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Workshop Report: Identification of Research Needs in Breast Cancer Etiology

Christine Friedenreich, Loraine D Marrett, Members of the Canadian Breast Cancer Initiative Working Group on Primary Prevention of Breast Cancer and an Expert Panel

Abstract

A workshop to evaluate the scientific evidence for the etiologic associations between modifiable lifestyle and environmental risk factors and to identify areas for future research in breast cancer etiology was sponsored jointly by the Canadian Breast Cancer Initiative and the Canadian Breast Cancer Research Initiative in May, 2001. Reviews of the scientific evidence in these topics were commissioned and an expert panel was convened to consider the reviews and make recommendations for research. The panel concluded that there was substantial evidence to proceed with additional research in several areas of breast cancer etiology. Particular support for future research for several lifestyle and environmental risk factors including alcohol, diet, physical activity, anthropometric factors, hormonally active agents and occupational exposures was identified. Several emerging hypotheses for breast cancer etiology were also considered and recommendations made in these areas. Specific areas for future consideration included: insulin-like growth factors, pharmaceuticals, viruses, psychosocial factors, and functional polymorphisms. The panel also identified common themes for future research including: studies of exposures across the life cycle; research in populations with unusual exposure levels; consideration of effect modification; development of improved exposure assessment methods and use of intermediate endpoints; separation of disease subtypes by hormone receptor status, stage and tumour markers; and consideration of biological mechanisms in breast cancer etiology.

Key words: breast cancer etiology; environment; lifestyle; primary prevention; risk factors

Introduction

Breast cancer in Canada

Breast cancer is the most common cancer in Canadian women: an estimated 19,500 Canadian women will be diagnosed with breast cancer in 2001, and 5,500 will die from the disease.[1] About one in every 10 women in Canada can expect to develop breast cancer in her lifetime. Canada, along with Australia, Western Europe and the United States, has the highest incidence in the world, with rates more than four times those in low-incidence countries in Asia and Africa.[2]

Furthermore, incidence has been increasing over at least the past 20 years in Canada; it is now about 25% higher than that in the early 1980s.[3] Fortunately, mortality has been declining in recent years, probably because of intensive efforts to implement organized mammographic screening programs in most provinces and territories and improvements in treatment.

Despite the importance of the disease and substantial international research into its etiology, only about 25–40% of breast cancer incidence in Canadian women can be attributed to identifiable risk factors.[4] Unfortunately, many of these factors are not directly modifiable (e.g., family history of breast cancer, menstrual characteristics, age at first pregnancy).

The Canadian Breast Cancer Initiative Working Group on Primary Prevention

In 1993, Health Canada established the Canadian Breast Cancer Initiative (CBCI) with a mandate to reduce breast cancer morbidity and mortality. One

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component of the CBCI is the Canadian Breast Cancer Research Initiative (CBCRI). The CBCRI is a separate alliance of the public, private and charitable sectors, including the major funders of medical and cancer research, fundraisers, breast cancer survivors and advocates who have worked collaboratively to promote and fund breast cancer research in Canada.

In February 2000, the CBCI established the Working Group on Primary Prevention to provide advice on priority areas for research and prevention initiatives. Given the urgent need to identify means of reducing breast cancer incidence and the lack of etiologic information that would allow such primary prevention initiatives to occur, the Working Group decided to begin by identifying research needs around *modifiable* risk factors. "Modifiable" includes those risk factors and behaviours that individuals as well as public health policies might be able to modify or control in some way. The Working Group explicitly excluded chemoprevention because this issue is being addressed by another CBCI component. The limited time and resources available to the Working Group also prevented the review of exogenous hormone use.

Identification of Priority Research Needs for Modifiable Breast Cancer Risk Factors

Goal, objectives and approach

The Working Group established as its first goal *the identification of gaps in knowledge and research needs for breast cancer in women that will inform primary prevention research (excluding research about chemoprevention).*

It identified two specific objectives, namely:

1. To evaluate scientific data on the etiology of breast cancer;

2. To provide recommendations for future research on modifiable risk factors, with particular emphasis on lifestyle and environmental risk factors and the underlying biological mechanisms involved in the etiology of breast cancer.

