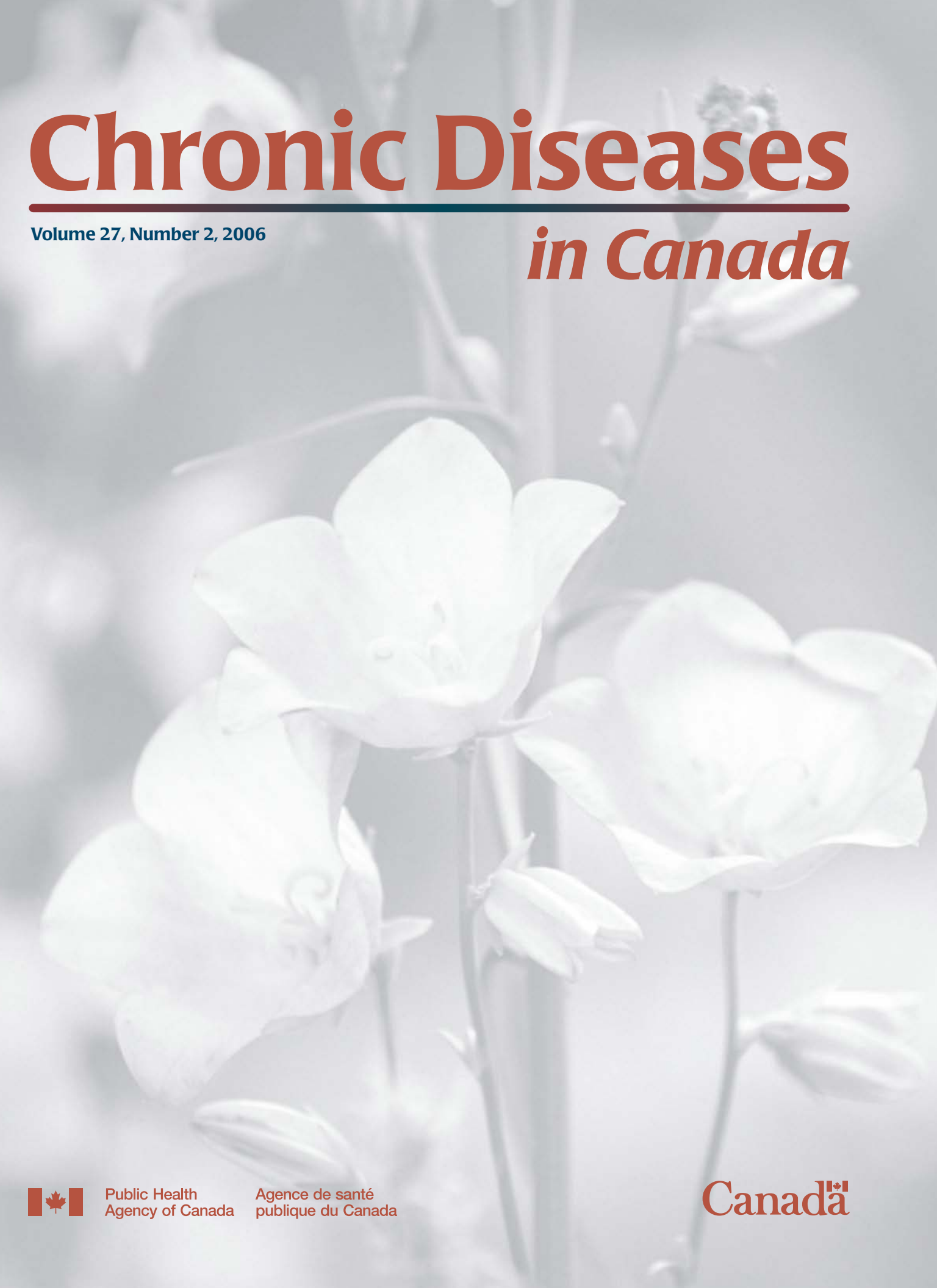

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Farewell to Dr. Steve Hotz

On behalf of all those associated with Chronic Diseases in Canada over the past several years, we would like to express our appreciation for the significant contribution of Dr. Steve Hotz to the journal. Dr. Hotz has been one of our associate scientific editors, providing expertise on manuscripts that have a behavioural sciences thrust. Dr. Hotz has decided to step down from his associate scientific editor post to devote more time to his research, teaching activities and professional practice. We are very grateful to have benefited from our colleague's integrity, exacting standards and extensive knowledge. We are sorry to see him leave.

The Editors

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Population-based cohort development in Alberta, Canada: A feasibility study

Heather Bryant, Paula J Robson, Ruth Ullman, Christine Friedenreich and Ursula Dawe

Abstract

In a climate of increasing privacy concerns, the feasibility of establishing new cohorts to examine chronic disease etiology has been debated. Our primary aim was to ascertain the feasibility of enrolling a geographically dispersed, population-based cohort in Alberta. We also examined whether enrolees would grant access to provincial health care utilization data and consider providing blood for future analysis. Using random digit dialling, 22,652 men and women aged 35 to 69 years, without diagnosed cancer, were recruited. Of these, 52.4 percent (N = 11,865) enrolled; 84 percent of Alberta communities were represented. Approximately 97 percent of enrolees consented to linkage with health care data, and 91 percent indicated willingness to consider future blood sampling. Comparisons between the cohort and the Canadian Community Health Survey (Cycle 1.1) for Alberta demonstrated similarities in marital status and income. However, the cohort had a smaller proportion who had not finished high school, a greater proportion of non-smokers and a higher prevalence of obesity. These findings indicate that establishment of a geographically dispersed cohort is feasible in the Canadian context, and that data linkage and biomarker studies may be viable.

Key words: *Alberta, cohort studies, feasibility studies, questionnaires*

Introduction

Prospective cohort studies are potentially powerful tools to examine chronic disease etiology. Because they collect exposure information prior to disease diagnosis, they are free from the potential differential biases that may occur recalling this information when cases are compared with controls. Further, because the exposure information is collected relatively contemporaneously, rather than having subjects recall distant past exposures, there is potential for increased accuracy of reporting. The high value of these research resources has resulted in the construction of a number of cohorts internationally. It has also resulted in national discussion of the potential for development of a large Canadian cohort for the study of chronic disease.

While the value of such cohorts is acknowledged, their drawbacks are equally well known. Ongoing cost, of course, is one issue that would have to be addressed in the consideration of developing a full-scale national cohort. In addition, although the cohort design minimizes recall bias, other biases are now known to exist: dietary assessment is hampered to some degree by mis-reporting,[1] and there is controversy concerning the impacts of socially desirable responding on the validity of data obtained from self-administered questionnaires.[2-5] Similarly, loss to follow-up over time may also contribute to bias.[6] Moreover, quantifying the precise impact of such biases in cohort studies remains challenging. Furthermore, because cohort studies need a large sample size and extensive exposure data collection to have sufficient study power, their construction and maintenance is expensive. For etiological hypotheses, investigators must wait for a sufficient number of cases to be identified before analysis is worthwhile, delaying results and adding to the expense. This time delay of results makes cohort construction an unattractive research endeavour for investigators who live under the "publish or perish" paradigm.

In 1999, the population health research group at the Alberta Cancer Board (ACB) began to explore the feasibility of the construction of such a cohort in Canada. There were two underlying themes in the development of the cohort concept: the creation of a research legacy, identified by our team as a population health laboratory, and the maximization of both short-term and long-term potential benefits.

The research legacy term refers to the concept of developing a rich data resource that could be used by current researchers, but which would be of even more value to researchers who may enter the field in several years, when the cohort is "maturing" and disease outcomes become frequent. This resource would be more valuable with increased depth and volume of behavioural, biochemical, socio-demographic and environmental data available on each participant. Thus, collection of exposure information with detailed, validated tools, re-collection of data at reasonable time intervals, individual biological specimens and the potential to link data with complete health care utilization files were seen to be key components of such a population laboratory. While it was envisioned that the primary focus would be research into cancer etiology, many of the risk factors examined are also potentially important in the etiology of other chronic diseases, thereby ensuring that the cohort would also be valuable for research in other areas.

Author References

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The long-term benefit of the cohort would be linked most closely to the complete and accurate collection of the information noted above. However, we discussed several ways in which shorter term outcomes could also be of value. Clearly, a cohort that was representative of the general population, rather than one composed of population sub-groups defined by occupation or educational status, or people recruited as volunteers, could provide more insight into general trends in cancer-related prevention and screening behaviours. While we were interested in examining the degree to which a population-based cohort could be constructed, we projected that, even if the cohort was somewhat unrepresentative with respect to demographic or behavioural characteristics, within-cohort comparisons of predictors of behaviours or behaviour change could still produce valuable insights into cancer control. In addition, such a cohort could provide an opportunity for the evaluation of "natural policy experiments" that might occur over its longitudinal course. If, for example, smoke-free public spaces were mandated in some communities and not others over the course of the study, a baseline group would already be in place with recorded smoking behaviours and other characteristics prior to the policy change. By using the rich data available to distinguish the characteristics of smokers who changed their behaviours from those who did not, we could provide excellent analysis of the predictors for success of such policies.

In this paper, we address three questions that relate to the legacy potential and the probability of collecting reasonably geographically representative data from "average" individuals. These questions are,

1. Could we enrol a cohort of randomly selected individuals across a dispersed geographic population which could adequately represent the distribution of demographics and health behaviours within a province?
2. What proportion of these individuals, in a world of increasing privacy concerns, would be prepared to give access to health care utilization files for further research?
3. What proportion of these individuals would be willing to consider providing a blood sample for storage and future analysis?

This paper reports on the findings related to these questions.

Methods

The target population for the feasibility study was men and women aged 35 to 69 years. Other enrolment criteria were as follows: 1) no known history of cancer other than non-melanoma skin cancer; 2) plans to reside in Alberta for at least one year; and 3) English speaking, to allow for collection of self-report data. Approvals to conduct the feasibility studies were obtained from the ACB and University of Calgary ethics review boards.

Subject selection and enrolment

A two-stage sampling design was used to identify eligible individuals. The first stage used a random digit dial (RDD) procedure.[7] Since 97 percent of Alberta households in the year 2000 had at least one telephone line,[8] a telephone-based sampling method ensured that almost all households were included in the theoretical sampling frame. The first stage of sampling selected households in the 17 regional health authorities (RHAs) extant in Alberta in 2000, and the second stage selected one eligible adult within each household. A household was defined as one or more persons, related or otherwise, who occupy the same private or collective dwelling.[9] The sampling and RDD were done by an experienced social research laboratory at the University of Alberta.[10]

The recruitment for the feasibility component was done in four waves, in order to evaluate and, if necessary, change procedures as a result of early experience in the study. These four stages are referred to as RDD1 through RDD4, respectively.

First-stage random selection

Standardized procedures were used to ensure methodological and ethical integrity of the RDD approach.[7] An electronic database of randomly generated telephone numbers, mapped to RHAs, was used for calling purposes. Trained interviewers, working with a computer assisted telephone interviewing (CATI) system and standard script, called selected households to screen for eligible individuals who would be willing to consider enrolment into the Alberta Cohort Study (The Tomorrow Project®).

In order to maximize the likelihood of contacting residents in the selected households, calls for RDD1 were made up to 20 times over a variety of times and days of the week before abandoning the number. Because of diminishing returns with subsequent calls, this total was reduced to 15 in RDD2 and to 12 calls in each of RDDs 3 and 4. Disproportionate sampling was done to ensure a sufficient number of participants from rural and remote regions.

Second-stage random selection

The first adult householder answering the telephone was given a description of the study purpose, eligibility criteria, conditions for participation (i.e., voluntary; long term with periodic follow-ups and repeated data requests), and examples of information asked on baseline questionnaires. In households with more than one potential study participant, the person with the most recent birthday was selected for possible enrolment, to reduce selection bias towards groups more likely to be available to answer the telephone.[7]

As part of our feasibility exploration, a second household member of the opposite sex was selected for possible enrolment when the first respondent was eligible and interested in considering cohort enrolment. This approach was attempted in 2527 households (RDD1) to assess the

impact on rate of accrual.

At the conclusion of the RDD process, all telephone numbers/households were assigned one of the following codes: "recruited" (target respondent was eligible and interested in considering study enrolment); "ineligible"; "undetermined eligibility" (efforts at contacting the target householder were unsuccessful and/or a screening interview was not completed); or "refused".

Subject enrolment and retention

A self-administered baseline health and lifestyle questionnaire (HLQ) and a detailed consent form were sent by regular mail to individuals interested in study enrolment. Participants were classified as enrolled if they completed and returned the HLQ and the consent form. Approximately three months after enrolment into the study, two additional questionnaires concerning habitual diet and past year physical activity were mailed to participants.

As part of their written consent, participants were asked for permission for data linkage with the Alberta Cancer Registry. They were also asked to voluntarily provide their Alberta Personal Health Number (PHN) and signed authorization allowing the Alberta Cohort Study to request health services utilization data held by Alberta Health and Wellness. Specifically, subjects were informed that the study would seek data from Alberta Health concerning types of health care services accessed (defined by "billing codes"), frequency of use of such services and whether services were provided in doctors' offices or hospitals. Subjects were invited to consent to periodic linkages for the duration of their participation in the study. Individuals were allowed to participate in the overall study even if they denied access to Alberta Health or to Alberta Cancer Registry data.

Participants were also asked if they would be willing to consider providing a blood sample for study purposes, should they be asked for one in the future. They were also informed that, if such a request were made, a full explanation of the blood collection's purpose would be provided and that further written consent would be required before any sample could be collected.

The final page of the consent form provided subjects with the study's contact details, and encouraged subjects to use any of several contact methods if they moved away from the address from which they were originally recruited. Specifically, we provided a "change of address" form on the study Web site (www.thetomorrowproject.org), as well as toll-free and collect-call telephone numbers, in order to ensure that subjects who moved out of province or out of Canada had access to a variety of free and convenient methods of keeping in contact. In addition, subjects were asked to provide their cellular telephone number and e-mail address (if applicable), as well as contact details for two people outside their household. These contacts would be used in the event that the subject could not be contacted using any other means. Furthermore, regular contact continues to be maintained with subjects by means of a biannual newsletter, which also serves to provide feedback on study progress and news.

Baseline data collection

Baseline information about lifestyle-related risk factors and exposures was collected using three self-administered, mailed questionnaires. The instruments were selected on the basis of 1) relevance to factors with potential high attributable risk for cancer and other chronic diseases; 2) the suitability/adaptability of the measure for self-administered surveys; and one of the following: 1) previous use in established epidemiologic studies; and/or 2) published data describing the measure's psychometric properties.

Health and Lifestyle Questionnaire (HLQ)

The HLQ is a composite of existing items used in other large studies relating to

TABLE 1
Number of people recruited and enrolled into the Alberta Cohort Study, described by the regional health authority (RHA) of residence at time of recruitment

	Alberta RHA[a]	Number of people recruited[b]	Number of participants enrolled[c]			Response[d]
			Male	Female	Total	%
1	Chinook	1,628	360	554	914	56.1
2	Palliser	1,491	376	471	847	56.8
3	Headwaters	1,279	277	411	688	53.8
4	Calgary	3,729	822	1,193	2,015	54.0
5	RHA 5	1,111	252	346	598	53.8
6	David Thompson	1,255	317	419	736	58.6
7	East Central	1,369	302	403	705	51.5
8	Westview	1,559	316	489	805	51.6
9	Crossroads	732	157	217	374	51.1
10	Capital	4,036	807	1,138	1,945	48.2
11	Aspen	1,089	237	332	569	52.2
12	Lakeland	1,434	291	419	710	49.5
13	Mistahia	1,029	218	338	556	54.0
14–17	Northern Regions	911	175	226	401	44.0
Total		**22,652**	**4,907**	**6,956**	**11,863[e]**	**52.4**

[a] RHAs extant in Alberta in 2000.

[b] Defined as eligible and interested in receiving enrolment package (consent form and Health & Lifestyle Questionnaire).

[c] Defined as completed and returned Health & Lifestyle Questionnaire.

[d] Calculated as number enrolled/number recruited (%).

[e] Excludes two transgender individuals.

personal health and reproductive history, family history, psychosocial factors, anthropometric measures, use of cancer screening services, smoking behaviour, sun exposure and socio-demographic characteristics. Some items were developed for the Alberta Cohort Study if other sources were not available.

Items concerning personal health history, male and female reproductive information, and family history of chronic illness and longevity were adapted from questions used in the Prostate, Lung, Colorectal and Ovarian (PLCO) Screening Trial,[11] the Women's Health Initiative (WHI) Study[12] and the 2000/01 Canadian Community Health Survey (CCHS; cycle 1.1).[13]

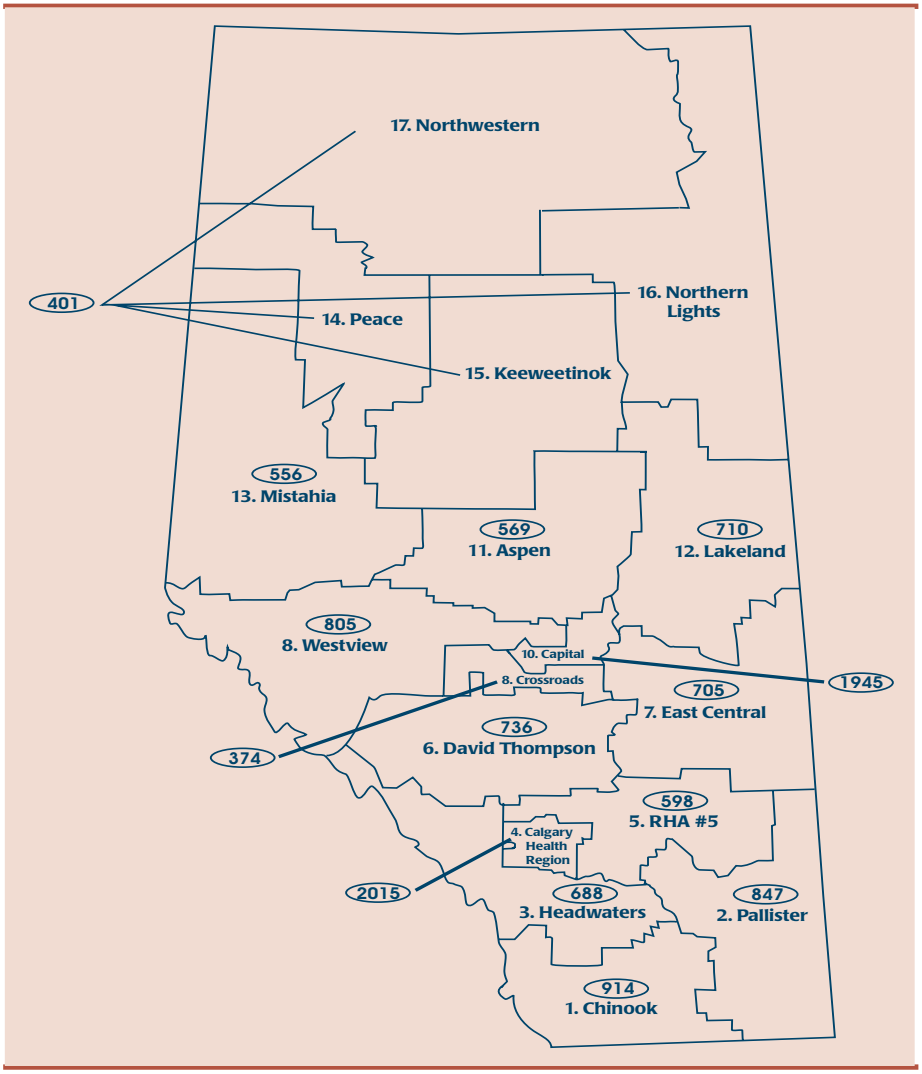
Items concerning Pap tests, mammograms, clinical breast examination, breast self-examination and PSA tests originated with the CCHS.[13] Items about colorectal screening with digital rectal examination, sigmoidoscopy/colonoscopy and stool collection for occult blood testing were adapted from the CCHS and the California Health Interview Survey 2001.[14]

Questions about tobacco exposure were based on a recommended set of measures for monitoring tobacco use in Canada as developed through the Canadian Workshop on Data for Monitoring Tobacco Use.[15] Sun exposure was measured using selected items recommended by the Canadian National Workshop on Measurement of Sun-Related Behaviours for monitoring sun exposure and protective behaviours.[16]

Social support was measured using questions from the Medical Outcomes Study (MOS)[17] questionnaire. Items proposed for the CCHS 2000/2001 were used for measuring stress. Spirituality was measured with three items taken from the CCHS and one created for the baseline survey.