The Working Group adopted a two-step approach to accomplish these objectives:

1. Literature reviews covering specific topics;

2. An expert workshop.

Literature Reviews

Reviews were conducted by members of the Working Group and two additional scientists on the topics shown in Table 1.[5-15] Most of the reviews related to known or suspected modifiable lifestyle or environmental risk factors for breast cancer. A separate review considered new hypotheses and methodologic approaches in breast cancer etiology.[13] This review included topics that were not covered by the other reviews but which may warrant further research. Two special reviews were conducted on the biological aspects of breast cancer to provide back-

TABLE 1 Topics of literature reviews	
General area	**Reviews conducted**
Known or suspected modifiable lifestyle and environmental risk factors	• Smoking (active and passive) • Alcohol • Diet • Physical activity • Anthropometric factors • Electromagnetic fields (EMF) • Organochlorines • Occupation
New etiologic hypotheses	• New and emerging hypotheses and methodologic approaches
Biology	• Biological mechanisms • Evolutionary aspects of etiology

ground relevant to the identification of additional fruitful avenues of research.[14,15]

Each review summarized the literature and made recommendations on substantive and methodological research needs for that topic. In making their recommendations, the authors tried to identify areas that might not have traditionally received adequate or complete research attention, as these might prove particularly valuable in terms of preventive potential.

The literature reviews were presented to and critiqued by other members of the Working Group, then revised accordingly. A summary report[16] was prepared to provide the highlights of each literature review and its main research recommendations. The summary report and the more detailed reviews were provided as background material for the second step of the process, the expert workshop.

Expert Workshop on Primary Prevention of Breast Cancer

Nine American and Canadian experts in various areas related to breast cancer etiology were invited to attend a workshop in Quebec City on May 3, 2001. The workshop immediately preceded the Canadian Breast Cancer Research Initiative's *"Reasons for Hope 2001"* Second Scientific Breast Cancer Research Conference. Members of the Working Group also attended the workshop. The names of the workshop participants are listed in the appendix.

Goal

The goal of the workshop was to develop consensus recommendations for etiologic research needs that might ultimately lead to the primary prevention of breast cancer.

Process

The workshop process is outlined in Table 2. The experts received the literature reviews prior to the workshop and were asked to

• read the reviews and consider the research recommendations in them;

TABLE 2 **Workshop process**	
Pre-Workshop	• Literature reviews conducted by Working Group members, with research recommendations • Experts read reviews and revise/identify additional research recommendations
Workshop	• Small group review and consolidation of research recommendations for specific topic areas (excluding new hypotheses and approaches) • Full group discussion of consolidated recommendations in specific topic areas • Full group brainstorming regarding recommendations for new hypotheses and approaches • Full group review of all recommendations and development of consensus for inclusion/exclusion

TABLE 3 **General discussion areas**	
Issue	**Resolution**
• Lack of review of hormone replacement therapy as a modifiable risk factor	• To be discussed as part of "new hypotheses and approaches"
• Etiology of breast cancer vs. other chronic diseases	• Although overlap of risk factors with those of other chronic diseases, stay focused on breast cancer
• Role of multidisciplinary research	• To be encouraged
• Human vs. animal research into mechanistic pathways	• Human studies favoured

- recommend needed revisions to these recommendations; and,
- identify additional recommendations for both substantive and methodological research.

Each expert was individually asked to pay particular attention to three specific topics and to provide his or her recommendations for research on these prior to the workshop. The experts were *not* expected to critique the reviews.

At the workshop, participants were initially asked to meet in small groups focused on a set of related topics (smoking and alcohol; diet, physical activity and anthropometric factors; EMF, organochlorines and occupation; and biological mechanisms and evolutionary aspects of etiology). Each group was asked to discuss and consolidate the recommendations made by the original reviewers and by the experts. The consolidated recommendations on each topic were then presented to the remaining workshop participants and discussed. New hypotheses and methodological approaches were discussed by all workshop participants in a plenary session. Finally, workshop participants reviewed all research recommendations and agreed on areas and topics worthy of further consideration.

TABLE 4 **Criteria for recommending priorities** **for future research**
• Biological plausibility
• Potential for modifiability
• Magnitude of the problem (i.e. strength of the association and exposure prevalence)
• Feasibility to conduct research in the short term with limited budget
• Level of current supportive evidence
• Ability to study or measure
• Unique opportunity or need within Canada

Workshop Results

General Discussion

There were some general issues of clarification that required discussion and resolution at the start of the workshop (Table 3).

There was also discussion of the criteria that should be used for recommending an area as worthy of consideration for future research. The criteria selected are shown in Table 4; not all need to be satisfied simultaneously.

Research Recommendations Resulting from the Workshop

The recommendations and discussion identified a number of common methodologic themes, such as suggestions for types of research or approaches to research that were considered relevant for several of the specific risk factors or exposures (Table 5). Many of these themes are expanded upon in the topic-specific recommendations.

Table 6 summarizes the recommendations for each of the specific topics for which background literature reviews were prepared. It includes recommendations from both the reviews and the workshop. Table 7 identifies recommendations for research in emerging topic areas. Many of these topics have not been fully investigated. Although some areas may be more speculative, most are supported by suggestive evidence and/or biological plausibility.

An examination of the biological mechanisms and evolutionary aspects of breast cancer etiology was included in the workshop discussions because understanding these areas will enhance the development and testing of relevant hypotheses on the etiology of breast cancer. For example, the importance of reproductive hormones in the etiology of breast cancer is accepted yet the relationship between levels of hormones and breast cancer is not well understood. It may be important to improve understanding of how hormones are regulated through the complex internal feedback loops, and how these loops are affected by exogenous hormonally active agents. Furthermore, it will be important to consider simultaneously effects on breast cancer risk of the range of hormones in a given metabolic pathway. These

TABLE 5
Common methodologic research recommendations for studies of breast cancer etiology

Recommendation	Rationale

• Consideration/study of effect modification of lifestyle/environmental exposures by:
  • Genetic predisposition (polymorphisms, specific mutations, etc)
  • Race/ethnicity
  • Menopausal status
  • Other lifestyle/environmental exposures
	Effects might vary across subgroups of the population or in conjunction with other exposures.

• Research in specific populations with "unusual" exposure levels or "unusual" disease risks	Populations with particularly high levels of exposure might provide more power to detect effects and might help extend the dose-response curve. Populations at high or low risk of breast cancer might also be informative.

• Study of exposures across the life cycle, particularly:
  • In "susceptible" periods (e.g., puberty, pregnancy)
  • In utero (i.e., intergenerational studies)
  • Early in life
	Response of breast tissue to exposures must vary over the life cycle, as do exposures themselves. There might be some periods of particular susceptibility.

• Improved exposure assessment:
  • Better questionnaires
  • Use of biomarkers
  • Objective measures
  • Statistical methods for measurement error
	Poor assessment of exposure and lack of consideration of measurement error might mask true effects.

• Development and use of intermediate endpoints as indicators of breast cancer risk:
  • Mammographic breast density
  • Early thelarche (start of breast development)
	Often, longitudinal and mechanistic studies cannot wait for breast cancers to develop, so relevant short-term endpoints are needed. This type of research can lead to a better understanding of the natural history of the disease.

• Separation into disease subtypes according to:
  • Hormone receptor status
  • Stage
  • Tumour markers
	Exposures might relate to risk differentially by tumour characteristics.

• Consideration/study of biological mechanisms • Determinants of steroid hormones, prolactin and insulin-like growth factor (IGF)-related growth factors across the life cycle	Understanding the biological mechanisms for various risk factors may lead to more fruitful avenues for etiologic research.

• Use of all types of study designs where appropriate:
  • Longitudinal
  • Case-control
  • Cross-sectional
  • Descriptive
  • Interventions with intermediate endpoints
	Each design will have some limitations, thus, the most appropriate designs need to be considered and used.

mechanisms are intrinsic to establishing biological plausibility (Table 8).

Conclusions

Although the Working Group's focus is primary prevention, the research needs identified were actually in the area of *etiology*, rather than primary prevention interventions. The Working Group felt that additional research into etiologic factors with potential for primary prevention was required before large-scale intervention prevention research could be recommended. Even for those risk factors where strong evidence of association exists, much information essential to the application of practical risk reduction interventions remains unknown.