Subjects were also provided with detailed instructions and a 183 cm (72 inch) tape measure for obtaining accurate height, buttock and waist measures using a self-administered method that had been tested for reliability and validity.[18] Instructions were also given for recording body weight

FIGURE 1
Numbers of participants enrolled in the Alberta Cohort Study from the 17 regional health authorities (RHAs) extant in the Province of Alberta in 2000



using a scale accessible to the respondent. The HLQ comprised 32 pages and took an estimated 40 minutes to complete.

Diet History Questionnaire (DHQ)

The DHQ is a cognitive-based food frequency questionnaire (FFQ) developed by the US National Cancer Institute (NCI) as a tool for assessing diet over the preceding 12-month interval.[19] There is evidence that the DHQ was comparable to, or superior to other FFQs that have been used in other large cohort studies.[20,21] The instrument, which takes about 60 minutes to complete, has questions about 124 food items and dietary supplements, with additional embedded questions within 44 of these items. In collaboration with the NCI, changes were made to the questionnaire and nutrient database to account for differences between the US and Canada in food availability, brand names, nutrient composition and fortification practices.[22]

Past Year Total Physical Activity Questionnaire (PYTPAQ)

The PYTPAQ was based on a questionnaire developed to measure lifetime total physical activity (LTPAQ). The LTPAQ is an interviewer-administered questionnaire that provides a reliable lifetime measure of occupational, household and recreational activities from childhood to present.[23] Frequency, duration and intensity of all types of activity (i.e., occupational, household, recreational) are recorded to yield

TABLE 2
Comparison of participant baseline characteristics reported in the Alberta Cohort Study and the Canadian Community Health Survey (CCHS; Cycle 1.1) for the Province of Alberta

		Alberta Cohort Study [a] Raw data		Alberta Cohort Study-Weighted [b]	Canadian Community Health Survey (1.1) [c]
		N	%	%	%
Marital status	Living with a partner	9,059	78.9	76.8	76.4
Household income before tax	<$50K per year	4,330	38.7	35.0	38.2
	$50K–79.9K per year	3,383	30.2	29.8	30.8
	≥$80K per year	3,480	31.1	35.2	31.0
Education	Less than high school	1,435	12.5	10.5	19.3
	Completed high school	4,808	41.9	39.2	27.1
	Completed post-secondary education	5,229	45.6	50.3	53.6

[a] Includes only the first person recruited in each household and excludes transgender individuals.

[b] Reflects percentage of responses in each category following weighting by regional health authority of residence, age and sex.

[c] Data for CCHS subjects aged 35–69 years, living in the province of Alberta.

measures of expended energy within each type of activity area and an overall measure of the energy cost of physical activity. To produce the PYTPAQ, the LTPAQ was adapted for self-administration and the reference period was changed from lifetime to the 12-month period preceding questionnaire completion. A separate study to evaluate the reliability and validity of the PYTPAQ was also conducted.[24]

All questionnaires are available on request.

Data handling and analysis

TeleForm® software (TeleForm V8.1; Verity, Sunnyvale CA USA) was used for automated optical scanning and data capture of HLQ and DHQ data, while PYTPAQ data were entered using Blaise® software (Westat, Rockville, MD USA). Routine quality checks were performed before and after data entry, and telephone follow-up was used to clarify ambiguous data. HLQ, DHQ and PYTPAQ data were linked by subject identification number, and no subject identifiers were stored with questionnaire data. To ensure security, all electronic data were stored on servers with limited access, and all files were password protected and backed up on a daily basis.

Data cleaning and analyses were done using the SAS® statistical software program (SAS V9 2003; SAS Institute Inc, Cary NC USA).

Results

Recruitment and enrolment

The four waves of telephone calling were carried out between October 2000 and June 2002, resulting in 77,327 randomly selected households being contacted. A screening interview to identify eligible residents was not completed in 38.9 percent of households; in most of these cases, the person answering the telephone could not be engaged in the interview. A screening interview was completed in 61.1 percent of selected households, and an eligible individual was recruited for possible study enrolment (i.e. willing to consider study participation) in 47.9 percent of these households. The remainder were ineligible and thus excluded for reasons of age outside the target range (89.4 percent); history of cancer (7.2 percent); expecting to move away from Alberta within the following year (2.9 percent); and, unable to understand and complete the study material in English (0.5 percent).

In 2,527 households in RDD1, we attempted to select a second participant of the opposite sex in households where a first person was successfully recruited. As a result, 711 subjects were recruited as "second in household". Of these, 384 (54 percent) enrolled in the cohort; this double recruitment strategy required, on average, two additional telephone calls to the household. The combined response from eligible first and second contacts was 56.7 percent compared to a response of 47 percent in households where only one person was recruited.

Of the 22,652 eligible individuals who were recruited, 52.4 percent (N = 11,865) enrolled in the cohort between February 25, 2001 and June 30, 2003. It is estimated that the enrolled sample represents about 32 percent of all potential participants; exact percentages cannot be given as we do not know the eligibility of those who did not complete the screening interview.

The enrolled sample of 11,865 represents about one per cent of the Alberta population aged 35 to 69 (based on the population estimate for 2002) and 84 percent of Alberta communities and municipalities are represented. Figure 1 shows regional study enrolment; enrolment outside the major metropolitan areas ranged from 44 percent to 58.6 percent and in the urban areas it was similar at 48.2 percent and 54.0 percent in the Edmonton and Calgary regions, respectively (Table 1). Non-urban participants were selectively overrepresented as planned.

Of those enrolled, approximately 88 percent returned completed DHQs and PYTPAQs.

Baseline characteristics of cohort participants

The cohort was made up of 4,907 men (41.4 percent), 6,956 women (58.6 percent) and two transgender individuals.

In order to examine whether or not the cohort was similar to the Alberta population, a comparison was made between the

TABLE 3
Health related behaviours reported by participants in the Alberta Cohort Study and the Canadian Community Health Survey (CCHS; Cycle 1.1) for the Province of Alberta

		Alberta Cohort Study[a] Raw data		Alberta Cohort Study-Weighted[b]	Canadian Community Health Survey (1.1)[c]
		N	%	%	%
Smoking status	Non-smoker	8,337	78.2	79.2	71.2
Mammogram ever	Females ≥50yr	2,518	94.2	94.4	85.6
Pap test ever	All females	6,247	99.1	99.0	94.6
PSA test ever	Males ≥50yr	830	50.3	54.0	43.3
Obesity (BMI ≥ 30)	Males	1,148	28.3	25.5	19.4
	Females	1,447	24.8	23.5	17.4

[a] Includes only the first person recruited in each household and excludes transgender individuals.

[b] Reflects percentage of responses in each category following weighting by regional health authority of residence, age and sex, and further weighted by household income and educational attainment.

[c] Data for CCHS subjects living in the Province of Alberta.

Alberta cohort respondents and Alberta respondents from the CCHS (cycle 1.1)[25] carried out between September 2000 and November 2001. The latter survey has a response rate of about 85.1 percent in Alberta,[26] and is commonly used to reflect population-based estimates of health and behaviours.

Following exclusion of the transgender subjects and those recruited as "second in household", the cohort data were weighted to the CCHS population frequency estimate, stratified by sex, age and RHA of residence at the time of recruitment. As shown in Table 2, the cohort sample was comparable to the CCHS sample in terms of marital status and annual household income below and above $50,000; the median family income in Alberta in 2001 was $60,100.[27] The proportion of the samples with post-secondary education was similar, but there were fewer individuals in the cohort with less than high school education.

Prior to comparing health behaviours, the cohort sample was further weighted by educational and income levels (Table 3). Even following this adjustment, the cohort group had more non-smokers than the CCHS group and a higher prevalence of obesity (body mass index ≥ 30). In both groups, the majority of women had had at least one Pap smear. For women over 50, a greater proportion from the cohort reported having had at least one mammogram (94.4 percent versus 85.6 percent). Similarly, for men over 50, prostate specific antigen history was higher in the cohort group (54.0 percent versus 43.3 percent).

These comparative analyses were based on Statistics Canada's Canadian Community Health Survey, Cycle 1.1, Public Use Microdata File, which contains anonymized data collected in the year 2000/2001. All computations on these microdata were carried out by staff employed by the Division of Population Health and Information at the Alberta Cancer Board, and the responsibility for the use and interpretation of these data is entirely that of the authors.

Consent for health file linkage

As part of their written consent, participants agreed to periodic data linkages with the Alberta Cancer Registry to identify incident cases of cancer. The consent also specifically asked participants for authorization to allow the Alberta Cohort Study to request health services utilization data held by the provincial health ministry (Alberta Health and Wellness); if they agreed, they were asked to provide their PHN. The majority of men (95.8 percent) and women (98.1 percent) consented to this aspect of the study and provided their PHN (Table 4).

Willingness to participate in blood collection studies

A separate form included in the enrolment package asked participants to indicate their willingness to be contacted in the future to consider providing a blood sample for study purposes. Approximately 91 percent of men and women gave a positive response to this proposal (Table 4).

Discussion

One of our primary questions was whether Canadian individuals not affiliated with any particular profession, association or known registries would agree to be part of a long-term prospective study. While cohort studies of this type have been initiated in Europe, there was considerable question as to whether North Americans, in an environment of increasing concern over privacy in the 21st century, would be willing to participate. Our results have shown that enrolment of such a cohort is indeed possible, and that the response rate obtained (32 percent), although lower than would be desirable for simple cross-sectional studies, is comparable to cohort studies elsewhere in the world, recruited in earlier time periods. For example, a Swedish population-based cohort reported 40 percent participation,[28] the Utrecht EPIC study reported a 34.5 percent participation rate,[29] and the German EPIC study reported enrolment of 22.7 percent in Potsdam and 38.3 percent in Heidelberg.[30] Among single-sex cohorts, a national sample of Dutch women had a response rate of 35.5 percent,[31] the Iowa Women's Health Study reported 42 percent enrolment,[32] 57.1 percent participation was reported in a women's cohort in Norway[33] and 51.3 percent was reported in Sweden.[33]

Since the main rationale for most cohort studies is the investigation of etiologic hypotheses, such response rates are not of concern for internal validity.[34] In fact, even when restricted populations, such as the cohort of women in the Nurses' Health Study, are used as the enrolment sampling frame, about 51 percent of the letters sent resulted in enrolment.[35] This has not precluded using these data for the investigation of etiologic hypotheses.

TABLE 4
Alberta Cohort Study: Consent rates for linkage with provincial health care utilization data, and for future blood studies

		Total N[a]	Provided personal health number and consent for data linkage % yes[b]	Willing to be contacted about providing future blood sample % yes[b]
Males	35–44 yr	1,806	94.1	89.6
	45–54 yr	1,720	96.5	91.6
	55–64 yr	1,011	97.5	93.6
	65–69 yr	370	96.8	92.2
	Total	**4,907**	**95.8**	**91.3**
Females	35–44 yr	2,519	97.9	90.8
	45–54 yr	2,398	98.2	93.3
	55–64 yr	1,477	98.2	92.0
	65–69 yr	562	98.0	90.2
	Total	**6,956**	**98.1**	**91.9**

[a] Total number of participants in each age category (includes participants recruited as 'second in household').

[b] % of participants within each age category who responded positively to request.

The lower response rates for cohort studies, as opposed to case-control or cross-sectional studies, are hardly surprising given the far higher degree of commitment asked of cohort participants. In our study, we asked participants to be willing to be followed until the age of 85 or until death, and similar commitments are expected in other prospective studies. In fact, we believe that the intensity of the questionnaire process used in this first enrolment phase of the study was a useful study component, not unlike the "run-in" period used in long-term randomized controlled trial designs.[36,37] Other cohort investigators have also noted this potential advantage,[30] since those who do enrol are more likely to continue the follow-up over a number of years. Indeed, this possibility has now been corroborated by the results of our first follow-up survey, in which approximately 92 percent of those fully enrolled did complete and return the questionnaire. Such a response bodes well for future follow-up.

However, thought has to be given to what degree the cohort data can be used to determine answers to population-based questions. Our data indicate that some caution would need to be used in attempting to use cohort data to reflect prevalence of health behaviours. There is a slight tendency towards "healthy enrolee" effects in a long-term study group, and our group had higher non-smoker rates and slightly higher "ever use" of screening tests. However, the fact that the cohort group had a higher prevalence of obesity than the CCHS group indicates the presence of more than a simple bias. It is possible that some, although not all, of the difference between the two groups reflects the later secular time period for collecting the cohort data; smoking rates have been decreasing in Alberta, while use of screening tests and obesity rates have been increasing. It is interesting to note that the differences between the two groups in health practices reflect the same differences in secular trend.

In fact, some would argue that the use of such population-based information can be extrapolated further, to the calculation of population attributable risk and preventable proportions, provided known exposure rates in the general population are applied to the cohort data in question.[28,30] It should be noted that even obtaining such point estimates in current cross-sectional studies is becoming more difficult; the participation rates for such cross-sectional studies as the Behavioural Risk Factor Surveillance Study, used to gauge health behaviours in the USA, has participation rates of 42.4 percent overall and as low as 24.0 percent in some states.[38] Thus, any surveillance of population-based risk factors is likely to be subject to increased selection pressure in future.

One of the intended applications of this cohort is the ability to observe the outcome of "natural experiments". By this we mean that we will have the ability to observe how local changes in policy or environmental conditions affect cohort participants in the affected environment as compared to the "controls" in the stable environments. This particular application of population data will not be affected by the population selection pressure since one will be able to either select matched controls or control for potential effect modifiers within the entire design; the same arguments about internal validity apply as when one is using the cohort data to examine etiologic hypotheses. In addition, the fact that we were able to successfully enrol over the entire geographic area of the province, with similar uptake rates around the province, predicts that we will be able to apply the cohort results to monitor such effects in future.

Other recruitment strategies, such as collaborating with Statistics Canada's CCHS or using health care insurance files, were explored but found to be not feasible. The CCHS-affiliated method would have introduced a selection bias, as consent for cohort enrolment could only be asked for at the conclusion of CCHS interviews, and then only when the interview was concluded face to face. Because Statistics Canada indicated that many interviews were concluded on the telephone, it was decided to try a method where at least the first approach to the individual was carried out via random selection. Timeliness issues argued against attempting to use health care insurance files. Thus, random digit dialing (RDD) became our primary recruitment method. Because RDD does not depend on telephone directory listings, all households with telephone lines had an equal chance of being called. Since 97 percent of households had at least one telephone line at the time, using a telephone-based, RDD sampling method meant that almost all households were included in the theoretical sampling frame. However, we are now starting to become concerned that the RDD telephone-based recruitment

approach is likely to become less effective in the near future. In the USA, there is growing disquiet concerning declining response rates to RDD methods of contacting potential subjects.[39,40] Declining response rates not only raise concerns about the ability of the resulting cohort to reflect even broadly the characteristics of the general population, but may also have severe fiscal consequences as the length of time and amount of effort required to achieve the desired sample numbers increases. It is therefore possible that other recruitment methods may be evaluated against the RDD experience before future waves of enrolment are undertaken in the Alberta Cohort Study.

While the double recruitment strategy was somewhat effective in increasing the number of potential study subjects, the potential "cost" of this approach outweighed the benefit. That is, a relatively high portion of the RDD1 sample (28.1 percent) shared a household with another person recruited for the study (most often a spouse). If a large proportion of these individuals along with their household partners were to enrol in the study, the potential of having a high degree of correlation present in the study data, especially on measures of exposure would outweigh the advantages of using the "second person" strategy. Despite the added efficiency, the method was not used in subsequent RDD recruitment waves.

There had been considerable media discussion about information privacy concerns and the potential ethical challenges surrounding the collection of biologic samples for long-term storage and use prior to and during our study enrolment. In fact, the Office of the Information and Privacy Commissioner was established in Alberta in 1995, presumably—and in part—to address public concerns in this area. However, the consent for use of personal health information and to be contacted for biologic specimens was extremely high. Undoubtedly, some of this is due to the research-oriented nature of individuals who choose to participate in such a study; those who were uncomfortable with this request may have elected not to participate in the cohort at all. Furthermore, the somewhat abstract notion that they may be contacted in the future to consider providing a blood sample may have encouraged some subjects to respond positively to the request without thinking it through. In order to estimate how many people would provide a sample if asked, we subsequently conducted a small pilot study, which demonstrated that approximately two thirds of those approached would give a 50 mL sample of blood for banking. These samples (N = 769) have been processed to provide multiple aliquots of serum, plasma, red blood cells and buffy coat, which are being stored at -85°C in mechanical freezers. The planned collection of further blood samples will necessitate expansion of the existing bio-repository and establishment of protocols to guide the granting of access to the samples for further research. All further research on the samples will be subject to full ethical approval.

In conclusion, the results of this feasibility study suggest that cohort development in the Canadian context is feasible, and that the potential for future studies using biological samples to determine prevalence of biomarkers—or correlations between reported exposures or disease outcomes with biomarkers—appears to be high. It is our hope that the information in this feasibility study will be of use to the much more extensive discussions that will need to take place in proposing the ultimate design, funding and administration of a full-scale cohort.

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Copies of the research instruments are available on request.

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Patients' opinions on privacy, consent and the disclosure of health information for medical research

Stacey A Page and Ian Mitchell

Abstract

A structured survey of patients in three illness groups (acquired immune deficiency syndrome, multiple sclerosis and mental disorders) was undertaken to describe patients' perspectives on privacy, consent and the use of their health information for medical research. The survey was distributed by mail to subjects in the AIDS and MS groups and was completed in a clinic waiting room by people in the mental disorders group. Of the 478 patients approached for participation, 235 returned completed surveys (response rate 49.2 percent). Most subjects were concerned about privacy and they valued opportunities to provide consent for the use of their personal health information for research. Contextual factors, such as identification, type of illness and who was conducting the research, were important to individuals' preferences in granting consent. When health information was used specifically for research, the majority of subjects wanted to be asked for their consent unless anonymity was assured. Privacy and control over personal health information were important to patients in these groups. Patients prefer to be asked for research access to their health information.

Key words: *confidentiality, informed consent, privacy, secondary data*

Introduction

Personal health information includes patient sociodemographic information, as well as that for diagnostic, treatment, care and scheduling/billing. Patients share information with health care professionals in the belief it is related directly to their health care. However, it is also of considerable interest to others, including health researchers.

Most literature considering the secondary use of health information for medical research consists of editorials and theoretical discussions.[1–5] Privacy advocates believe the values of privacy and autonomy make it morally unacceptable to use personal health information for other than direct patient benefit without patients' knowledge or consent. In contrast, those espousing a more communitarian view argue that the right to medical care should generally include a collateral responsibility to allow information gained in its course to be used for the benefit of others. These proponents suggest that it is unethical to hinder legitimate research by placing onerous restrictions on access to personal health information since this compromises the research benefits that both society and the individual may reap.[3,4,6]

US opinion surveys conducted in the early 1990s suggested that respondents were uncomfortable with the unauthorized use of personal health information even with assurances of confidentiality and REB oversight.[7,8] A more recent poll revealed that most adults opposed non-consensual access to health information by any group (e.g., MDs, pharmacists, police/lawyers, health departments, banks, employers, insurance companies, government agencies). Two thirds of respondents were opposed to medical researchers accessing their medical records without consent.[9]

The National Health System (NHS) Information Authority in Great Britain conducted a study examining confidentiality and consent issues from the perspectives of patients and the public.[10] Although respondents expressed a high level of trust in the NHS to protect confidentiality, many were also unaware of how information was actually used. Who used the information and whether it was anonymous was of greater concern than how the information was used. Respondents believed that information used outside the NHS or for reasons other than treatment should be anonymized or that consent should be sought for its use. Women and people identifying themselves as being of Caucasian origin were likely to set more stringent requirements for consent.

A recent Canadian survey examined patients' preferred methods of obtaining consent for the use of electronic medical records.[11] Again, few people had thought about how their health information was used. Most subjects were willing to allow their health information to be used for research, although they preferred that, out of respect, consent be obtained first.

Understanding patients' opinions and expectations in these matters is important to the continued development, evolution and implementation of regulations governing access to personal health information in both the clinical and research contexts.[1,10,12]

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The purpose of this study was to examine perspectives of members of three patient groups regarding consent and the disclosure of health information for medical research. Its conduct was approved by the Conjoint Health Research Ethics Board, University of Calgary.

Methods

Sample

Approximately 200 adults from each of three patient groups (people with AIDS, a mental disorder or multiple sclerosis) were approached to participate.

Instruments

A five-page, fixed-choice questionnaire was developed for the purpose of this study based on a literature review and consultation with members of each patient group. The questionnaire was locally peer reviewed by experts in research ethics, law and privacy. A small number of individuals from each patient group were asked to review the questionnaire. Recommendations were incorporated into a subsequent version.

Questions focused on sociodemographics, experience with medical research, and opinions and experiences relating to privacy, health information and medical research. Subjects were invited to make additional comments about the use of personal health information for medical research. Questionnaire completion was anonymous.

Procedure

The procedure for administering the questionnaire varied slightly between the three participating organizations and reflected differences in client characteristics, interim response rates and organizational human resources.

AIDS support agency

A community-based organization providing supportive resources for people with AIDS mailed the survey package (covering letter, questionnaire, postage-paid return envelope) to all clients with contact information in its database (N = 200) in late November, 2003. Approximately six weeks later, survey recipients received a post card reminder and, two weeks after that, a second questionnaire package. Posters promoting survey participation were placed on the organization's bulletin boards.

Inner city mental health clinic

This clinic serves an outpatient population with a range of mental disorder diagnoses. At the time of questionnaire distribution, the clinic served approximately 450 clients. Consecutive patients presenting to the clinic reception over a five-month period beginning in November 2003 were asked if they would be willing to complete the questionnaire. If they agreed, clients were given the survey package and asked to return it before leaving the clinic. Clients were excluded if they were actively psychotic.

Multiple sclerosis support agency

A community-based organization providing supportive services to clients with multiple sclerosis mailed a survey package to 200 of 751 active clients in its database in November 2003. Every third client was selected until a total of 200 was reached. Approximately eight weeks later, a second questionnaire package was mailed.

Analyses

Data were analyzed using STATA 6.[13] Descriptive statistics were used to summarize responses from each patient group as appropriate. Thirteen items addressing opinions on consent (see Table 3) were used to construct an individual "consent index". These items examined whether or not respondents believed individual consent should be obtained for medical research using their health information under a variety of conditions. Responses indicating that consent should be obtained were scored +1, those indicating consent need not be obtained were scored –1 and those indicating the respondent was unsure were scored 0. These items were summed for each respondent. Positive sums characterized those who more often believed consent was necessary while negative sums characterized those who did not believe consent was always necessary. Respondents with a score of zero were considered undecided. This method identified two groups of respondents holding relatively strong opinions on consent and access to personal health information for medical research. Multiple logistic regression methods were used to identify factors predictive of these perspectives (alpha = 0.05). Demographic characteristics (age,

FIGURE 1
Consent index of study participants: Distribution of participants' individual summary scores for items in Table 3. Positive integers indicate preference for consent

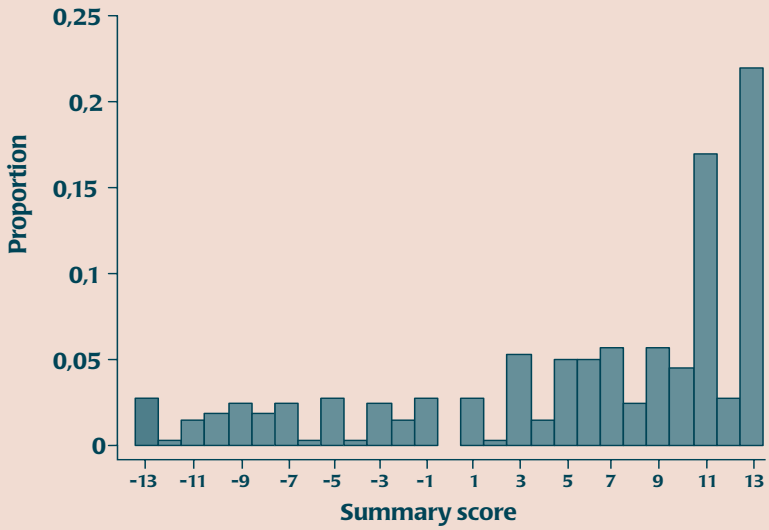

TABLE 1
Sociodemographic characteristics of paticipants by patient group

Characteristic	Patient groups: AIDS N (%)	Mental disorders N (%)	Multiple sclerosis N (%)	Total N (%)
Sex				
Male	22 (84.6)	33 (38.4)	31 (25.2)	86 (36.6)
Female	4 (15.4)	53 (61.6)	92 (74.8)	149 (63.4)
Age group				
20–39	9 (34.6)	39 (45.4)	20 (16.3)	68 (28.9)
40–59	15 (57.6)	37 (43.0)	77 (62.5)	129 (54.9)
≥ 60	1 (3.9)	7 (8.1)	26 (21.2)	34 (14.5)
≥ Unknown	1 (3.9)	3 (3.5)	0	4 (1.7)
Currently working?				
No	19 (73.1)	59 (68.6)	96 (78.1)	174 (74.0)
Yes	7 (26.9)	27 (31.4)	27 (22.0)	61 (26.0)
Education				
< Grade 12	5 (19.2)	20 (23.3)	14 (11.4)	39 (16.6)
Grade 12	3 (11.5)	15 (17.4)	17 (13.8)	35 (14.9)
Attended/finished post secondary	18 (69.2)	47 (54.6)	91 (74.0)	156 (66.4)
Refused to answer	0	4 (4.7)	1 (0.8)	5 (2.1)
Marital status				
Single*	24 (92.3)	67 (77.9)	46 (37.4)	137 (58.3)
Partnered (common-law, married)	2 (7.7)	19 (22.1)	77 (62.6)	98 (41.7)
Annual gross income				
< $20,000	12 (46.2)	34 (39.5)	28 (22.8)	74 (31.5)
$20,000 – $39,999	8 (30.8)	14 (16.3)	21 (17.1)	43 (18.3)
$40,000 – $59,999	1 (3.9)	7 (8.1)	15 (12.2)	23 (9.8)
$60,000 – $79,999	0	4 (4.7)	17 (13.8)	21 (8.9)
≥ $80,000	0	1 (1.2)	10 (8.1)	11 (4.7)
Don't know/refused to answer	5 (19.3)	26 (30.3)	32 (26.1)	63 (26.9)
Previous experience with medical research				
Yes	15 (57.7)	29 (33.7)	83 (67.5)	127 (54.0)
No	9 (34.6)	43 (50.0)	33 (26.8)	85 (36.2)
Not sure	2 (7.7)	14 (16.3)	7 (5.7)	23 (9.8)

* Includes never married, separated, divorced and widowed.

sex, marital status, employment status, education, income, research experience and illness group) were considered. Backwards, stepwise regression was performed with the resulting models evaluated using the likelihood ratio chi-square test. Textual comments were summarized using content analysis.

Results

Subjects

Questionnaires were returned by 244 people. Nine returns had substantial missing data and were excluded. The overall response rate was therefore 235/478 (49.2 percent). The response rate varied significantly by group.

AIDS patient group

Of the 200 surveys sent out, 78 were returned as undeliverable. Twenty-six questionnaires were returned for a response rate of 21 percent (26/122).

Mental disorders patient group

One hundred and eighty-seven people were approached for survey participation. Seventeen people were too ill to participate and were excluded (possible N = 170). Two people returned a questionnaire with excessive missing data and 82 people refused. Eighty-six useable questionnaires were returned (response rate 86/170: 51 percent).

Multiple sclerosis patient group

Twelve of 200 surveys were returned as undeliverable. One hundred and thirty surveys were returned, of which seven had excessive missing data. Thus, 123 useable questionnaires were received for a response rate of 65 percent (123/188).

Sociodemographic characteristics are given in Table 1.

Opinions and experiences about privacy and health information

Patients from all three groups were most concerned with the privacy of their financial and health information, whereas the privacy of their religious and political beliefs concerned them least. Across groups, the majority of patients (96 to 100 percent) believed people should be able to access their own health information. However, few had tried to do so.

From a list of people/organizations, patients were asked to identify those whom they felt could access their health information without consent. As Table 2 shows, respondents felt that physicians involved in their care were the only ones who should be able to access their health information without their consent. Most patients believed all others, including spouses and other close relatives, should have access to such information only with consent.

TABLE 2
Access to health information without consent by various stakeholders: Number and percentages of patients who would agree to it, by patient group

	Patient groups			
	AIDS N (%)	Mental disorders N (%)	Multiple sclerosis N (%)	Total N (%)
Spouse/partner	4 (15.4)	18 (20.9)	57 (46.3)	79 (33.6)
Close relatives	6 (23.1)	26 (30.2)	35 (28.5)	67 (28.5)
Physicians involved in your care	24 (92.3)	71 (82.6)	108 (87.8)	203 (86.4)
Physicians not involved in your care	4 (15.4)	6 (7.0)	11 (8.9)	21 (8.9)
Medical researchers	8 (30.8)	28 (32.6)	42 (34.2)	78 (33.2)
Pharmacists	8 (30.8)	29 (33.7)	24 (19.5)	61 (26.0)
Drug companies	2 (7.7)	5 (5.8)	3 (2.4)	10 (4.3)
Your employer	1 (3.9)	7 (8.1)	2 (1.6)	10 (4.3)
Your insurance company	2 (7.7)	10 (11.6)	7 (5.7)	19 (8.1)
The government	2 (7.7)	8 (9.3)	4 (3.3)	14 (6.0)

Contextual factors influencing need for consent to be obtained

Subjects were given the following scenario and asked if they felt that consent should be required to use their personal health information. It was emphasized that the question pertained to whether consent should be obtained, not whether they would actually grant consent.

"Medical researchers at a university are conducting a study about a medical condition that has affected you. They would like to use your personal health information in their study. This information is stored with your name on it."

Subsequently, a number of variables in this scenario were changed and in each case subjects were asked if their consent needed to be obtained. These results appear in Table 3.

Across conditions, there was variation in the proportions of subjects needing consent to be sought. Except in one variation of the scenario, the majority of subjects believed that consent should be required to use their health information for medical research. Subjects demonstrated greatest consensus on this when it was possible to identify the person whose information was used (>78 percent across groups believed consent was needed) and when the information was sensitive in nature (>84 percent across groups believed consent was needed). Subjects were most undecided about the need to seek consent when doing so was not feasible for the researchers (17 percent not sure). The only condition under which the majority of respondents believed consent for personal information access did not need to be sought was when the information was anonymous (63 percent seeking of consent not required).

The items in Table 3 were used to construct the consent index, as described earlier. Scores on the consent index revealed that for the sample as a whole, 184 (78.3 percent) were consent advocates—more often of the opinion that consent was necessary for access to personal health information. The difference in proportions between patient groups was not significant (Pearson $chi^2 = 3.5$; $p = 0.17$). The range of scores determined for the consent index is shown in Figure 1.

Sex and employment status were the only factors found to predict consent support. Specifically, women and those who were employed were more likely to be consent advocates ([OR = 1.96; 95% CI: 1.04-3.71] and [OR = 2.29; 95% CI: 1.00-5.25], respectively).

Opinions about health research

Subjects rated on a five-point Likert scale the extent to which they agreed or disagreed with six statements about health research, and when consent for research access to personal health information should be required. Since illness group was not found to influence opinion on consent and the disclosure of health information for research purposes, the data for the three groups were combined. These results are shown in Table 4.

Textual comments

Nineteen people made additional comments relating to privacy issues. Most of these comments reflected concern for privacy and emphasized the need for consent to be obtained prior to the use of personal health information for medical research.

Some of the comments are included below:

> "I strongly believe that a person's medical history must be protected, as this information could be harmful if obtained by others who could use it in a damaging way." (resp 96)

> "I am more interested in doing research if I am being interviewed or answering questions. I am somewhat leery of having my info out there accessible to many people. Somehow there is always a trust issue there ... it is much easier to trust if you are involved and meet someone involved in the research. The idea of any medical professional accessing my info bothers me." (resp 233)

> "Basically, I believe a person should be asked always. Personally, in most situations I would say 'yes', but I should always be asked." (resp 50)

Discussion

Patients are clearly concerned about the privacy and security of their personal health information. Most respondents preferred that their consent be obtained before using their information for health

TABLE 3
The influence of different factors on patients' consent preferences for medical research, by patient group

Factor	Patient groups			
"Would you like to be asked for your consent if . . . "	AIDS N (%)	Mental disorders N (%)	Multiple sclerosis N (%)	Total N (%)
. . . the information is stored with your name on it (i.e., you could be identified).				
Yes	23 (88.5)	75 (87.2)	97 (78.9)	195 (83.0)
No	3 (11.5)	9 (10.5)	23 (18.7)	35 (14.9)
Unsure	0	2 (2.3)	3 (2.4)	5 (2.1)
. . . the medical condition is something very serious (e.g., cancer, heart disease, Alzheimer's disease).				
Yes	21 (80.8)	53 (61.6)	88 (71.5)	162 (68.9)
No	4 (15.4)	27 (31.4)	28 (22.8)	59 (25.1)
Unsure	1 (3.9)	6 (7.0)	7 (5.7)	14 (6.0)
. . . the medical condition is something relatively minor (e.g., ear infection, muscle strain, headache).				
Yes	18 (69.2)	60 (69.8)	79 (64.2)	157 (66.8)
No	7 (26.9)	24 (27.9)	41 (33.3)	72 (30.6)
Unsure	1 (3.9)	2 (2.3)	3 (2.4)	6 (2.6)
. . . the medical condition is something "sensitive" (e.g., sexual problems, sexually transmitted disease, mental illness).				
Yes	22 (84.6)	80 (93.0)	106 (86.2)	208 (88.5)
No	3 (11.5)	4 (4.7)	14 (11.4)	21 (8.9)
Unsure	1 (3.9)	2 (2.3)	3 (2.4)	6 (2.6)
. . . the research is likely to help you directly.				
Yes	21 (80.8)	69 (80.2)	91 (74.0)	181 (77.0)
No	4 (15.4)	12 (14.0)	28 (22.8)	44 (18.7)
Unsure	1 (3.9)	5 (5.8)	4 (3.3)	10 (4.3)
. . . although the research will not help you, it is likely to help others.				
Yes	18 (69.2)	68 (79.1)	87 (70.7)	173 (73.6)
No	7 (26.9)	13 (15.1)	31 (25.2)	51 (21.7)
Unsure	1 (3.9)	5 (5.8)	5 (4.1)	11 (4.7)
. . . the information can be obtained by questioning another member of your family.				
Yes	23 (88.5)	69 (80.2)	92 (74.8)	184 (78.3)
No	3 (11.5)	12 (14.0)	21 (17.1)	36 (15.3)
Unsure	0	5 (5.8)	10 (8.1)	15 (6.4)
. . . the information is in a database and is identified only by a number. That is, the information is not linked directly to your name and your identity will remain unknown to the researchers.				
Yes	8 (30.8)	35 (40.7)	34 (27.6)	77 (32.8)
No	17 (65.4)	45 (52.3)	85 (69.1)	147 (62.6)
Unsure	1 (3.9)	6 (7.0)	4 (3.3)	11 (4.7)
. . . the researchers have assured you that your information is secure. Although your identity will be known to them, they will keep this information secure and your identity will never be revealed to others outside the research team.				
Yes	15 (57.7)	62 (72.1)	79 (64.2)	156 (66.4)
No	9 (34.6)	22 (25.6)	38 (30.9)	69 (29.4)
Unsure	2 (7.7)	2 (2.3)	6 (4.9)	10 (4.3)
. . . the information must be obtained from another 5,000 patients like yourself. The researchers have stated that getting consent from everybody is not feasible and they will not be able to carry out the research if they have to get consents.				
Yes	15 (57.7)	49 (57.0)	57 (46.3)	121 (51.5)
No	10 (38.5)	22 (25.6)	42 (34.2)	74 (31.5)
Unsure	1 (3.9)	15 (17.4)	24 (19.5)	40 (17.0)

TABLE 3 (continued)
The influence of different factors on patients' consent preferences for medical research, by patient group

Factor	Patient groups			
"Would you like to be asked for your consent if . . . "	AIDS N (%)	Mental disorders N (%)	Multiple sclerosis N (%)	Total N (%)
. . . your own doctor is conducting the research.				
Yes	13 (50.0)	58 (67.4)	76 (61.8)	147 (62.6)
No	13 (50.0)	25 (29.1)	44 (35.8)	82 (34.9)
Unsure	0	3 (3.5)	3 (2.4)	6 (2.6)
. . . a drug company is conducting the research.				
Yes	19 (73.1)	74 (86.1)	102(82.9)	195 (83.0)
No	4 (15.4)	11 (12.8)	17 (13.8)	32 (13.6)
Unsure	3 (11.5)	1 (1.2)	4 (3.3)	8 (3.4)
. . . a research ethics board, made up of doctors, lawyers, nurses, research experts and ordinary people from the community, has looked at the research proposal. They have decided it is an important study and will not cause you any harm.				
Yes	16 (61.5)	57 (66.3)	86 (69.9)	159 (67.7)
No	10 (38.5)	20 (23.3)	26 (21.1)	56 (23.8)
Unsure	0	9 (10.5)	11 (8.9)	20 (8.5)

research. Although the response rates by the patients with mental disorders and the patients with multiple sclerosis were reasonable, non-response bias is a threat to the generalizability of these findings. In particular, the AIDS patient group had a very low response rate. This was likely due to the transient nature of the population served by this community-based organization, evidenced by the high proportion of questionnaires returned as undeliverable. Those who responded from this group did, however, express views similar to those of individuals in the other two groups.

Access to health information

Respondents felt that soley the physicians directly involved in their health care should be able to access patient personal health information without consent. This reflects the fiduciary nature of the physician-patient relationship, in which patients provide intimate details about their physical and mental health based on the assumptions that information will be kept confidential and that it will be used for purpose of providing care. This is consistent with previous findings indicating that patients value the inter-professional exchange of their health information and view such disclosure as vital to their care.[14]

Not surprisingly, treatment has been rated as the most important use of health information by patients.[10] When asked about their physicians having access to health information for research, the proportion of patients indicating they would agree to waive the requirement for consent fell considerably from the levels reached in the clinical circumstance.

For all other third parties, most respondents would deny access to their health information without consent, a view that strengthened as the described relationship with the third party grew more remote. Consistent with findings from previous studies,[7,11,15] a majority of respondents objected to the use of their health information for research without consent.

Conditions that influenced need for consent

Across most items, the majority (63 to 89 percent) felt their consent should be sought when using their health information for medical research. Variation in these proportions was observed, however, indicating that contextual factors are important to individuals. Respondents were most likely to shift their views from "requiring" to "waiving" consent when the information sought was anonymous and individual identification was impossible. Nevertheless, one third of respondents maintained the perspective that their consent be required even if the data were unidentifiable.

A similar, though smaller, shift occurred when researchers presented the situation where obtaining consent would not be feasible and the research could not proceed if individual consents were needed. Moreover, a substantial proportion of respondents were undecided on this issue. Possibly, further details on other conditions of the research would be necessary for subjects to come to an opinion under this condition.

Assurances that confidentiality would be maintained or that an REB had reviewed and approved the research did not alter the majority view that consent should be sought. Even with the provision that the research might directly benefit them or others, most respondents still believed their consent for use of personal information should be requested.

Opinions on consent and medical research

Most respondents were "consent advocates" around the issue of use of personal health information for medical research.