A large number of areas for potentially fruitful research into the etiology of breast cancer were identified through the background reviews and the workshop. There was no attempt to prioritize these, other than to identify those for which the need for research was considered very low. This decision was made because the Working Group wanted to stimulate innovative research in a broad range of topics potentially relevant for breast cancer etiology.

The overlap of modifiable risk factors for breast cancer with those for other common chronic diseases was noted (e.g., body weight and diabetes; physical activity and heart disease), and the resultant importance of societal change and public health policy in effecting behaviour modification that would have multiple benefits was stressed. It was agreed that general directions for public health policies that may promote breast cancer risk reduction can be developed at this time despite the fact that only limited scientific evidence regarding specific interventions for the primary prevention of breast cancer at the population or individual clinical level currently exists.

Next Steps: Stimulating Innovative Breast Cancer Etiology Research in Canada

One of the key strategic areas identified by the CBCRI is breast cancer etiology research. In its 1998 Strategic Research Agenda CBCRI identified both primary prevention and environmental agents as breast cancer risk factors of special concern. CBCRI has closely followed the progress and work of the CBCI's Working Group on Primary Prevention of Breast Cancer since its inception. Following the workshop, on May 4, 2001, a draft request for applications for research in breast cancer etiology was approved in principle by the CBCRI Board of Directors and announced at its "Reasons for Hope 2001" conference. This major initiative in breast cancer research is intended to respond to the gaps in breast cancer etiology research identified through the literature reviews and the workshop. These literature reviews and recommendations for future research will be used as the basis for a special competition in breast cancer etiology research in Canada that the CBCRI hopes to launch in the near future.

TABLE 6
Summary of research recommendations for specific modifiable risk factors

Risk factor	Summary consensus of evidence	Specific recommendations
Smoking	Little evidence of effect. Weak biological rationale. Methodological problems with assessment of passive smoking.	• Overall, low priority for research • Studies in genetically defined susceptible sub-populations • Assessment of *in utero* and early life exposures where data exist to perform efficient studies (e.g., by record linkage)

Alcohol	Risks well known and quantified for moderate levels of drinking.
	
	More research needed for high consumption and to assist in developing appropriate primary prevention strategies (e.g., identification of high-risk subgroups). Full understanding of the biological mechanisms underlying the association still needed.
		• Gene-exposure interaction studies
		  • Genotypes involved in alcohol metabolism
		  • Pooling across studies/centres to gain power
		• Mechanistic studies
		  • Effect on endogenous hormones and proteins, including those not well examined (e.g., progesterone, prolactin, IGF, androgens)
		  • Effect on target tissue
		• Study of populations with high levels of consumption
		• Effect modification by diet, particularly folate, body mass, and physical activity
		• More comprehensive exposure assessment
		  • Exposures early in life and over the lifecycle
		  • Role of binge drinking
		• Study of effect modification by ethnicity
		• Association with intermediate endpoints (e.g., breast density)
		• Effect in relation to tumour stage and hormone receptor status

Diet, physical activity and anthropometry*	Weight control and physical activity probably linked to breast cancer. Evidence specific enough to plan interventions on weight control but not yet adequate for physical activity interventions. Evidence for a link with diet less strong. More research needed to clarify interrelationships between diet, physical activity and anthropometry and breast cancer risk.	• Clinical metabolic studies of effects of specific diet, physical activity and weight control interventions on sex hormones, prolactin and IGFs • Intervention trials of specific changes in these risk factors, using both intermediate and long term endpoints, that examine the relative contribution of each risk factor to breast cancer risk reduction • Incorporation of high quality measures of all three factors, including relevant parameters such as frequency, intensity and duration of physical activity • Characterization and effects of patterns of exposure (vs. individual, specific exposures) within and between the three factors • Effects of exposures at different periods of life, particularly during fetal development and juvenile development before first pregnancy • Gene-environment interactions for physical activity and anthropometric measures • Study of effects of weight change during different periods of life • Role of some specific dietary constituents, such as phytoestrogens
Electromagnetic fields (EMF)	Little solid evidence of a link with breast cancer, but studies to date had serious methodological flaws, limiting ability to draw firm conclusions. Since EMF exposure is ubiquitous, some additional research desirable.	• Study of populations with high levels of exposure, such as in occupational settings (although assessment of non-occupational exposures also needed) • High quality exposure assessment • Mechanistic approaches • Large studies needed so that small excess risks detectable

Hormonally active agents/environmental chemicals (HAAs)†	Although some chemicals in this class (e.g., DDT/DDE or total PCBs) are not associated with increased breast cancer risk, many others not studied are potentially important. Since exposure prevalence could be high, particularly in some population subgroups, more research needed. Particular attention to agents well recognized as highly estrogenic, e.g., atrazine.	• Development of screening methods to select agents for further study
		• Development of good exposure assessment methods
		  • Low level exposures
		  • Interrelations between compounds
		  • Measurement during critical life periods
		• Development of analytic methods (computation models) for examining effects of complex mixtures of exposures
		• Study in populations with very high potential exposure levels (e.g., Inuit, those living close to toxic waste sites)
		• Studies to characterize exposure levels, susceptibility and interactions
		  • Surveillance of population exposure levels
		  • Longitudinal exposure characterization (e.g., through blood and human milk banks)
		  • Understudied HAA exposures (including individual PCB congeners)
		  • Toxic equivalency approach
		  • Identification of modifiers of HAA metabolism (polymorphisms, etc.)
		• Effects on possible intermediate endpoints such as premature puberty or thelarche
		• Effect modification by body mass, parity, lactation

TABLE 6 *(cont'd)*
Summary of research recommendations for specific modifiable risk factors

Risk factor	Summary consensus of evidence	Specific recommendations
Occupational exposures	Methodological flaws common in studies conducted to date. High quality studies needed. Possible effects of women's work on breast cancer risk understudied. The female workforce traditionally concentrated in administrative, teaching, clerical and other indoor occupations. Some of these groups have an excess of breast cancer. Further study of exposures and other less traditional jobs relevant to breast cancer in this group may be useful.	• Studies employing "best practices" in terms of 　• specificity of exposure (e.g., active ingredients vs. job title) 　• occupational exposure assessment 　• power 　• measurement and control of non-occupational exposures and potential confounders such as menopausal status 　• examination of dose-response relationships • Explore use of biomarkers such as DNA adducts in nipple aspirates or breast milk • Focus on substances with a biological rationale 　• Animal bioassays indicate potential for mammary carcinogenesis (e.g., dyes, solvents, metal oxides) • Study of occupations with high proportions of women, including homemakers and administrative/clerical workers 　• Solvents and other household chemicals 　• Indoor air quality or other possible explanations for high breast cancer risk in some of these groups (occupational exposures vs. lifestyle or other factors)

* These risk factors were considered together because of their very strong interrelationships
† The topic organochlorines was expanded to hormonally active agents/environmental carcinogens during the workshop discussions.

TABLE 7
Summary of research recommendations for new and emerging hypotheses and methodologic approaches

Risk factor	Summary consensus of evidence	Specific recommendations
Insulin-like growth factors	Growing area of research; much literature on the mechanistic relationship between IGF and ovarian function but not yet on the epidemiological side. Multidisciplinary research will be required.	• Relation of tissue levels to circulating levels • Relation to mammographic density • Effects pre- vs. post-menopause • Determination if associations independent of steroid hormone levels 　• Relation to steroid hormone levels • Role of genetic polymorphisms • Study of how other risk factors influence IGF levels, e.g., physical activity • Study to determine what affects binding proteins and receptors
Hormone replacement therapy (HRT)	Potentially an important modifiable risk factor to study, given the large number of women using various forms of HRT. Evidence that the addition of progestin to HRT further increases breast cancer risk.	• Effect of very low dosage HRT on breast cancer risk • Effect on breast tissue of delivery by patch and other methods (e.g., intravaginal), cyclical and continuous 　• use biomarkers and intermediate endpoints (serum levels, pharmacokinetic studies, mammographic density) • Effect modification in relation to other risk factors, particularly physical activity and body mass • Evaluation of interventions that might beneficially affect hormone levels
Other pharmaceuticals	Suggestive evidence of association with breast cancer for some classes of drugs. Further investigation needed that establishes biological plausibility.	• Study of tranquilizers for possible causal mechanisms through hormonal or hormone-receptor effects • Study of cholesterol-lowering drugs because they have been shown to affect IGF receptors • Study of ovulation-stimulating drugs (e.g., clomiphene) because it might induce premature thelarche • Drug studies must include: 　• Control for other risk factors, including HRT, and the condition for which the drug is prescribed 　• Duration of use