Nevertheless, almost one third of respondents felt that it would be sufficient to simply be informed that their information was being used for research, paralleling earlier Canadian findings.[11]

Respondents were more in favour of providing consent for each new research project than of giving blanket consent for research purposes in general. Similarly, the majority of respondents in previous surveys preferred to give consent each time their health information was accessed, whether this was for treatment or other purposes.[7,10]

As previously described, even when provided with the assurance their data was not associated with their name and that their identity would remain unknown, 33 percent of the respondents believed researchers should be required to obtain their consent before using their health information for medical research (e.g., item 8, Table 3). In contrast, when this question was presented more generally (e.g., item 2, Table 4), the proportion of respondents requiring consent increased to 46 percent and an increasing proportion were unsure. It may be that respondents needed the additional detail provided in the first presentation of the question in order to provide a response. Alternatively, the first question may have reflected a more personal response, while the latter, more conservative, response reflected what they perceived should happen in society generally. Ambiguities and inconsistencies in patient reports in this area have been reported previously.[11]

Conclusion

The secondary use of personal health information for research has provoked much discourse, centering on the balance between individual rights and societal benefit.[1–5,16–18] Privacy and autonomy are core values in the population, reflected in the expressed concerns around use and disclosure of personal information of any sort. These findings indicate that respecting these rights in a research context means requiring consent for research access to health information. However, the community also values the benefits that accrue to individuals and society from medical research. These findings suggest that this valuing does not extend to a widespread willingness to trade loss of privacy for the public good.

Previous research that has shown that people have little awareness of how personal health information is used,[10,11] and it is possible that this lack of understanding is what mediates the reluctance to allow greater access to personal health information. There may be a need for public education to foster trust in the health research process and to provide the public with a broader, societal perspective on the uses of personal health information. Future studies should focus on understanding the rationale behind the public's perspectives.

For their part, researchers must be sensitive to the public's desire to control the use of their individual health information. Research ethics boards in Alberta and elsewhere have the legislative and institutional authority to permit researchers access to identifiable information without individual consent under certain conditions. These include factors directly related to the study, such as risk being minimal and study outcomes being in the public's interest. Consent may also be waived if, in addition to the previous considerations, the process of obtaining consent is deemed to be unreasonable or impractical.[4,16,19–22] What actually constitutes unreasonableness or impracticality is often unclear and may be a consequence, in part and in some cases, of insufficient resources. Increasing the budget amounts dedicated to obtaining individual consents is a potential means of addressing this issue.

TABLE 4
Opinions on matters of research and consent of three sampled groups (combined)

Issue	Agree/ strongly agree N (%)	Neutral N (%)	Disagree/ strongly disagree N (%)
Research using individually identified health information is important to the development of medical care.	114 (48.5)	58 (24.7)	52 (22.1)
Researchers should be able to use unidentifiable personal health information without a person's consent.	80 (34.0)	43 (18.3)	109 (46.4)
Researchers must always get a person's consent to use identifiable health information.	182 (77.4)	28 (11.9)	22 (9.4)
Consent to use health information should be obtained each time a new research project is starting.	165 (70.2)	32 (13.6)	34 (14.4)
Consent to use health information need only be obtained once for all future research projects.	53 (22.5)	37 (15.7)	142 (60.5)
People should be informed that their health information is being used: They do not have to give consent.	70 (38.3)	33 (14.0)	106 (45.1)

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Statistical disease cluster surveillance of medically treated self-inflicted injuries in Alberta, Canada

Rhonda J Rosychuk, Cynthia Yau, Ian Colman, Don Schopflocher and Brian H Rowe

Abstract

Routine surveillance of cases of disease can highlight geographic regions that need further study and intervention. Statistical disease cluster detection methods are one way to statistically assess the number of cases in administrative areas. Traditionally, disease cluster detection methods are used to monitor the incident cases of disease. We review a statistical cluster detection method that is applicable for regions with diverse administrative area population sizes. We apply the method to assess clustering of self-inflicted injury presentations to emergency departments in Alberta, Canada. Analyses focus on the pediatric population and are adjusted by the age and gender distributions of sub-regional health authorities. Fifteen clusters of self-inflicted injuries are identified and, based on age and gender distributions, the clusters are not likely chance occurrences. We believe that these clusters represent areas of excessive self-inflicted injury and that special intervention programs should be considered.

Key words: *emergency service, hospital, injuries, space-time clustering, wounds*

Introduction

Health authorities are often alerted to areas of suspected high disease rates by medical personnel and members of the general public. Investigations, costly in time and resources, are likely to produce false alarms. Even in the absence of local reports, public health offices routinely monitor regions for new cases of illness and disease. This surveillance can identify geographic regions where further investigations or interventions can be focused. Statistical disease cluster detection methods are one way to assess if any geographic regions have more cases than could be expected by chance alone.

Statistical disease cluster detection methods identify regions with excessive cases either in space (i.e., close to each other), over time (i.e., temporal proximity) or both. The aggregation of cases are referred to as "clusters". These detection methods can involve general or focused testing.[1] General tests identify any clusters with elevated cases, whereas focused tests identify areas of excess cases near potential point sources of influence, such as environmental contaminants.

Traditionally, statistical disease cluster detection methods are exploratory tools that have been developed for and applied to disease incidence data. Examples include identification of clusters of squamous cell carcinoma,[2] leukemia,[1,3–5] various cancers,[6] serious cardiac birth defects[7] and childhood diabetes.[8]

There are a variety of methods available that are suitable for testing specific hypotheses in particular data situations. For identifying the most likely cluster in a geographic area, tests by Turnbull et al.[4] and Kulldorff and Nagarwalla[6] can be used. On the other hand, Besag and Newell[1] and Tango[10] provide methods that are designed to identify regions with a tendency to cluster. For an overview of these methods and the more general topic of disease mapping, readers are directed to Lawson et al.[11]

In this paper, we apply one statistical disease cluster technique in a non-traditional way. The definition of a case is based on a presentation of a self-inflicted injury to an emergency department (ED). These injuries are often referred to as "suicide attempts" and are relatively common ED presentations. Three to five percent of individuals indicate they have attempted suicide in their lifetime.[12] Individuals who harm themselves may suffer from a variety of problems including abuse, chronic illness, psychiatric disorders and financial difficulties.[13–18]

As these individuals are at risk for further self-inflicted injuries and suicide,[19,20] identification of geographic areas of excess self-inflicted injuries can be an important first step in directing research efforts and interventions to high-risk populations. Our analyses incorporate the age and gender distributions of sub-Regional Health Authorities (sRHAs) to identify clusters of medically treated self-inflicted injuries.

Materials and methods

Study population

The study population represented all 785,079 individuals under 18 years of age

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residing in the western Canadian province of Alberta between April 1, 1998 and March 31, 1999. Health care is the responsibility of a government ministry called Alberta Health and Wellness. The province was divided into nine Regional Health Authorities that served as administrative units for the delivery of health services and which were further sub-divided into 68 sRHAs[21] (Figure 1). The sRHA population sizes ranged from 2,658 to 36,632 individuals (Figures 2a and 2b). The two major metropolitan centers, Calgary and Capital, had the highest populations and the smallest geographical regions. Conversely, the geographically large sRHAs had sparse population counts.

Data

The Ambulatory Care Classification System (ACCS) was established by Alberta Health and Wellness in 1997 and was the first of its kind in Canada. ACCS is an extensive database which records augmented information on all services provided by hospitals on an outpatient basis, including ED presentations to acute care hospitals. This replaced an older limited data set which was collected through the Hospital Reporting System of the Canadian Institute for Health Information. Since the ACCS is used to assign funding to Regional Health Authorities for emergency care, there is a strong incentive to insure that the system is accurate. The history of the system and its validation are detailed by Alberta Health and Wellness.[22]

ACCS contained 1.5 million records during the study period. All records of ED presentations were coded with up to five diagnoses using the International Classification of Diseases, 9th revision, Clinical Modification,[23] by trained medical records nosologists as part of the data collection procedure.

For each individual, ACCS includes a unique lifetime identifier, which allows the record to be linked to the Alberta Health Care Insurance Plan Stakeholder Registry to retrieve demographic information. The ACCS records and the Stakeholder Registry were linked to create a research data set. The Alberta Health Care Insurance Plan charges a premium for health care insurance. As a result, there is a strong motivation to maintain the accuracy of the database. Comparisons of population estimates between the Stakeholder Registry and the Canadian Census have been made and show that the Registry functions as a dynamic population registry. Statistics Canada now uses the Registry populations to adjust the census population estimates for intercensal projections.[24]

The research data set included the age, gender, health care insurance premium subsidy status, sRHA of residence, and number of ED presentations for each patient as well as other detailed information about each presentation, such as the date and time of the encounter. The self-inflicted injury presentations used in the analysis are based on External Causes of Injury codes E950 to E959 ("Suicide and Self-Inflicted Poisoning", "Suicide and Self-Inflicted Injury"). The median number of self-inflicted injuries per person was one (range 1 to 18). Individuals may present with more than one self-inflicted injury during the study period; we restrict our analyses and define "a case" as an individual with at least one self-inflicted injury. There were 827 pediatric cases in the province and the number of cases in each sRHA ranged between 0 and 45 (Figure 2). A full description of the self-inflicted injury ACCS data appears in Colman et al.[25]

For each sRHA, the population, a population-based centroid and the intercentroid distances were calculated. The mid-year population sizes were stratified by age, gender and subsidy status for each sRHA. Key to the disease clustering technique is a simplified spatial relationship provided by a nearest neighbour (NN) matrix. For a sRHA, its first nearest neighbour is the sRHA located the shortest distance away, the second nearest neighbour is the sRHA within the next shortest distance, and so on. The distance between any two sRHAs determined the Euclidean distance—that distance "as the crow flies" between the centroids of the sRHAs. All analyses are based on sRHA of residence, although we recognize that individuals may seek care from EDs outside their sRHA of residence.

Table 1 provides demographic summaries for the population and cases. The analysis was conducted with 18 age groups based on the year of age (0, 1, 2, . . . , 17).

TABLE 1
Gender proportions, age medians and age interquartile ranges of cases of medically treated self-inflicted injury in the population of Albertans under 18 years of age

Pediatric population		**N = 785,079**
Gender	Males	402,394 (51.3%)
	Females	382,685 (48.7%)
Age (years)	Median	9
	Interquartile range	4 to 13
Pediatric cases		**N = 827**
Gender	Males	255 (30.8%)
	Females	572 (69.2%)
Age (years)	Median	16
	Interquartile range	14 to 17

Data collected between April 1, 1998 and March 31, 1999.

Statistical disease cluster detection method

We focus our description and analyses on Le et al.'s cluster-testing algorithm,[7] based on a test by Besag and Newell.[1] The Besag and Newell technique individually tests sRHAs to see if every person is equally likely to acquire the "disease" independently of other cases and the location of their residence. Since our focus is on clustering in the entire province, we chose a method that detects sRHAs with the tendency to cluster and also tests each sRHA. In addition, the method has

several appealing features, such as ease of understanding, straightforward covariate adjustment, overall testing capability, analogous focused test version and statistical appropriateness.

The method requires knowing the population and the number of cases in each sRHA as well as the nearest neighbour matrix. Stratification by important demographic variables, such as age and gender, is easily done. For sRHA i, let c_{iag} and n_{iag} denote the number of self-inflicted injuries and population, respectively, from group a and gender g, $a = 0,\ldots,17$, $g = 1,2$, $i = 1,\ldots,68$. The total number of self-inflicted injuries is $c_{\cdot ag} = \sum_{i=1}^{68} c_{iag}$ by age group and sex and similarly, the total population in the province by these categories is $n_{\cdot ag} = \sum_{i=1}^{68} n_{iag}$.

The method tests for a particular cluster size, k. The observed test statistic, ℓ, is the number of sRHAs that must be added to sRHA i in order to have at least k cases. The population of sRHA i with its ℓ nearest neighbours stratified by age group and gender is calculated, $m_{i\ell ag}$. In a rare disease, the significance level is approximated by a Poisson distribution with the p-value determined as

$$1 - \sum_{x=0}^{k-1} \exp(-\lambda_i)\lambda_i^x / x! \qquad (1)$$

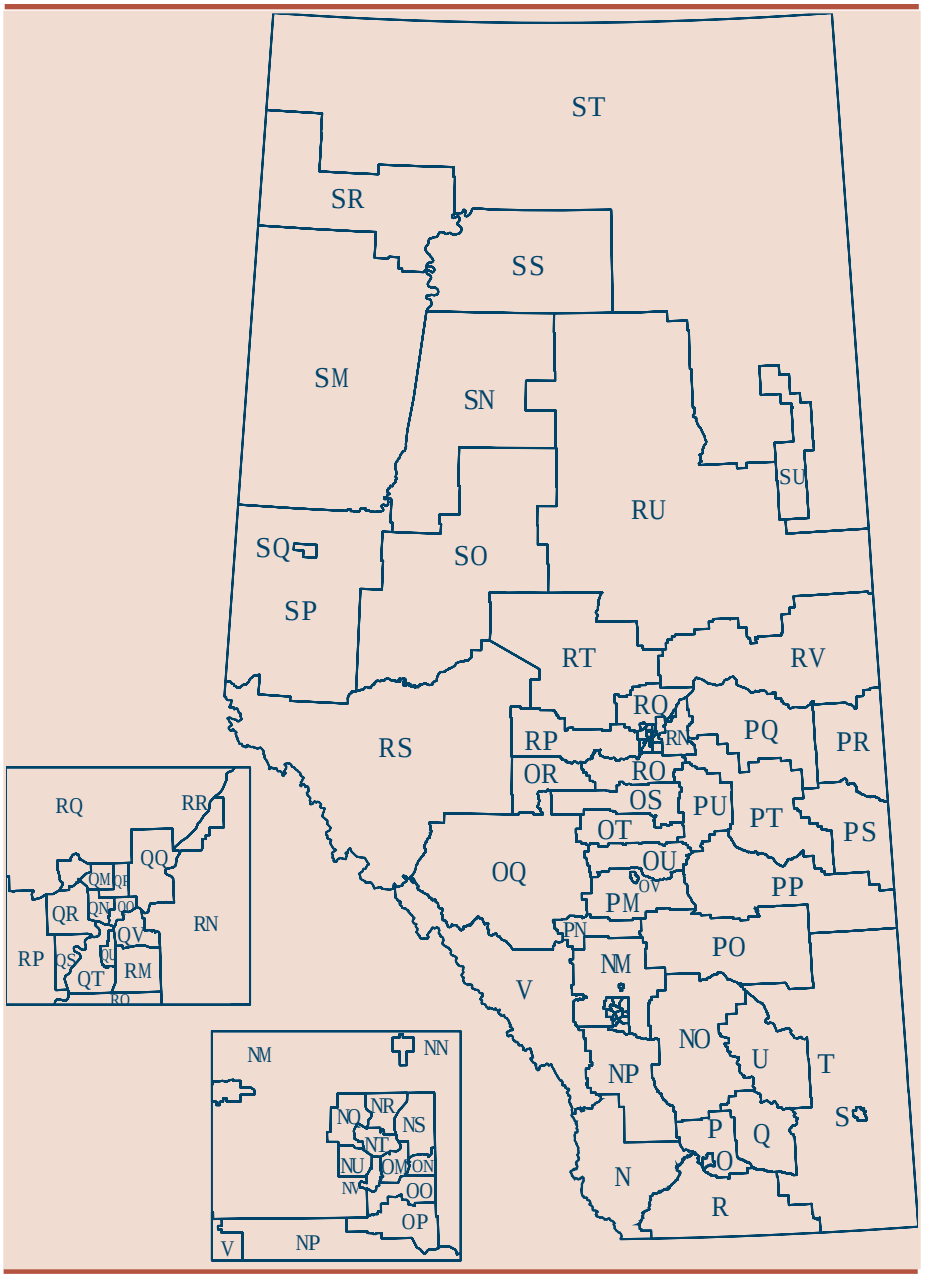
where $\lambda_{i\ell} = \sum_{a=0}^{17} \sum_{g=1}^{2} m_{i\ell ag} c_{\cdot ag} / n_{\cdot ag}$.

Small ℓ indicates that k cases are nearby and reflects the degree of clustering. This testing is done for each sRHA separately and statistically significant sRHAs are called "clusters".

Since population sizes differ substantially from one sRHA to another, one cluster size may not be appropriate for all sRHAs. Instead, the cluster size can be based on the sRHA's population and its nearest neighbours' population through a testing algorithm.[7] For sRHA i, the cluster size k_{i0} is chosen such that $k_{i0} - 1$ is the 95th percentile of the Poisson distribution with mean λ_{i0}. This cluster size can be interpreted as the minimum number of cases necessary to get a significant cluster at the five percent level, based on the population in sRHA i. If the test is insignificant, a new cluster size, k_{i1}, is chosen, similarly based on λ_{i1}. Hence, the algorithm is composed of a sequence of tests at cluster sizes k_{i0}, k_{i1}, k_{i2} ….

Practically speaking, some small number of tests can be chosen and if a cluster is not found at lower cluster sizes, larger cluster sizes can then be tested. In our application, we test at k_{iw} for $w = 0,1,2,3$ and refer to this approach as "modified Besag and Newell".

FIGURE 1
Alberta sub-Regional Health Authorities (sRHAs)



The clustering technique can also provide an overall assessment of clustering based on simulation. This aspect is particularly important when many tests are conducted in the testing algorithm. For the overall assessment, cases are simulated and allocated to sRHAs based on population proportions. The analyses are repeated on the simulated data in the same manner as was done for the original data and the number of clusters identified is recorded.

When case data are simulated many times, an overall clustering *p*-value is calculated, representing the number of simulations that had at least as many clusters as the original data.

Ethics approval

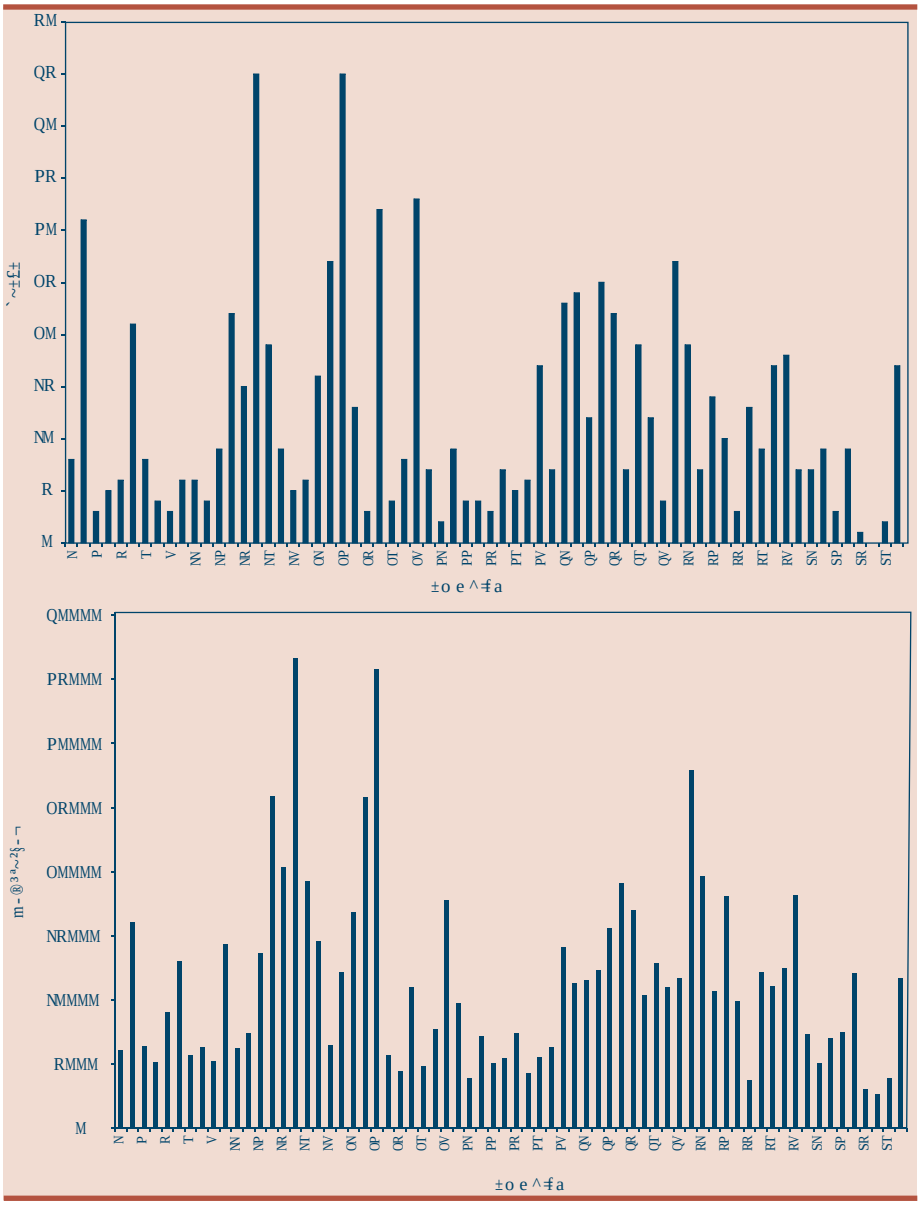
This project was part of an ED Atlas project, which examined a variety of diseases presenting to EDs over a number of years. The projects and analyses were approved by the University of Alberta Ethics Review Panel. No patient contacts were made and no informed consent was obtained. Patient identifiers were scrambled prior to off-site transfer to protect privacy and ensure anonymity.