TABLE 7 *(cont'd)*
Summary of research recommendations for new and emerging hypotheses and methodologic approaches

Risk factor	Summary consensus of evidence	Specific recommendations

Early life exposure	Considerable interest in this area of research, as indicated in the common research themes and specific modifiable risk factors.	• Use existing national (or other) longitudinal studies of children
		  • add biological sample collection
		  • measure important risk factors (physical activity, diet, etc.)
		  • use biomarkers (e.g., hormone levels) and intermediate endpoints (e.g., mammographic breast density)
		• Breast cancer in children born from pre-eclampsic pregnancies
		  • record linkage studies
		• Effect of feeding with soy formula on age at menarche and other indicators
		• Effect of birth weight, being a twin on hormone levels, breast density

Viruses	Possible fruitful area for future research. Study of viruses found in tumour tissue not recommended.	• Epstein-Barr virus and mouse mammary tumour virus are leading viral candidates for research currently • Investigations using stored serum samples
Functional polymorphisms and gene-gene interactions	Polymorphisms and mutations are likely to be important in identifying susceptible and non-susceptible individuals and the potential for gene-environment and gene-gene interactions. Area will be exploratory, initially, and iterative.	• Development of methods to identify *functional* polymorphisms for study • Interaction between epidemiologists and laboratory scientists will be important • Improved statistical methods for analysing multiple markers • Establishment of banks of biological material for use once relevant markers have been identified • Need for studies with large sample sizes

Other diseases	Number of diseases might be associated with altered androgen levels that might be involved in breast cancer etiology.	• Incorporation of high quality diagnostic data into studies of other diseases
		  • record linkage studies
		  • consider possible underlying mechanisms linking the diseases

Psychosocial factors/ Stress	Stress been shown to induce immune suppression that might be linked to breast cancer. However, study in this area is methodologically challenging.	• Use of high quality measures of stress, including objective ones • Role of chronic vs. acute stress • Relation of acute stress to tumour progression • Role of pregnancy as a major immune-system suppressor

TABLE 8
Summary of research recommendations for biological mechanisms and evolutionary aspects of breast cancer etiology

Biological Mechanisms:

• Study of risk factors in relation to mutation rate, mitotic indices and mammary tissue differentiation

• Study of known mutagens and carcinogens in human mammary epithelial cells

• Study of molecular and cellular mechanisms in humans

• Methods of distinguishing parous versus nulliparous breast tissue

• Role of apoptosis in breast cell cultures

Evolutionary Aspects:

• Meta-analyses of existing studies that examine data from an evolutionary perspective including factors such as mutation rate, number of cell divisions that occur during menarche, pregnancy, menopause

• Translation of known risk factors into shared biological mechanisms (e.g., estrogen/cell division/ life span to date)

• Multidisciplinary collaborations to apply basic science models to human data of breast cancer

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APPENDIX
Participants: Canadian Breast Cancer Initiative (CBCI) Workshop
on the Primary Prevention of Breast Cancer, May 3, 2001

Expert Panel

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Assessing the Surveillance Capability of Canada's National Health Surveys

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Abstract

We assessed Canada's national health surveys as surveillance instruments, with emphasis on comparing the temporal structure of data sets with those generated by the US Behavioral Risk Factor Surveillance System (BRFSS). Only the Canadian Tobacco Use Monitoring Survey (CTUMS) has the BRFSS capability to generate continuous, uniform time series with monthly intervals. These time series can offer substantial extra value for retrospective analysis such as program evaluation in addition to surveillance. Expanding CTUMS is a simple option for providing an ongoing, uniform monthly survey instrument for non-tobacco variables. The Canadian Community Health Survey (CCHS) will generate monthly data, and could potentially generate useful continuous time series even though surveys at the health region and provincial levels will alternate annually. Reconfiguring the CCHS, or even implementing a provincial surveillance survey based on the BRFSS model are other viable options, but each option has associated tradeoffs or obstacles.