Results

The results of the cluster detection tests for each sRHA are presented in Table 2. The Regional Health Authority names are provided as well as sRHA identification numbers (IDs) to ease discussion. For each sRHA, the results are presented for the cluster size kw, based on the population of the sRHA and its w nearest neighbours. The number of nearest neighbours, ℓ, that must be combined to have at least kw cases is presented as well as the IDs of the combined nearest neighbours (NNs). The actual number of cases in the sRHA and its ℓ nearest neighbours are denoted by o_ℓ and the expected number (and Poisson mean used for testing) is λ_ℓ. The ratio of the observed-to-expected numbers of cases are provided as a comparison. The *p*-value associated with each sRHA is listed and a double asterisk highlights $p < 0.05$. The simulation-based overall *p*-value associated with the number of clusters identified is $p_S = 0.007$.

The analysis identifies 15 sRHAs as clusters for individuals presenting with self-inflicted injuries. For Chinook 2 (R102) to be identified as a cluster, it needed at least 26 cases. With 31 cases in this sRHA, it qualifies as a significant cluster. Chinook 3 is identified as a cluster when it is combined with Chinook 2. Similarly, when these two sRHAs are combined with Chinook 4, this becomes a cluster. Pallisar 6 is a cluster on its own and when it is combined with

FIGURES 2a and 2b
Population and case counts of medically treated self-inflicted injuries in Albertans under 18 years of age for each sub-regional health authority (sRHA)



Data collected between April 1, 1998 and March 31, 1999.

Pallisar 7. David Thompson 24, 26 and 29 have enough cases to be clusters on their own. David Thompson 26 has 2.79 times more cases than expected. When David Thompson 28 and 30 are combined with David Thompson 29, they, too, become significant. East Central 38 needs at least 27 cases to be considered a cluster; 38 are observed when it is combined with its first nearest neighbour, David Thompson 26. David Thompson 27, when combined with its first two nearest neighbours, has 1.75 times more cases than expected by its age and gender distribution; it thus is identified as a cluster. Capital 41 and 42 are clusters on their own but Capital 45 needs to be combined with Capital 47 and 41 to be detected as a cluster. No other sRHAs had enough cases to be clusters by themselves nor when combined with their first, second or third NNs. Hence, $\ell > 3$ for these sRHAs and the associated combined population sizes are large enough for the *p*-values to be insignificant.

TABLE 2

Medically treated self-inflicted injury clustering results of Albertans under 18 years of age by regional health authority (RHA), stratified by age group and gender.* Nearest neighbour IDs (NNs) are provided for significant sRHAs.

RHA and sub-RHA ID number *i*		*w*	k_w	ℓ	NNs	o_ℓ	λ_ℓ	o_ℓ/λ_ℓ	*p*
Chinook	1	3	53	4		57	57.1	1.00	0.724
	2	0	26	0		31	17.6	1.76	0.036 **
	3	1	33	1	2	34	24.0	1.42	0.047 **
	4	2	39	2	3,2	39	29.3	1.33	0.050 **
	5	3	51	4		53	46.7	1.13	0.285
Pallisar	6	0	21	0		21	14.0	1.50	0.048 **
	7	1	29	1	6	29	20.3	1.43	0.041 **
	8	3	36	5		45	46.4	0.97	0.950
Calgary	9	3	66	5		73	90.3	0.81	0.997
	10	3	59	4		74	83.9	0.88	0.998
	11	3	99	4		107	110.7	0.97	0.878
	12	3	92	4		119	114.1	1.04	0.985
	13	3	104	5		108	114.3	0.95	0.843
	14	3	96	6		121	134.6	0.90	1.000
	15	3	121	6		121	134.6	0.90	0.889
	16	3	100	4		101	102.7	0.98	0.618
	17	3	78	6		121	134.6	0.90	1.000
	18	3	81	5		88	106.2	0.83	0.995
	19	3	73	5		82	94.0	0.87	0.989
	20	3	75	5		82	94.0	0.87	0.981
	21	3	110	5		118	117.6	1.00	0.770
	22	3	99	4		99	98.6	1.00	0.497
	21	3	99	4		99	98.6	1.00	0.497
David Thompson	24	0	12	0		13	6.3	2.06	0.028 **
	25	3	54	5		76	65.9	1.15	0.941
	26	0	18	0		32	11.5	2.79	0.045 **
	27	2	35	2	28,26	44	25.2	1.75	0.036 **
	28	1	37	1	29	41	27.2	1.51	0.043 **
	29	0	27	0		33	19.0	1.74	0.047 **
	30	1	39	1	29	40	29.2	1.37	0.047 **
	31	3	62	5		69	75.6	0.91	0.951
	32	3	49	7		71	78.0	0.91	1.000
	33	3	38	5		39	40.4	0.97	0.669
East Central	34	3	50	5		55	65.1	0.85	0.977
	35	3	34	4		37	43.4	0.85	0.939
	36	3	34	6		47	56.7	0.83	1.000
	37	3	25	6		61	49.6	1.23	0.988
	38	1	27	1	26	38	18.9	2.01	0.046 **

TABLE 2 (continued)
Medically treated self-inflicted injury clustering results of Albertans under 18 years of age by regional health authority (RHA), stratified by age group and gender.* Nearest neighbour IDs (NNs) are provided for significant sRHAs.

RHA and sub-RHA ID number *i*		*w*	k_w	ℓ	NNs	o_ℓ	λ_ℓ	o_ℓ/λ_ℓ	*p*
Capital	39	3	70	4		81	73.4	1.10	0.671
	40	3	70	4		83	67.7	1.23	0.406
	41	0	18	0		23	10.9	2.11	0.030 **
	42	0	19	0		24	11.8	2.04	0.032 **
	43	3	73	4		91	70.4	1.29	0.395
	44	3	84	4		84	80.0	1.05	0.340
	45	2	55	2	47,41	64	42.8	1.50	0.040 **
	46	3	68	4		83	65.4	1.27	0.390
	47	3	68	4		83	65.4	1.27	0.390
	48	3	81	4		86	78.3	1.10	0.397
	49	3	78	4		90	74.9	1.20	0.374
	50	3	81	4		86	78.3	1.10	0.397
	51	3	83	5		87	84.4	1.03	0.576
	52	3	79	6		98	107.1	0.92	0.998
	53	3	80	5		102	91.1	1.12	0.889
	54	3	69	4		71	74.9	0.95	0.769
	55	3	70	5		76	84.8	0.90	0.955
Aspen	56	3	41	4		47	43.0	1.09	0.642
	57	3	74	6		91	106.1	0.86	1.000
	58	3	58	5		64	70.7	0.91	0.946
	59	3	53	4		61	63.3	0.96	0.916
Peace	60	3	45	8		51	62.2	0.82	0.991
	61	3	44	8		47	62.2	0.76	0.994
	62	3	44	5		48	53.7	0.89	0.922
	63	3	47	5		48	53.7	0.89	0.838
	64	3	47	5		48	53.7	0.89	0.838
Northern Lights	65	3	21	5		26	29.0	0.90	0.948
	66	3	21	5		26	29.0	0.90	0.948
	67	3	21	5		34	35.1	0.90	0.996
	68	3	60	4		63	59.8	1.05	0.506

*p_s = 0.007 – The simulation-based overall *p*-value associated with number of identified clusters.

** $p < 0.05$

Data collected between April 1, 1998 and March 31, 1999.

Note that the significant *p*-values are generally close to 0.05 because of the testing algorithm used. The *p*-value is based on the cluster size tested and not the number of cases observed. For example, Chinook 2 was tested at a cluster size of 26 cases though 31 cases were actually observed. When tested at the cluster size of 26, the *p*-value is 0.036. Had a cluster size of 31 been tested, the test statistic would remain the same but the *p*-value would be 0.002. However, a larger cluster size of 31 could not be chosen *a priori* without knowing the actual number of cases in Chinook 2. The testing algorithm determines cluster sizes based on the population distribution and not the observed cases in an sRHA.

The cluster sizes were calculated to be the minimum value necessary to achieve significance at the 0.05 level. For Chinook 2,

the cluster size was calculated to be 26. Hence, all of the significant clusters have *p*-values close to 0.05. How close the *p*-value is to 0.05 depends on the discreteness of the Poisson distribution. A significant *p*-value needs to be interpreted with the observed to expected ratio to reflect the degree of clustering. For example, Chinook 3 (when combined with Chinook 2) and David Thompson 29 both have $p = 0.047$ with observed to expected ratios 1.42 and 1.74, respectively. The latter ratio would suggest more clustering than the former, even though the *p*-values are identical.

Additionally, the testing algorithm also does not allow a significant cluster to be tested at larger cluster sizes. Capital 41 is significant when tested at a cluster size determined by its population but is not tested at a cluster size determined by its population and its first nearest neighbour, its second nearest neighbour, and so on.

Since provincial self-inflicted injury rates are used in the *p*-value calculation through λ_ℓ, it is surprising that 15 of 68 sRHAs are identified as clusters. In 1000 simulated data sets, seven had at least 15 clusters identified ($p_s = 0.007$). These simulations suggest that it is unlikely to identify 15 clusters by chance alone.

Note that when sRHAs had to be combined with neighouring sRHAs to form clusters (Chinook 2,3; Pallisar 7; David Thompson 27,28,30; East Central 38; Capital 45), they were combined with sRHAs that were clusters by themselves. In particular, no sRHAs that were insignificant individually were significant when combined together. Such a situation can occur in practice and did occur during the simulations. However, since all of the clusters identified in our analysis were based on combinations of individually identified clusters (Chinook 2; Pallisar 6; David Thompson 24, 26, 29; Capital 41, 42), we examined the gender and age-adjusted rates for each sRHA (Table 3). Most of these individual clusters (Chinook 2; David Thompson 24, 26, 29; Capital 41, 42) had 95 percent confidence intervals[26] for adjusted rates above the provincial rate. However, the 95 percent confidence interval for the adjusted rate of Pallisar 6 did include the provincial rate. Thus, Pallisar 6 would not be identified as an area with significantly higher rates when examining only its adjusted confidence interval.

TABLE 3
Population, cases and gender-age adjusted rates of medically treated self-inflicted injuries per 1,000 pediatric population under 18 years of age for selected Alberta sub-Regional Health Authorities (sRHAs)*

RHA and sub-RHA ID number		Cases	Population	Adjusted rate	95% confidence interval
Chinook	2	31	16,055	1.83	1.24, 2.62 *
Pallisar	6	21	13,015	1.58	0.98, 2.43
Calgary	14	22	25,911	0.83	0.52, 1.26
	16	45	36,632	1.25	0.91, 1.68
	17	19	19,242	1.06	0.64, 1.66
	21	16	16,855	1.08	0.62, 1.77
	22	27	25,818	1.00	0.66, 1.47
	23	45	35,813	1.27	0.92, 1.70
David Thompson	24	13	5,671	2.16	1.15, 3.77 *
	26	32	10,989	2.95	2.01, 4.12 *
	29	33	17,732	1.83	1.26, 2.58 *
Capital	39	17	14,104	1.08	0.64, 1.78
	41	23	11,572	2.20	1.39, 3.32 *
	42	24	12,327	2.11	1.35, 3.18 *
	44	25	19,082	1.35	0.88, 2.01
	45	22	16,989	1.34	0.84, 2.04
	47	19	12,822	1.34	0.81, 2.14
	50	27	27,912	0.95	0.63, 1.39
	51	19	19,610	0.89	0.54,1.43
Aspen	58	17	12,445	1.43	0.83, 2.31
	59	18	18,178	1.02	0.61, 1.62
Northern Lights	68	17	11,674	1.38	0.81, 2.24
Province (combined)		**827**	**785,079**	**1.05**	

* The sRHAs presented have more than 15 cases or have rates significantly higher than the provincial rate.
Data collected between April 1, 1998 and March 31, 1999.

Discussion

We identified 15 clusters of sRHAs with statistically significant excess self-inflicted injury cases that could not be explained by differences in age and gender distribution. Simulations based on sRHA age and gender distributions suggested that these clusters are not likely chance occurrences. The diversity in Alberta's sRHA population sizes, as with other large regional jurisdictions, warranted a method that had the capability of incorporating such diverse sizes as well as the age- and gender-distribution differences within each sRHA.

These clusters may, in fact, represent areas of excess self-inflicted injuries or may be false clusters that are identified because of differing distributions of unmeasured variables key to the development of self-inflicted injury. We believe, however, that these clusters represent areas of excess self-inflicted injuries, and special intervention programs should be considered for them. Any variable whose distribution differs among sRHAs would provide a potential explanation of observed clustering. For example, it is known that some of the sRHAs forming clusters have populations with higher proportions of aboriginals, a group that has been identified as having higher rates of self-inflicted injury.[27] Another possibility might be differing regional distributions of income level.

There are several limitations to the analysis presented. The method has been generally applied to very small areas where several might have to be combined to identify a cluster of size 5, 10 or 15. The majority of sRHAs we dealt with were from relatively large geographic areas and the cluster detection method has a restricted ability to detect very small-sized clusters. On the other hand, the modified Besag and Newell approach is appealing because it removes the need to specify the cluster size *a priori* and enables areas to be combined for testing.

That being said, the appeal of the modified Besag and Newell approach is diminished slightly when, in analyses like ours, significant sRHAs that were not clusters on their own were only significant when combined with sRHAs that were. The combining and testing of different sRHAs prevents a straightforward, two-dimensional graphical presentation of self-inflicted injury clusters. The geographic areas can have relatively large populations and the cluster sizes can be larger than 100. The geographic areas were created by the provincial health agency and analyses based on sRHAs could change if the data were aggregated into different geographic areas. This possibility is referred to as the "modifiable areal unit problem"[28] and is a concern for any surveillance study. As such, researchers should avoid making inferences about individuals based on aggregate data.[29] Here, the analysis was simplified by our definition of a case as being an individual with at least one self-inflicted injury during the study period. The cluster detection method used by our study could not incorporate the possibility of multiple self-inflicted injuries per person. However, a method that could use such correlated data may identify other clusters where multiple self-inflicted injuries are occurring.

These cluster analyses are a relatively rapid and efficient exploratory method to statistically identify areas with excess cases. The modified Besag and Newell method requires minimal information on the cases and population within administrative areas, permits the detection of statistically significant clusters when population sizes are diverse, incorporates age- and gender-distribution differences, automates the cluster size choice and provides an assessment of global clustering. While there may be several explanations for the excesses detected, this analysis can be used to answer local reports and direct further investigations into identified sRHAs. Further analyses could be performed with other cluster detection methods on specific types of injuries within the definition of self-inflicted injury, and on smaller administrative areas, if available. Public health officials and health authority administrators could further use these results with their current knowledge of self-inflicted injury to more appropriately focus self-inflicted injury prevention interventions.

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The impact of a quit smoking contest on smoking behaviour in Ontario

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Abstract

Community-based smoking cessation initiatives target large numbers of people, are highly visible and have the potential for great impact. Ontario's Quit Smoking (2002) Contest was evaluated one year after its implementation to measure behaviour change among adult smokers participating in the contest. The registration database of 15,521 contest participants provided the basis for a random sample of 700 participants throughout Ontario who were contacted for a follow-up telephone survey. A total of 347 surveys were completed, of which 60 percent were women. Almost one third (31.4 percent) of the survey respondents reported that they had not smoked since the start of the contest. Participation in the contest also may have delayed relapse by as much as five months for 31.3 percent of respondents who resumed smoking. Older respondents, men, those who had previously attempted to quit and people who said their cessation "buddy" was helpful were more likely to stop smoking.

Key words: *cessation, contests, health behaviour, health promotion, incentives, smoking, tobacco control*

Introduction

Smoking is a significant, preventable cause of death and disease worldwide.[1] Smoking has been identified as a significant risk factor for many forms of cancer, cardiovascular diseases, respiratory diseases, diabetes mellitus and adverse pregnancy outcomes. In 2001, approximately one out of every four people over the age of 18 living in Ontario smoked daily or occasionally.[2] It is estimated that 15,969 Ontarians died in 1998 from a smoking-related disease.[1]

Smoking cessation is difficult, due to the addictive properties of nicotine, especially for longer-term smokers.[3,4,5] Different methods to quit smoking have evolved over the past few decades. Some of the more successful strategies include nicotine replacement therapies ("the patch", gum, inhalers and nasal sprays), smoking cessation programs that offer counseling and teach stress management, and other coping strategies including telephone counseling, prescription medications to help control cravings and self-help quitting methods. Quitting "cold turkey" (i.e., no intervention) has been described to be a successful strategy by some former smokers; however, only a very small percentage sustain cessation.[6,7,8] A "buddy" to provide social support has also been a successful method for smoking cessation.[9,10]

Evaluations of smoking cessation contests have shown how they facilitate the link between intentions to quit and a cessation attempt.[11,12] Post-contest quit rates between 10 and 35 percent have typically been demonstrated.[15,20,21,22] These "quit and win" contests originated in the United States. In the mid-1980s, Elder and colleagues implemented a lottery to stimulate participation in the "Up in Smoke" smoking-cessation initiatives.[13] The evaluation of the lottery revealed that, of the 103 enrollees, seven percent of smokers had quit at the three-month follow-up. Nevertheless, the Pawtucket Heart Health Program adopted the quit and win contest approach and, at one month, reported that one in five smokers had quit. Elder et al. concluded that lotteries were effective to recruit participants into community-wide smoking cessation programs. Lai and colleagues adapted a quit and win contest for implementation in a Vietnamese community in California. Of the 89 contest participants, 84.2 percent self-reported abstinence at six months.[14] The authors did not report any significant predictors of this high abstinence rate, but they concluded that quit and win contests can be adapted to different cultures successfully. Of 802 participants in a smoking cessation contest promoted to two million residents of San Diego County, California, 35 percent reported they were smoke-free at two months.[15] The authors reported that television promotion was the most effective medium, and contest enrollment was somewhat higher for those who received

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a promotional flyer compared to those who heard of the contest through other sources.

Incentive-based cessation programs have been adapted in several jurisdictions internationally. For example, in the fall of 1986, the North Karelia Project in Finland, in cooperation with Finnish national television, arranged a nation-wide, televised smoking cessation program and coincident contest, offered in eight parts. The results of a national survey[16] showed that 7.7 percent of those who smoked and had watched at least one of the eight parts of the program reported they had tried to quit smoking (the national rate was 7.5 percent). At six months, just over one in five persons who smoked and resided in North Karelia (22 percent) reported they had quit (17 percent nationally). In 1988, Sweden implemented a national quit and win contest that attracted 12, 840 participants.[17] A panel of 557 randomly selected participants responded to a self-report survey at 12 months post contest. One in five (21 percent) reported they had been tobacco-free for the entire year. Another nine percent had relapsed but quit again and were tobacco-free at 12 months. Sun and colleagues[18] evaluated the International Quit and Win '96 contests of China and Finland and compared results of the two countries (a total of 25 countries participated). Of those who participated in China (N = 13,848), more than one third (38 percent) had been abstinent at one year, compared to only12 percent of Finnish participants (N = 6,038 participants). China's comparatively better success at 12 months is attributed to greater measures to maintain cessation, compared to the Finnish maintenance initiatives. The authors suggested that countries with comparatively less experience in anti-smoking policy development programming will need different strategies to facilitate the implementation of smoking-cessation interventions.

Quit and win contests promote understanding of the different methods of smoking cessation identified above, including access to information and resources to encourage cessation, identifying a "buddy" and other social support mechanisms. These considerations informed the design of Ontario's Quit Smoking 2002 Contest (and subsequent contest planning). Smoking cessation rates were significantly better among individuals who had chosen to identify a support person compared to those who did not, according to Pirie and colleagues.[19] The relationship of the support person to the participant (e.g., family member, friend, relative, colleague) did not influence the probability of quitting smoking. Having a support person was particularly important for those people who had a spouse who smoked. This finding highlights the importance of contest provisions of incentives to smokers to identify a support person for their quit attempt. Bains and colleagues[20] reported on the evaluation of a contest in eastern Ontario. They measured the smoker's motivation to quit using the stages-of-change model. The authors determined that smokers who were in the action stage were six times more likely to have quit smoking than those in all other stages combined (although the significant test result was borderline). No other sociodemographic or smoking factors were predictive of cessation. A study of a quit and win contest in Quebec[21] demonstrated that persons who had successfully quit rated the social support they received from their buddy to be more useful than persons who had relapsed. These "buddies" included non-smoking family members, friends and colleagues. Overall, 72.4 percent of respondents indicated that social support was important in their quit attempt. The majority of the smokers (60 percent) chose to quit without the assistance of any aids other than the social support of the buddy. There was no difference in the perceived utility of the social support between those who did and those who did not use pharmacological aids. A community-based cessation program in Olmsted County, Minnesota reported that 11 percent of the 304 contest participants were abstinent at one year, based on a self-report survey.[22] They found that having a person enrolled in the contest to support the person who smoked, an absence of other people who smoked where they resided and educational attainment beyond high school were significant predictors of abstinence.

Ontario's third annual smoking cessation contest was launched in October 2002. Paid television and radio media were used to promote the contest provincially. In addition, community-based contest promotions utilized a variety of promotional strategies, including newspapers, billboards and direct mailings. Entrants registered by mail, fax and on the contest Web site. Eligibility for the contest was restricted to Ontario residents, 19 years of age or older, who were daily smokers. Contest registrants were required to sign up a non-smoking buddy, pledge to quit smoking for a minimum of four weeks (October 14th to November 11th) for a chance to win a prize. Potential winners were drawn randomly from the list of contest registrants. The potential winners participated in an interview followed by a urine test to measure cotinine in order to confirm their smoke-free status. In addition, the registrant's buddy was interviewed prior to the urine test. Winners also agreed to participate in the media announcement regarding the awarding of prizes. This public process discouraged false representation by contest participants. The purpose of this paper is to report on the results of the contest evaluation at 12 months post-registration. The objectives of the evaluation were to measure the potential impact of Ontario's 2002 Quit Smoking Contest at one year post-implementation; to measure the effectiveness of different strategies to promote the quit smoking contest; and to quantify contest participants' main smoking cessation concerns. .

Methods

The three evaluation objectives (program effect, promotion strategies and cessation issues) were measured using a telephone survey of randomly selected contest participants. Demographic and smoking behaviour data were captured on the contest registration form. The telephone survey methodology followed the design described by Dillman.[23] The self-report survey was modified from a pre-existing tool used in earlier evaluations of the Ontario contest. The modifications were based on input from the contest team and included modest changes in wording, the

addition of a seven-day point prevalence measure and a question to quantify quit attempts.[24] This revised survey was pilot tested among 11 randomly selected contest participants and was finalized with minor changes.

While many of the questions used in the survey were closed ended, some questions allowed the respondent to provide answers in her/his own words, which were reviewed and coded at the time of data entry. As part of the quality control procedure, all surveys were reviewed for completeness prior to data entry. Half of the surveys were extracted and verified to ensure accuracy in data entry. Analyses were conducted using SPSS version 11.5 for Windows.[25]

Five trained interviewers conducted the surveys during the period November 17 through December 18, 2003. Interviews took approximately ten minutes to complete. Interviewers were instructed to make at least four attempts on different occasions to contact a potential respondent identified on the contact list before abandoning the attempt. Periodic follow-up was scheduled with the interviewers to ensure consistency in interview delivery and data collection.

A power analysis was done during the study planning stage to anticipate the likelihood that the contest would yield a significant effect. The sample size was calculated based on cessation rates achieved in earlier Ontario contests and smoking cessation contests in other jurisdictions compared to Ontario's spontaneous quit rate, which was reported to be just under ten percent in 2001.[26] Quit rates from other contests were reviewed to establish one for the Ontario contest. Using a conservative approach, a 17 percent quit rate was selected, which was based on Finland's contest experience. In order to test the null hypothesis that the proportion positive in the population is 0.100, the criterion for significance (alpha) was set at 0.01. The test was two tailed—permitting interpretation of an effect in either direction. Therefore, using STATA software, and assuming a one-sample comparison of a proportion to hypothesized value, the estimated sample size of 321 contest participants gives 90 percent power to detect a difference between the proportion of 10 percent (spontaneous quit rate) and the alternative proportion of 17 percent.

Descriptive analysis was supplemented by a multivariate analysis using logistic regression to study the determinants of the likelihood or probability of a survey respondent quitting smoking. The dependent variable was set up as a binary (i.e., 0-1) dummy variable equal to 1 for those survey respondents who reported that they stopped smoking. It was set equal to 0 for those respondents who had not stopped smoking.

Based on examination of correlation coefficients and existing literature, a number of potential explanatory variables for smoking cessation available in the survey were selected and entered simultaneously for the regression modeling exercise. In order to compare smoking cessation across gender, the variable "male" was created and set equal to 1 for male survey respondents and otherwise set equal to 0. Hence, the reference category in this case is females. For education, "high school" was the reference category, and for the help provided by a buddy, "not at all helpful" was the reference category. For those respondents who had previously attempted to quit smoking, the reference group is those who had not made previous attempts. Similarly, for those who reported receiving support from others, their reference category was not receiving support from others. Finally, the two other potential determinants of smoking cessation modeled, "age" and "years smoking", are continuous variables whose impact was modeled as a linear function. Model estimation was performed with the STATA SE version 8 statistical analysis software package.

Results

Survey response rate

According to experiences of earlier Ontario quit smoking contest evaluations, approximately 50 percent of individuals contacted to participate in the evaluation would be expected to agree to participate in the telephone survey. A random sample of 700 contest participants was extracted from the registration database to ensure the required sample size (321 respondents). The sample of 700 was generated randomly using SPSS from the complete registrant database of 15,521 persons.

Of the original sample of 700, four individuals were dropped (one duplicate record, two individuals residing at the same address as two others were randomly removed, and one person was a "buddy"). There were 200 people who could not be reached due to incorrect telephone numbers (N = 107), who were away or deceased or had no recall of participation (N = 18), or, as per the protocol, who were still unreachable after four attempts (N = 75). Forty-seven individuals refused to participate in the survey.

A total of 348 interviews were completed and one survey was dropped from the analysis as less than 50 percent of the questions had a response, leaving a total of 347 completed interviews for analysis. The survey response rate was 58.3 percent. This calculation is based on the following formula: # completed surveys / [# completed surveys + # incomplete surveys + # refused + # attempted and not reached] = 348 / [348 + 1 + 47 + 200] = 0.583.

Respondent profile

The survey respondents were not statistically different from the individuals in the contest registrant database (see Table 1) in terms of age, sex, education and prior smoking behaviour (e.g., amount smoked daily and number of years smoking). Over half (60 percent) of the surveys were completed by women. The mean age of survey respondents was 42 years (median = 41 years) and half (49 percent) of the survey respondents had some form of post-secondary education (community college or university). With respect to smoking behaviour, 42.4 percent reported smoking more than 20 cigarettes per day prior to the contest and the mean number of years that they had smoked was 21 (median = 20 years).

TABLE 1
Comparison of Ontario 2002 Quit Smoking Contest registrants and survey respondents at 12 months post-registration by sex, age, education, and tobacco consumption and history

Characteristic	Registrants (N=14,824) %	Survey respondents (N=347) %	*p*-value
Sex			0.2807
Females	57.0	59.9	
Males	43.0	40.1	
Age			0.0570
< 30	24.1	18.4	
30 – 39	26.2	26.8	
40 – 49	27.4	31.2	
50 – 59	13.2	16.0	
60 – 69	3.9	2.3	
70 +	5.2	5.2	
Education			0.6100
High school	48.4	51.0	
Community college	35.7	33.4	
University	15.9	15.6	
Number of cigarettes smoked per day			0.1020
1 – 5	6.8	3.5	
6 – 10	15.5	16.7	
11 – 20	37.0	37.5	
20 +	40.7	42.4	
Years smoking			0.3680
< 5	5.6	4.3	
5 – 10	20.1	18.5	
11 – 15	14.7	12.1	
16 – 20	17.0	17.3	
21 – 25	13.3	15.8	
26 – 30	13.0	13.0	
31 – 35	6.8	9.2	
> 35	9.5	9.8	

The following findings are presented based on the three evaluation objectives: contest effect, promotion strategies and cessation concerns/issues.

Effect of contest

Three criteria were used to measure the effect of the contest:

1. the percentage of evaluation participants who self-reported they had stopped smoking;
2. the amount of time lapsed between smoking cessation and relapse; and,
3. the percentage of evaluation participants who self-reported they were smoking less than prior to the start of the contest.

Almost one third (31.4%; N = 109; 95% CI = 0.2650 – 0.3632) of the study participants reported they had not smoked since the start of the contest. This success rate is significantly higher than the 17 percent quit rate reported by Finland's contest experience (p = 0.00001). Of the 238 survey respondents who smoked since the contest started, eight percent said they were unable to quit during the contest. Another 9.7 percent said they had resumed smoking, but were not able to recall when they started smoking again. Of those who could recall when they resumed smoking (N = 196), 12.8 percent resumed smoking in October 2002, 20 percent resumed smoking in November 2002, another 20 percent resumed smoking in December 2002, 15.9 percent resumed smoking in January 2003, and almost one third (31.3 percent) resumed smoking in February 2003 or later.

We compared those survey respondents who quit smoking and those who continued to smoke according to sex, age, education, amount smoked and length of time smoked. We found no statistically significant differences across any of these strata.

Both those who stopped smoking and those who continued to smoke or relapsed were asked to describe the methods they used to try to stop smoking. Sixty-one percent of those who had continued to smoke or relapse had made at least one serious quit attempt since the end of the contest period. Non-intervention methods (e.g., quitting cold turkey or no method used) and intervention methods (e.g., nicotine patch or gum, anti-depressants) were reported. The data suggest that those who quit were somewhat more likely to use a non-intervention approach (e.g., cold turkey), compared to those who continued to smoke or relapsed and attempted to quit (51.4 vs. 40 percent). In comparing the two groups according to intervention strategies, those who continued to smoke or relapse reported using the patch to try to quit more often than those who were successful in quitting. This may suggest that those still struggling to quit are more likely to have perceived a need to try direct

interventions to help them stop smoking. Another possible explanation for the difference in types of techniques employed by quitters and continuers is that the continuing group may be heavier smokers.

Of the participants who were still smoking at the time of the interview, 49.8 percent (N = 100) reported they were smoking fewer cigarettes, compared to the amount smoked prior to the contest. Another 43.8 percent (N = 88) said they were smoking the same amount as they had smoked before participating in the contest, and only 13 people (6.5 percent) reported they were smoking more cigarettes than before the contest.

The survey also measured the extent to which those who continued to smoke were contemplating a quit attempt. Nearly two thirds (65 percent) said they planned to quit smoking in the next 30 days and 92 percent indicated that they planned to quit smoking within the following six months.

Promotional strategies

Ontario's Quit Smoking 2002 Contest was promoted through a wide variety of media, including television, newspapers, radio, brochures, posters and other strategies used by public health professionals at the community level throughout Ontario. Study participants were asked, without disclosing the types of strategies used by the contest, to recall which promotional method was most important in motivating them to enter the contest. Television, radio and newspapers were each equally effective (14 percent of respondents, respectively) as strategies to recruit people to participate in the contest. These promotional strategies were followed closely by "word-of-mouth/friends" (13 percent), flyers (9.2 percent) and the Internet (9.2 percent). About six percent of the respondents could not remember any specific promotional method.

Interestingly, more than half of the respondents (53.7 percent) said that the opportunity to win a prize by entering the contest was not at all or not very important in their decision to enter the contest. Only about one out of five (21 percent) of the study participants reported that the prize was important or very important in motivating them to enter the contest. In fact, nearly two thirds (64.3 percent) of respondents reported that they didn't know which prize was most appealing (the list of potential prizes was not read aloud to respondents during the interview). Of the prizes available, about one in five (21.6 percent) said the car was the most appealing. There was no significant relationship between outcome (i.e., smoking cessation) and the importance or interest in the prize offered (chi square(5) = 4.63, $p = 0.463$).

Cessation issues

Two thirds of the contest participants continued to smoke or relapsed after contest registration. Respondents were asked which factors influenced them to continue to smoke or resume smoking following the contest. The main reason cited for smoking was "stressful situations", according to one third of the respondents. Other factors included finding themselves in social situations where others were smoking (14 percent), the impact of withdrawal symptoms (12 percent), and for 6.4 percent of respondents, drinking alcohol was accompanied by smoking as part of their behavioural pattern.

All contest participants were required to sign up a buddy. Importantly, half (52 percent) said the buddy was helpful or very helpful in their efforts to stop smoking. In addition, there was no statistically significant difference in the perceived helpfulness of a buddy to stop smoking when comparing those who had quit with those who continued to smoke or relapsed.

Contest participants were also asked to describe any sources of social support they received in their efforts to stop smoking. Respondents could provide more than one answer. Nearly one quarter (23 percent) of the respondents said they did not receive any support from others. In contrast, 61 percent reported that family members provided support to quit smoking, and more than one third (38 percent) said that friends and co-workers supported them. About five percent of the study participants said health professionals provided them with support to quit smoking.

It is worth noting that 63 percent of the study participants reported that they sug-

TABLE 2
Likelihood of quitting smoking of Ontario 2002 Quit Smoking Contest survey respondents by age, sex, quitting history and perceived buddy helpfulness – Logistic regression model

	Odds ratio	Robust standard error	*p*-value
Age	1.0191**	0.0106	0.075
Sex *[Female]:*			
Male	1.5374**	0.2426	0.076
Tried quitting before *[No]:*			
Yes	2.0857**	0.4223	0.082
Helpful buddy *[Not at all helpful]*:			
Not very helpful	0.7889	0.4873	0.627
Somewhat helpful	1.4017	0.3979	0.396
Helpful	2.4099*	0.3810	0.021
Very helpful	1.2775	0.3449	0.484
Log likelihood	- 207.54		
Sample size	347		

* Significant at 5% level.

** Significant at 10% level.

The comparison group is shown in italics and square parentheses [ ].

gested entering the contest to other smokers. More than 9 out of 10 respondents (91 percent) said they would recommend this same type of contest to other smokers who are trying to stop smoking.

We learned from additional feedback made by study participants that they would prefer to have more follow-up, including additional information about available support, and the results of this and other cessation contests and programs. Communication with contest participants, therefore, is regarded highly. Apparently, more frequent communications and program content would add value as support to the participants.

Table 2 presents the results of the multivariate logistic regression model of the probability of quitting smoking. As noted earlier, the model included a number of potential explanatory variables collected by the survey, but in this table we present results for only those explanatory variables with $p < 0.10$. The likelihood of stopping smoking is positively related to the individual's age; older individuals are more likely to stop smoking than are younger ones (odds ratio = 1.019, $p = 0.075$). In addition, males are more likely than females to stop smoking (odds ratio = 1.537, $p = 0.076$). Individuals who previously attempted to quit smoking were more likely to stop smoking (odds ratio = 2.086, $p = 0.082$). Study participants who reported that their buddy was "helpful" were more likely to stop smoking, relative to those whose buddy was considered "not at all helpful" (odds ratio = 2.410, $p = 0.021$). Given the statistical significance levels of these results, some may regard these findings as only being suggestive. Other variables, such as education level and the length of time smoking, had no statistically significant impact on the likelihood of quitting smoking.

Discussion

Nearly one out of three contest participants who responded to the survey (31.4 percent) reported they had stopped smoking since the contest began and an additional 35 (14.8 percent) said they had since stopped smoking. Assuming the quit rate is the effect of the contest, the potential impact of this contest for Ontario's smoking population can be calculated. This is only an assumption and other assumptions are possible. For example, some may argue that attributing the full amount of the quit rate to the contest is excessive due to factors such "selection" or "volunteer" effects among contest registrants, which may bias the results. A more conservative approach, and the one we will adopt, is to use the lower bounds of the 95 percent confidence interval of the quit rate, 26.5 percent, rather than the 31.4 percent point estimate. The more than 15,000 participants who registered for the contest in 2002 represent about one percent of adult smokers in Ontario. By combining the registration rate with the lower bounds of the quit rate, it can be extrapolated that 1 in 377 adult smokers were motivated to quit because of the contest.

Nearly half of the people surveyed reported that the possibility of winning a prize did not influence their decision to enter the contest. Nevertheless, the contest itself represents an important opportunity for smokers who were contemplating quitting. The participants were motivated to enter the contest to quit smoking and improve their health. Winning a prize may be secondary to these motivations.

While the smoking cessation rate of 31.4 percent is within the range of quit rates reported in published studies evaluating incentive-based cessation programs, a large proportion of respondents were not successful in quitting. This underscores the challenges of smoking cessation among even highly motivated individuals, as Bains and colleagues reported in their evaluation of a more geographically limited quit smoking contest.[18]

The contest may have contributed to a delay in smoking relapse until February 2003 for nearly one third of the contest participants who had resumed smoking, although we were unable to find any comparable data on the rate at which quitters relapse in the absence of a smoking cessation program. This period of delay, approximately four months from the start of the contest, may be an important window of opportunity, in which follow-up reinforcement strategies could be introduced to sustain a quit attempt. However, it is noteworthy that a slightly higher proportion of those who continued to smoke or who relapsed, reported they smoked more than 20 cigarettes per day (although this is not statistically significant). Perhaps this group comprises a higher proportion of people who could be considered heavily addicted to tobacco.

More than four out of ten people surveyed who had continued to smoke or who relapsed during the contest reported they smoked fewer cigarettes after the contest than before it. Other studies of incentive

TABLE 3
Comparison of respondents to Canadian Tobacco Use Monitoring Survey (CTUMS) and Ontario 2002 Quit Smoking Contest survey respondents, aged 25 years and over, by sex and age

Characteristic	CTUMS (N=1,536,580) %	Survey respondents (N=310) %	*p*-value
Sex			0.00001
Females	46.1	59.7	
Males	53.9	40.3	
	(N=1,744,930) %	(N=345) %	
Age *(years)*			0.1040
20 – 24	13.1	10.1	
25 – 44	52.4	50.7	
45 +	34.5	39.1	

smoking cessation programs have demonstrated a similar reduction. A controlled study of a quit and win contest in Kentucky reported a statistically significant difference in the reduction of the number of cigarettes smoked in favour of participants in the treatment group.[28]

The results of this study are based on responses to a self-report survey. Biochemical verification of smoking cessation was not possible due to resource limitations. It is worth noting, however, that an expert work group formed by The Society for Research in Nicotine and Tobacco recently reported that biochemical validation is not necessary in the case of interventions for general populations of adult smokers.[29] In addition, population surveys have shown that self-report smoking status is generally accurate among smokers.[30,31,32,33]

It is important to note that the study participants in the evaluation of the Ontario Quit Smoking 2002 Contest were not incented (financially or otherwise) to complete a survey. Also, contest prizes were awarded and communicated to participants several months prior to the interview, and, as such, should not impact participants' responses. There is no evidence to suggest an adjustment to the self-report rate is required, when measuring smoking cessation in incentive-based programs. If there is a perception that an adjustment to self-reported quit rates is necessary for evaluations of smoking contests, no published studies were found to give direction in the amount of adjustment required.

The evaluation design did not include a formal control group due to resource limitations. Documents on surveys of smokers conducted in Ontario were used to compare the respondent sample to the samples in these surveys (e.g., Canadian Tobacco Use Monitoring Survey – [CTUMS], Canadian Community Health Survey [CCHS] and the Ontario Tobacco Research Unit [OTRU] monitoring reports). Some of these data were not broken down by province. In addition, many of the demographic and smoking behaviour variables were defined differently than those variables collected in the registration database (e.g., previous quit attempts, number of cigarettes smoked per day) and as such, opportunities for comparison were limited. It was possible to compare the respondents with data from the CTUMS[27] with respect to age and sex distribution. As Table 3 shows, the sex distribution of participants in the evaluation was significantly different than that of the general population of Ontario smokers, as reported in the CTUMS. However, there was no significant difference with respect to age between respondents in either survey. These findings are consistent with the evaluation of other quit smoking contests.

Finally, the timing of a quit smoking contest could be an important factor influencing smoking cessation. Contest participants suggested that contests offered late in a calendar year could be influenced by the fact that this time of year is particularly stressful (preparing for the holiday season), resulting in higher rates of relapse or delayed cessation. Also, it is possible that the high percentage of respondents who "planned" to quit in the near future may be the result of the timing of the evaluation. People participated in this evaluation during late November through mid-December. Perhaps they were "predicting" their resolutions to stop smoking to coincide with the onset of a new year.

Conclusions

The Ontario Quit Smoking 2002 Contest was successful in helping a significant proportion of smokers quit and remain abstinent at 12 months. In addition, the research revealed the contest may have contributed to delaying relapse by several months in individuals who resumed smoking. This was an unexpected subset of smokers who reported heavier daily smoking. Experience and data suggest this group requires more investigation to identify better strategies that would reinforce cessation. Overall, contests that reinforce multiple strategies and aids for smoking cessation, such as they exist in Ontario, can attract a large number of participants and are an effective public and community health intervention.

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Trends in mortality from ischemic heart disease in Canada, 1986–2000

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Abstract

This study examined trends in ischemic heart disease (IHD) mortality rates in Canada from 1986 to 2000, including analyses at the county level. The study population comprised Canadians aged 35 and over. Age-standardized mortality rates (ASMRs) were computed. Linear regression and Poisson regression were used to calculate average annual percentage change (AAPC) by age, sex, county and province. A substantial decrease in mortality rates was observed in those aged 35 and over for both sexes; the AAPC indicated a decline of 3.44 percent for males and 3.42 percent for females. The ASMRs were plotted for three time periods; the rates increased with each successive age group and decreased with each consecutive time period for both sexes. A significant decline in the IHD mortality rate was found in 47.2 percent and 46.9 percent of the counties among males and females, respectively; those counties had a statistically significant lower prevalence of daily smoking in both genders, and obese in females only. Only two counties showed a significant increase in the ASMRs of IHD in males and females, respectively. Enhanced prevention and control strategies should be considered to address IHD in countries where more modest decreases (or no decrease at all) in IHD mortality have been observed.

Key words: *Canada, ischemic heart disease, mortality*

Introduction

Cardiovascular disease is the leading cause of death in Canada, accounting for almost one third of all deaths each year. In 1982, 60 percent of the deaths attributed to cardiovascular disease (CVD) were caused by ischemic heart disease (IHD),[1] by 2003, this had fallen to 55 percent.[2] Coronary heart disease (CHD) mortality rates have decreased in many countries,[3–6] including Canada.[3,4] However, the mortality from IHD in Canadian counties has not been reported yet.

The purpose of this paper is to examine trends in the Canadian mortality rates of IHD from 1986 to 2000 and to address the decline in mortality at the national, provincial or territorial and county (census division) levels.

Methods

Data on mortality from IHD (ICD-9 codes 410–414 and ICD-10 in 2000) were provided by Statistics Canada from annual Canadian mortality files.[7] The data contained information on age, sex, and province and county of residence for the entire study period (1986–2000). Population counts were obtained from the Canadian Census.[8] Age-standardized mortality rates (ASMRs) were computed using the 1991 Canadian population as the standard.

The study population included Canadians aged 35 and over. ASMRs were computed at three levels of geographic aggregation: Canada; the provinces and territories (Newfoundland and Labrador, Prince Edward Island, Nova Scotia, New Brunswick, Quebec, Ontario, Manitoba, Saskatchewan, Alberta, British Columbia; Yukon Territory, Northwest Territories); and counties. We compared ASMRs calculated over a five-year period by sex for 1986–1990, 1991–1995 and 1996–2000.

Linear regression was used to determine the average annual percentage change (AAPC) in mortality rates by age, sex and province. Poisson regression was used to calculate the AAPC by sex and county. The AAPC values were computed by fitting a model that assumed a constant rate of change in the ASMRs. That is, a linear or Poisson model was applied to the ASMRs after logarithmic transformation. Because of the generally small number of deaths in each county, Poisson regression models (the log of the age-specific rates) were used. Ninety-five percent confidence intervals (CIs) for the AAPC were calculated by province only. Trends were investigated by ten-year age groups (35–44, 45–54, 55–64, 65–74, 75–84, 85+) and for the summary grouping (35+).

The Canadian Community Health Survey, Cycle 1.1 (CCHS in 2000–01) data were used to examine the difference between the AAPC and risk factors.[9] The present study was restricted to individuals aged over 35. The counties were categorized based on the quintile cutoff point defined by AAPC in IHD mortality. The prevalence of each factor among counties was averaged in each AAPC category.

For further analysis of risk factors in counties, the first through the fourth quintiles were grouped as group 1; the fifth quintile was group 2. *P*-values for differences between the two groups were calculated using CCHS bootstrap weights. A nor-

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mal distribution one-tailed test for higher proportions of risk factors in group 2 was performed. Three risk factors (i.e., daily smoking, obesity and low income) for IHD were selected from other Canadian studies. All statistical analyses were performed using SAS software.

Results

Figure 1 shows a substantial decrease in annual Canadian mortality rates of IHD among men and women aged 35 and over. The ASMRs fell from 581.1 per 100,000 in 1986 to 345.9 per 100,000 in 2000 among males, and from 299.3 per 100,000 in 1986 to 177.4 per 100, 000 in 2000 among females.

For the entire 1986-2000 period, the overall ASMRs of IHD for Canadians aged 35 and over were 439.0 per 100,000 among males and 226.8 per 100,000 among females (Table 1). The highest provincial rates during this period were found among men and women in Newfoundland and Labrador (531.3 per 100,000 and 292.2 per 100,000, respectively). The AAPC indicated a significant national decline of 3.4 percent for both genders between 1986 and 2000. On average, the significant decreases in rates were greater than 3.4 percent per year among men in Prince Edward Island, Nova Scotia, New Brunswick, Quebec, Ontario, British Columbia and Yukon Territories, and among women in Nova Scotia, New Brunswick, Ontario, Saskatchewan, British Columbia and Yukon Territories.

TABLE 1
Average annual percentage change in age-standardized mortality rates of ischemic heart disease by sex and province, ages 35 and over (Canada, 1986–2000)

	Males			Females		
Province/territory	Mortality rate per 100,000	AAPC	95% CI	Mortality rate per 100,000	AAPC	95% CI
Newfoundland & Labrador	531.25	-2.37	-3.20, -1.53	292.15	-2.70	-3.25, -2.00
Prince Edward Island	506.26	-3.53	-4.96, -2.08	245.59	-2.45	-3.74, -1.13
Nova Scotia	466.55	-3.70	-4.33, -3.07	224.22	-4.04	-4.52, -3.48
New Brunswick	436.07	-4.14	-4.92, -3.36	225.01	-3.83	-4.39, -3.34
Quebec	455.87	-3.56	-3.95, -3.16	230.60	-3.16	-3.99, -2.92
Ontario	458.9	-3.54	-3.94, -3.14	244.76	-3.48	-3.73, -3.19
Manitoba	440.53	-2.34	-2.73, -1.96	221.42	-2.63	-3.31, -1.90
Saskatchewan	405.53	-3.03	-3.63, -2.41	192.09	-3.47	-3.84, -2.77
Alberta	405.72	-2.68	-3.12, -2.24	208.44	-2.41	-2.65, -1.64
British Columbia	365.54	-3.66	-3.99, -3.33	183.62	-3.95	-4.23, -3.50
Yukon Territory	400.89	-8.14	-14.36, -1.48	183.76	-3.99	-8.56, 1.90
Northwest Territories	243.48	-0.65	-4.08, -2.90	92.93	0.76	-6.30, 8.28
Canada	438.99	-3.44	-3.74, -3.10	226.81	-3.42	-3.66, -3.18

AAPC = average annual percentage change.
CI = confidence interval.

FIGURE 1
Age-standardized* mortality rates (per 100,000) of ischemic heart disease by sex (Canada, 1986–2000)

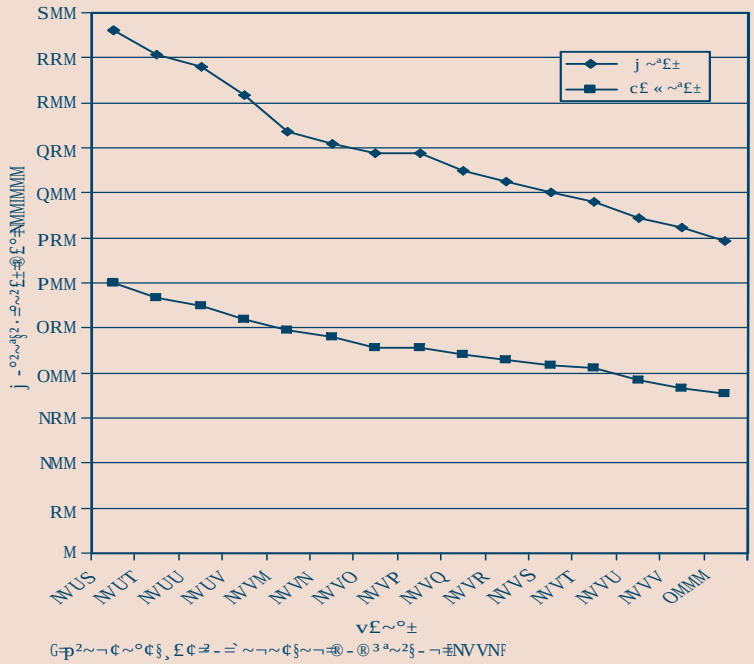
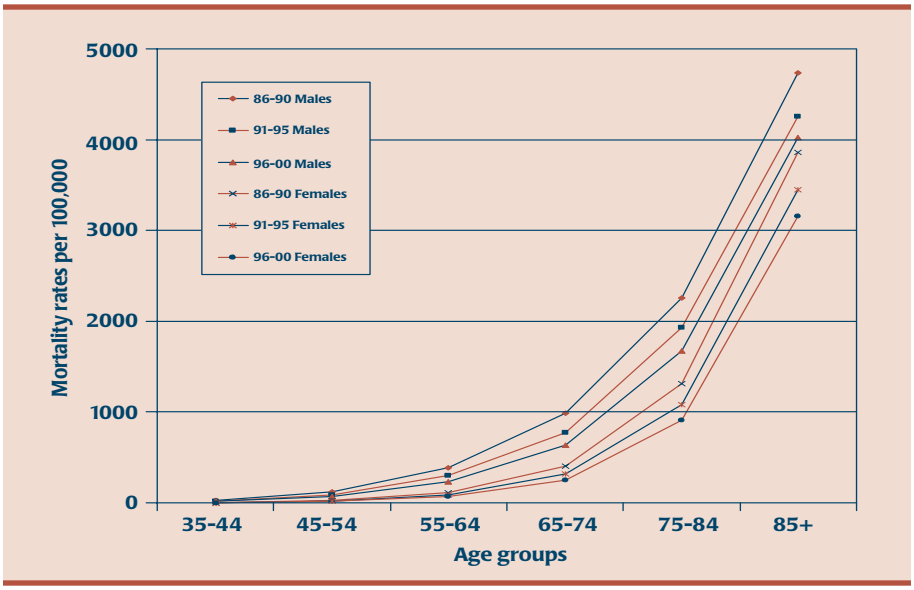


Table 2 presents the ASMRs and AAPC by sex and age for the complete study period (1986–2000). The ASMRs of IHD significantly increased with each age group among both sexes, particularly for those aged 75 and older. Once again, the high-

The IHD mortality rates within ten-year age groups were plotted for three time periods within the complete study period. For both sexes, the ASMRs increased with each successive age group. Mortality increased sharply for the 75-and-over age groups and the highest rates were observed among those aged 85 and over. On the other hand, the rates decreased with each consecutive time period. For each age group, the lowest ASMRs occurred in the most recent time period (1996–2000) among both men and women.

FIGURE 2
Age-standardized mortality rates (per 100,000) of ischemic heart disease by age, sex and five-year period (Canada, 1986–2000)



est rates were observed among those aged 85 and over (4,290.0 per 100,000 for males and 3,441.9 per 100, 000 for females). The AAPC showed a significant decline of 4.4 percent or more for males aged 35 to 74 and of 4.5 percent or more for females aged 45 to 74. In the 85-and-over age group, the average annual decreases were only 1.7 percent and 2.0 percent among men and women, respectively.

Figure 3 and 4 (not included here for technical reasons, but available by request) show maps of average annual percentage change in age-standardized mortality rates of IHD among males and females, aged 35 and over, by counties, between 1986–2000. The AAPC was categorized into three levels: the less-than-the-national AAPC (-15.149, -3.443); the less-than-0-and-larger-or-equal-to-the-national AAPC (-3.442, 0.000); and larger than 0 (0.001, 11.844) for males, and correspondingly for females (-33.884, -3.419), (-3.418, 0.000) and (0.001, 17.561). Each of the three categories was sub-divided into two sub-categories—significant and not significant—based on decreasing or increasing levels (i.e., $p \leq 0.05$). We used the 1996 census division boundaries.[10] The AAPC for four counties in females, Stikine Region, BC (5,957), Baffin Region, NWT (6,104), Keewatin Region, NWT (6,105), Kitikmeot Region, NWT (6,108) were not included in Figures 3 and 4 because of insufficient data.

For males, 47.2 percent of counties significantly decreased in IHD mortality relative to the national AAPC. The AAPC showed a significant decline of 6.5 percent or more in the following counties: Les Mitis, QC (2,411), L'île-d'Orleans, QC (2,420), La Haute-cote-Nord, QC (2,495), Skeena-Queen Charlotte Regional Di, BC (5,947), Yukon, YK (6,001), and Keewatin Region, NWT (6,105). There were 50.7 percent of counties whose AAPC did not reach the national level. Two percent of counties showed increased IHD mortality rates. In particular, the IHD mortality rate of Division No. 16, AB (4,816) increased significantly.

For females, 46.9 percent of counties showed a significant decrease in IHD mortality relative to national AAPC. The AAPC indicated a significant decline of 6.5 percent or more in La Matapedia, QC (2,407), Matane, QC (2,408), Matawinie, QC (2,462) and Division No. 10, MB (4,610). Forty-six (nine percent) of counties did not reach to the national AAPC and 4.1 percent of counties increased in IHD mortality. Specifically, one county, Division No.23, MB (4,623), significantly increased in IHD mortalit. Note that the majority of counties with the largest decreases in IHD rates (occurring in both sexes) were in Quebec. Detailed AAPC and age-standardized mortality rates in IHD by sex and county are obtainable by writing to the corresponding author of this article.

Table 3 shows the average prevalence of three characteristics/risk factors of counties, according to the AAPC distribution by quintile among males and females, respectively. For males, counties with a significant decline of IHD rates had a lower prevalence of daily smoking ($p < 0.0005$). For females, counties with significant decreases in IHD rates had a lower prevalence of obesity ($p = 0.0001$) and daily smoking ($p = 0.001$). No difference in

TABLE 2
Average annual percentage change in age-standardized mortality rates of ischemic heart disease by sex and age (Canada, 1986–2000)

Age groups	Males		Female	
	Mortality rate per 100,000	AAPC	Mortality rate per 100,000	AAPC
35-44	20.73	-4.59*	3.94	-1.26
45-54	90.28	-5.07*	19.06	-4.58*
55-64	302.84	-4.91*	88.66	-4.91*
65-74	783.88	-4.39*	319.15	-8.81*
75-84	1,918.92	-2.98*	1,081.31	-3.65*
85+	4,290.01	-1.68*	3,441.88	-2.04*
35+	438.98	-3.44*	226.81	-3.42*

AAPC = average annual percentage change.

* $p \leq 0.01$.

TABLE 3
Average prevalence of county characteristics/risk factors based on AAPC in age-standardized mortality of IHD by sex (Canada, 1986–2000)

Risk factor/ characteristic	Quintiles (AAPC) I (decreasing)	II	III	IV	V (increasing)	*p*-value[1]
Males	≤ -4.0708 *(N=56)*	> -4.0708 ≤ -3.735 *(N=57)*	> -3.735 ≤ -2.949 *(N=58)*	> -2.949 ≤ -1.964 *(N=57)*	> -1.964 *(N=57)*	
Daily smoking (%)	23.7	22.0	23.3	24.7	26.7	< 0.0005
Obesity (%)	17.3	16.9	16.5	17.8	20.9	0.44
Low income rate (%)	6.7	8.9	9.1	7.8	9.2	0.26
Females	≤ -4.581 *(N=56)*	> -4.581 ≤ -3.66 *(N=56)*	> -3.66 ≤ -2.97 *(N=56)*	> -2.97 ≤ -2.111 *(N=57)*	> -2.111 *(N=57)*	
Daily smoking (%)	19.6	17.9	18.6	21.7	21.8	< 0.001
Obesity (%)	15.9	18.5	16.3	20.1	20.3	< 0.0001
Low income rate (%)	12.9	12.2	13.9	13.8	13.7	0.92

AAPC = Annual average percent change.

Note: The data from three counties (area codes 2120, 2444 and 4623) in males and two counties (area codes 2420 and 2444) in females were not available for analysis.

[1] *p*-value measures the probability that quintiles have different proportions of risk factors.

low-income rates were observed in females or males. A detailed distribution of the AAPC in IHD by quintile and province is shown in Appendix 1.

Discussion

Our results show that mortality rates of IHD in Canada decreased on average by 3.4 percent per year for both males and females between 1986 and 2000. Significant decreases were also observed within each ten-year age group. Almost one half of all counties experienced significant declines in the IHD mortality rate. However, two counties had significantly increased rates among males and females.

Mortality from IHD has been declining steadily worldwide over the past several decades.[3,5,6] The results from the WHO MONICA project indicated that two thirds of the decline in CHD mortality may be attributed to decreases in coronary event attack rates, and one third to decreasing trends in case fatality.[11] Other studies also have reported declining incidence of acute myocardial infarction as well as improved secondary prevention, treatment and survival.[12,13] It is estimated that 25 percent of the decline in CHD mortality in the United States between 1980 and 1990 was due to efforts in primary prevention, 29 percent to secondary prevention and 43 percent to improvements in treatment.[14] A twenty-year population-based study indicated that the decline was significantly greater for in-hopital CHD deaths than for sudden cardiac deaths.[15]

The decreases in IHD rates which we found reflect those reported in other Canadian studies;[4,16] mortality reductions appear to be due to a decreasing incidence of acute myocardial infarction (AMI) and to improvement in cardiovascular treatment and care in Canada.[4,17] Canadian and provincial/teritorial utilization of evidence-based medications for treatment of AMI increased in 1999-2002.[18] Improvements in patient survival after AMI have been reported in British Columbia,[19] Alberta[20] and Ontario.[21]

The decline in IHD mortality can also be attributed to a reduction in risk factors. Studies from Great Britain, the United States and Australia have estimated that 30 to 75 percent of the observed decline in death rates from IHD was related to changes in lifestyle, which reduced the three major risk factors—hypertension, cigarette smoking and diet.[22,23] However, a recent follow-up study to evaluate the association of risk-factor time trends with CHD declines in Israeli males indicated that traditional risk factors appeared to play a limited role in the declining rates of CHD mortality.[24] In Canada, another follow-up study indicated that community and hospital factors explained no more than seven percent of the variation in the risk-adjusted outcomes across hospitals or regions.[25] Finally, a cross-sectional study in Scotland reported that conventional IHD risk factors do not explain the comparatively high rate of IHD.[26]

Cigarette smoking is one of the major risk factors for the development of CHD and is the major cause of preventable death in Canada. Results from the Canadian Community Health Survey, Cycle 1.1 (CCHS in 2000–01) indicated that health regions with high IHD mortality had a statistically significant higher prevalence of daily smoking.[27] Our results also indicated that counties with smaller decreases in IHD mortality had a significantly higher prevalence of daily smoking in both men and women. The proportion of Canadians aged 15 years or older that smoke cigarettes on a daily basis has fallen from 39 percent in 1977 to 24 percent in 1996–97.[28]

Our results show that counties with more modest decreases in IHD mortality rates

also had a higher proportion of their populations classified as low income. Socio-economic status has been demonstrated in previous studies to have a pronounced effect on access to specialized cardiac services. In subjects living in low-income areas In Ontario, there have been sharply higher mortality rates observed one year subsequent to hospitalization for AMI.[29] In another study, health regions with high IHD mortality in Canada were shown to be associated with a low-income rate and a lower prevalence of post secondary education.[27] Low socio-economic status can also exert a strong adverse influence on cardiovascular risk factors—for example, smoking habits, cholesterol level and blood pressure.[30] A large American cohort study showed that the risk of death from CVD and other disease for both men and women increased proportionately through a classification range of moderate to severe overweight.[31] In Canada, obesity rates have increased over the last decade.[32]

As for hypertension, the CCHS (2000–01) self-reported prevalence in Canada is 13.0 percent.[33] Filate et al. showed that regions in Canada with high IHD mortality also had a statistically significant higher prevalence of high blood pressure.[27]

Our results should be interpreted with caution. Population lifestyle changes and improvements in treatment and care may have contributed to the markedly declining mortality from IHD in Canada during the period 1986–2000 and it is possible that observed differential AAPC results can be explained by changes in risk factors. However, it must be admitted that there was not sufficient risk factor data to fully evaluate the association of risk factor longitudinal trends with IHD decline; we used only a single data point from a relatively recent source (CCHS, 2000–01). Thus our results indicate avenues for further research. Moreover, some of our observed differences in mortality at the county level may be the result of chance alone, particularly when a large number of comparisons are made.

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Appendix 1

Distribution of AAPC in ischemic heart disease by quintile and province, ages 35 and over (Canada, 1986–2000)

Province/ territory	Quintile I (decreasing)		II		III		IV		V (increasing)		Total
	N	%	N	%	N	%	N	%	N	%	
Males											
Newfoundland & Labrador	0	0.0	0	0.0	1	10.0	4	40.0	5	50.0	**10**
Prince Edward Island	1	33.3	0	0.0	1	33.3	1	33.3	0	0.0	**3**
Nova Scotia	2	11.1	5	27.8	6	33.3	3	16.7	2	11.1	**18**
New Brunswick	4	26.7	7	46.7	2	13.3	1	6.7	1	6.7	**15**
Quebec	30	30.3	24	24.2	17	17.2	12	12.1	16	16.2	**99**
Ontario	4	8.2	11	22.4	14	28.6	14	28.6	6	12.2	**49**
Manitoba	4	17.4	1	4.3	2	8.7	7	30.4	9	39.1	**23**
Saskatchewan	5	27.8	2	11.1	3	16.7	5	27.8	3	16.7	**18**
Alberta	1	5.3	4	21.1	4	21.1	4	21.1	6	31.6	**19**
British Columbia	5	17.9	3	10.7	8	28.6	6	21.4	6	21.4	**28**
Yukon	1	100.0	0	0.0	0	0.0	0	0.0	0	0.0	**1**
Northwest Territories	1	20.0	0	0.0	0	0.0	0	0.0	4	80.0	**5**
Total	**58**		**58**		**58**		**57**		**58**		**288**
Females											
Newfoundland & Labrador	0	0.0	1	10.0	1	10.0	5	50.0	3	30.0	**10**
Prince Edward Island	0	0.0	0	0.0	1	33.3	1	33.3	1	33.3	**3**
Nova Scotia	6	33.3	6	33.3	2	11.1	3	16.7	1	5.6	**18**
New Brunswick	3	20.0	5	33.3	3	20.0	2	13.3	2	13.3	**15**
Quebec	26	26.3	19	19.2	18	18.2	16	16.2	20	20.2	**99**
Ontario	5	10.2	13	26.5	14	28.6	9	18.4	8	16.3	**49**
Manitoba	7	30.4	1	4.3	5	21.7	2	8.7	8	34.8	**23**
Saskatchewan	4	22.2	5	27.8	2	11.1	6	33.3	1	5.6	**18**
Alberta	1	5.3	2	10.5	2	10.5	4	21.1	10	52.6	**19**
British Columbia	4	14.8	4	14.8	8	29.6	8	29.6	3	11.1	**27**
Yukon	0	0.0	1	100.0	0	0.0	0	0.0	0	0.0	**1**
Northwest Territories	0	0.0	0	0.0	0	0.0	1	50.0	1	50.0	**2**
Total	**56**		**57**		**56**		**57**		**58**		**284**

AAPC = average annual percentage change

Note: Four counties were removed because of insufficient data for females.

Seasonality of SIDS in Canada

Dear Editor,

Rusen et al.[1] recently claimed to have detected a change in the seasonal pattern of SIDS deaths in Canada, following a recommendation that infants should be placed in a supine sleeping position instead of prone. Their conclusion is based on a comparison of the association of cause of post-neonatal deaths (SIDS versus non-SIDS) and season (four quarters of the year) in 1985-89 vs. 1994–98. Rusen's interpretation was challenged in a new analysis by Mage,[2] who concluded that there had been no change in the seasonal pattern of SIDS.

In my opinion, the analyses by Rusen and Mage are both problematic because they do not address the most relevant epidemiologic question, which is whether the seasonal pattern of SIDS has changed over time in comparison to non-SIDS. Also, there remain methodological issues, which are outlined below.

Rusen's position is based on the fact that the association between season and cause of death is statistically significant in 1985-89, whereas in 1994-98 it is not significant. Unfortunately, I believe that type of comparison is invalid, because statistical significance is influenced not only by the strength of the association under study, but also by the sample sizes involved. There are considerably more deaths in 1985-89 than in 1994-98 (in fact, 61 percent more). Therefore, even if the seasonal pattern had remained exactly the same, we would expect to find a smaller *p*-value in the earlier time period—and that is what was actually observed. Because of the artifact of sample size, it is invalid to infer possible changes in seasonality from a comparison of *p*-values in this way.

Mage's own criticism of Rusen's position is actually based on somewhat different concerns. Mage correctly points out that Rusen's analysis does not adequately test the change in seasonal pattern because there could have been changes in either or both of the non-SIDS and SIDS death rates. In particular, it is possible that the non-SIDS seasonal distribution has changed while the pattern of SIDS deaths has not. Mage's own analysis compared the frequencies of SIDS deaths by season in the two time periods, ignoring the data from non-SIDS deaths. He also examined the fit of SIDS frequencies to predictions from a model based on external data. Non-significance occurred in the first comparison and non-significant deviations were found between the observed and predicted frequencies for both time periods. Mage therefore concluded that there has been no change in the seasonal pattern. (Even after Mage's criticism was published, Rusen et al.[3] re-iterated and continued to defend their method of analysis and the original interpretation).

By electing to analyse data only from SIDS deaths, Mage ignores the possibility of seasonal changes in infant mortality more generally. If such changes had happened, it would then be inappropriate to attribute any change in the seasonal pattern of SIDS to factors such as the recommendation on infant sleeping position.

Mage argues that the non-significant deviations of the SIDS frequencies from model predictions in each time period are evidence that the same seasonal pattern applies to both periods. In my opinion, this approach again falls into the trap of implicit comparisons of *p*-values and it does not provide any *direct* evaluation of possible differences in the seasonal pattern between periods.

A re-analysis

I feel that it is more appropriate to study the proportion of all deaths that are due to SIDS and to take the overall effects of seasons into account. Specifically, we should determine if the seasonal pattern of SIDS deaths has changed, relative to the pattern of non-SIDS deaths. To achieve this objective, I fitted a log-linear model to the data (as shown in Table 1), including the following: main effects for season, time period and cause of death; three two-factor interactions between these three variables; and one three-factor interaction.

The main effects take into account the unequal numbers of deaths observed in the various levels of each factor. For example, the main effect of period allows for the clearly larger number of deaths in 1985–89 compared to 1994–98. The two-factor interactions represent associations between pairs of variables. For instance, the interaction between season and period reflects the overall changes in the seasonality of post-neonatal mortality, but without regard to specific causes of death. Finally, the three-factor interaction examines whether the seasonal pattern of the SIDS to non-SIDS ratio has changed over time, which is the effect of primary interest here.

An informal examination of the data reveals that there are considerably more deaths in the earlier time period in both the SIDS and non-SIDS categories. Overall mortality appears slightly lower in the summer during both time periods. The percentage of deaths due to SIDS appears slightly higher in the winter and spring, and lower in the more recent time period.

A likelihood ratio test for the three-factor interaction was not significant ($p = 0.93$), indicating that there has been no meaningful change in the seasonal pattern of SIDS over time, compared to other causes of death. The two-factor associations of period and season with cause of death were both significant ($p = 0.02$ and $p = 0.02$), suggesting that the proportion of deaths from SIDS fell significantly over time (36 percent in 1985–89 vs. 31 per-

cent in 1994–98), and there was a relative excess of SIDS during the winter and spring. The association of period with season was not significant ($p = 0.13$), giving only weak evidence that the seasonal variation in overall mortality has changed (in the direction of less pronounced seasonal differences in 1994–98).

In summary, I conclude that although the proportion of SIDS deaths has varied between periods and between seasons, there has been no change in the seasonal pattern of SIDS deaths over time, compared to the changes experienced in overall post-neonatal mortality risk. This contradicts Rusen's original conclusion, but agrees with Mage (although for different reasons). My analytic approach has the advantage of taking other changes in post-neonatal mortality risk into account, whereas Mage's neglects data on other causes of death entirely. Although my conclusion qualitatively agrees with Mage in these particular data, in general I believe it is preferable to take the appropriate epidemiologic denominators (in this example, total deaths) into account.

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TABLE 1
Post-neonatal SIDS and non-SIDS deaths in Canada, 1985–89 and 1994–98

	Cause of death, 1985–89			Cause of death, 1994–98		
Season	SIDS	Non-SIDS	Total	SIDS	Non-SIDS	Total
Winter (Jan-Mar)	331 (36%*)	590 (64%)	921 (28%)+	173 (33%*)	352 (67%)	525 (25%)+
Spring (Apr-Jun)	283 (36%)	510 (64%)	793 (24%)	169 (31%)	371 (69%)	540 (26%)
Summer (Jul-Sep)	229 (31%)	502 (69%)	731 (22%)	139 (30%)	330 (70%)	469 (23%)
Autumn (Oct-Dec)	282 (31%)	622 (69%)	904 (27%)	155 (28%)	390 (72%)	545 (26%)
Total	1,125 (36%)	2,224 (64%)	3,349 (100%)	636 (31%)	1,443 (69%)	2,079 (100%)

* Percentage of all deaths that are due to SIDS, by season.

+ Percentage indicates seasonal distribution of all deaths.

Workshop Report

An invitation to develop Ontario's cancer research platform: Report of the Ontario Cancer Cohort Workshop

Fredrick D Ashbury, Victoria A Kirsh, Nancy Kreiger, Scott T Leatherdale and John R McLaughlin

Introduction

An emerging commitment among the Canadian government and funding agencies, and the scientific and clinical communities to reduce the growing cancer burden will enable enhancement in etiologic discovery, knowledge translation and control of cancer. Toward this end, the value of a large, longitudinal cancer research platform has become well recognized. Such a population-based and integrative program enables a wide range of innovative and high impact studies that relate to cancer prevention, early detection and outcomes. A group of multidisciplinary, experienced scientists and public health professionals (Appendix 1) met in a workshop on March 20-21, 2006 to take the first steps towards developing the research program.

The workshop goals were 1) to begin to develop the vision, direction and strategies for an integrated cancer research plan for Ontario; 2) to establish a transdisciplinary team of accomplished researchers from across Ontario to develop the research plan; 3) to determine the structures and processes needed to design a program; 4) to define the required developmental work; 5) to understand the available methods and the ones which will need to be developed; and 6) in broad terms, to identify the content of pilot and feasibility studies.

Participants were sent a draft proposal for a two-year research initiative (i.e., as the first phase of a large-scale cancer research plan for Ontario). An overview of key cancer cohort initiatives and selected readings[1-4] were also provided to participants.

Dr. Robert Hiatt gave the keynote address, "Cohorts: A vehicle for transdisciplinary science", followed by a moderated panel discussion to identify key considerations in developing an Ontario plan. Facilitated breakout groups organized by themes—community, epidemiology, genomics and health services—addressed specific questions related to research planning. Dr. John Potter presented "Towards the last cohort" and an all-delegate discussion focused on key considerations for the research plan, opportunities and recommendations.

Discussion

Dr. Hiatt discussed the international cancer research agenda and opportunities for enhancing large-scale science as well as identifing existing cohorts and discussing the role of cancer consortia. He reflected on the importance of transdiciplinary thinking and presented a research framework to address determinants of health and health disparities.

Dr. Hiatt stated that trandisciplinary science was relevant because a large population cohort is a platform for answering many different questions encompassing multiple disciplines; because the evidence derived from a major cohort study seeks application to a variety of settings; because knowledge exchange and uptake will require buy-in by organizations, governments and systems, and; finally, because a cohort study requires broader scope in planning and broad-based support. While a transdisciplinary approach may present obstacles— requiring larger financial commitment, logistical constraints, issues around tenure and merit review—this approach may contribute effectively to reduce the burden of cancer.

Dr. Potter's presentation focused on genetic susceptibilities and environmental exposures in cancer etiology. Rates of cancer vary 10- to 200-fold across different geographic locations around the world, and over 50 years we have seen up to ten-fold differences in rates within a geographic location. These differences can only be explained by differences in environmental exposures or interactions between genes and exposures.

Soon we will be able to characterize genotypes and haplotypes with increasing precision and efficiency, and we should be collecting high-quality information about both people and their exposure histories, biospecimens and information about disease outcomes. Also, better molecular characterization of cancer subtypes allows

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findings to be related to exposure, genetics and therapeutic responses. Homogeneous disease subsets may be associated with particular exposures and may carry different responses to therapy and prognoses.

Dr. Potter concluded that the time has come to establish new cohorts, possibly undertaken as an international collaboration. The danger is having the right technology in place to do high through-put genome sequencing and proteomics on very large populations, but without the requisite study infrastructure to best exploit those gains.

Following the speakers' addresses, delegates contemplated several important questions:

Do we build on existing studies or develop a new cohort?

Building on existing cohorts means limiting the breadth and depth of data to the lowest common denominators. In addition, many extant cohorts have limited ethno-racial heterogeneity, limiting potentially relevant studies of genetic variation or variation in environmental exposures. The age of existing cohort members limits questions; many of the cohorts have older-aged participants and outcomes of interest may already have occurred.

In contrast, while developing a new cohort requires substantial resources, it permits the greatest opportunity to address important research questions. Improved measurement, multigenerational approaches and a focus on younger people could ultimately lead to unique preventive interventions to reduce disease rates and yield savings in health care costs.

What is the added value of an Ontario-based cohort study?

Ontario has many advantages for hosting such an initiative, including the existence of a high-quality cancer registry, substantial record-linkage capability, technological advances in data collection and retrieval, and access to administrative databases that will assist in the detection of outcomes and patterns of health services utilization in the population. Blending an etiological and discovery focus with policy studies that have public health implications will advance such a cohort to the forefront of fostering new discoveries and applying knowledge to the Ontario population. Its value to government and health system decision makers would thereby be enhanced.

How important is it to incorporate uniform procedures and measures across studies?

Uniformity allows data sharing and consortium development that extends beyond Ontario, and increases utility in answering questions of global import. Uniform procedures of tissue collection may be more important than uniform questionnaire and other self-reported exposure data, since these latter must vary to cover the exposures relevant to the population under study (e.g., food frequency questionnaires which need to be tailored to the foods actually available).

Should the project be limited to cancer-relevant data or should it include data for understanding other diseases as well?

A strong consensus emerged regarding the need for breadth in data collection, to allow for the eventual analysis of disease outcomes in addition to cancer. Many of the biological processes (e.g., cell proliferation, inflammation, apoptosis, cell signaling) have relevance to many diseases. In addition, exposures of interest are common across the major diseases, or could be collected at little marginal cost once a cohort is assembled. Finally, interest in funding such an endeavour may be increased with broader scope and cohort participants may be more readily recruited.

Will the cohort support population intervention studies?

Incorporating analyses of natural experiments that might occur during the life of the cohort (e.g., initiatives from the public health units, local smoking cessation programs) was discussed. The drawbacks of evaluating direct interventions relate to data collection biases and potential increased respondent burden.

How does this cohort project map onto needs of future funders?

The leadership and governance of the program should include a scientific and a health impact focus. In the short term, there are two complementary directions: to identify leaders who can interact with or represent stakeholders, and who can negotiate with potential funders; and to identify leaders to lead the development of the overall platform and subsequent research proposals. These two groups of leaders are not mutually exclusive and must fully engage with each other.

Recommendations

The workshop strongly recommended a large-scale, longitudinal cohort that serves as an integrated cancer research platform in Ontario. A strong multidisciplinary team of experienced researchers from across Ontario supported the concept and is willing to collaborate to establish this major initiative. The workshop also underscored that transdisciplinary science is the key to leveraging maximum value from a cohort enterprise. This means inclusion of—and meaningful interaction between—scientists and public health leaders with multiple disciplinary backgrounds, working in the context of a population health laboratory. Substantial enthusiasm and commitment exist within the community of science and public health practitioners; the main challenges are sustaining required levels of interest and funding as developmental work evolves.

Developmental and pilot studies and preliminary explorations are needed to fill knowledge gaps and to demonstrate the optimal ways to proceed. Preliminary activities will relate to,

- environmental scans of major research platforms in order to identify organizational and governance structures which yield streamlined management with maximal efficiencies and participation, as well as maximal data utilization;
- methods for recruitment and retention of minority and often marginalized populations, including Ontario's aboriginal

communities, non-English speaking immigrants and homeless people;

- literature reviews to identify optimal methods for collecting information on behavioural and biological measures of exposure and outcome;
- exploration of technological developments which may facilitate information collection from large numbers of cohort participants;
- outreach to industrial and other private-sector partners to delineate the commercial value of this undertaking and identify mutually beneficial research areas;
- evaluation of sampling options.

Working groups are required to complete the preliminary developmental work to implement selected pilot studies and to begin writing the application for funding in the areas of, for example, exposure measurement, sampling and handling of biospecimens. The broad scientific community is invited to participate, including people working in the areas of epidemiology, biostatistics, public health, clinical research, technology transfer, history, economics, geography, anthropology, sociology and political science. The first year will involve identifying committed individuals (or teams), developing a governance structure, identifying a leadership team, creating working groups to focus on protocol development, identifying individual(s) responsible for liaison efforts with potential funding agencies and stakeholders, and selecting staff to assist the protocol development. The developmental phase requires completion of pilot studies and environmental scans, selection of data collection methodologies, selection of research centres and assessments of infrastructure requirements, and development of a detailed protocol. Governance and leadership structures will ensure that the work proceeds in a timely and scientifically sound fashion. A scientific advisory board will help to infuse the necessary breadth of vision, capitalize on transdisciplinary opportunities and develop fundraising plans.

This is an ambitious program of work. However, the potential is great for generating new research knowledge, for providing avenues for innovation and economic development and for affecting public health and health policy.

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Appendix 1
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