Key words: *BRFSS; CCHS; CTUMS; health information; health surveys; surveillance time series; temporal change*

Introduction

Improving surveillance has been a significant issue in recent discussions on upgrading Canada's health information infrastructure.[1–5] Specific actions to meet this demand include the introduction of two new national health surveys, the Canadian Tobacco Use Monitoring Survey (CTUMS) and the Canadian Community Health Survey (CCHS). While the new surveys will add greatly to our surveillance capability, we believe that there may still be scope for further cost-effective improvement. We will focus on data set temporal structure, since the way data are generated over time is a fundamental property of survey design configuration and has important implications for surveillance timeliness and statistical analysis.

National health surveys are not the only tools available for surveillance. Compelling cases can also be made, for example, for strengthening Canadian capability in the surveillance of policy implementation[6] and for making better use of numerous potential sources of surveillance data at local levels.[7] But by generating population-based data for a wide range of health-related variables, national health surveys contribute to surveillance by providing:

- point-in-time, demographic portraits of population health;

- a means for detecting temporal changes in health factors;

- information on associations among variables;

- a scientifically credible basis for cross-validating information from other surveillance instruments, especially those that generate "quick and cheap" data (e.g., sales records); and

- a basis of comparison for localized or specialized surveillance activities.

The national survey data sets tend to have lasting value. Longer time series with high, consistent data quality have greater analytic value, and this places a premium on establishing survey systems that will be maintained over time.

Definitions of public health surveillance typically emphasize that it must be ongoing, a prerequisite for detecting change over time, and it must be integrated with advancing public health goals.[3,8] At its root, proper surveillance involves "keeping a close watch", an idea

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that goes beyond simply monitoring for temporal change. How close the watch should be is dependent on what is being watched. Both incoming ballistic missiles and advancing glaciers can have enormous impacts, but it would be a waste of resources to conduct surveillance on the latter at the intensity required for the former.

Seismographs and smoke detectors are in one sense ideal surveillance instruments in that they operate continuously. Little would be saved by turning them off for intermittent periods brief enough that their function would not be compromised. Similar instruments for public health surveillance would, as noted by McQueen in reference to lifestyle factors in particular, "[collect] data continuously, producing a seamless flow of data that can detect subtle and long-term changes in the variables of interest at the population level".[9] However, the significant cost associated with each interview and statistical considerations relating to desired precision and power mean that, for a fixed cost, there are real trade-offs between the quantity and quality of information that can be acquired for each point-in-time cross-sectional survey and survey frequency.

The US Behavioral Risk Factor Surveillance System (BRFSS) conducts ongoing monthly surveys that can generate continuous uniform time series. This is likely as close to the ideal of seamless data flow as is practical for a national health survey. The same design for generating data over time has been adopted by CTUMS. Some similar design elements suggest that the BRFSS also influenced the CCHS. Some key differences remain between the BRFSS and the Canadian surveys, however, so it is part of our methodology to use the BRFSS as a comparative model.

In operation for over a quarter century, the BRFSS has been refined with experience and has built a substantial, well-documented record of use. The BRFSS has served as a model for surveillance surveys in other countries, such as Australia and China. In addition to being ongoing, the BRFSS has the flexibility to meet federal, state and local needs. There has always been a need in Canada for more focused provincial and local information as it is in the provincial and local mandates to develop their own policies and deliver public health services.

We take a broad view of surveillance. Differentiating the function of surveillance from other potential uses of data, such as research, is a useful point of distinction for assessing surveillance adequacy,[8,10] but the cost-benefit analysis for evaluating a surveillance instrument should include all potential benefits, whether or not they strictly constitute surveillance per se. In addition to the "hard" benefits arising from the data generated, the instrument can produce "soft" benefits by motivating information acquisition, dissemination and use.

Potential changes in a health information infrastructure or a component such as a survey can be assessed in terms of marginal utility. We propose that an optimal surveillance system would be one for which increasing or reallocating resources could not yield cost-effective gains in information value, nor could resources be reduced without a loss of information value that outweighs the savings achieved. In the cost-benefit analysis this entails, it is typically easier to quantify costs than benefits. Reductions in mortality and morbidity and program savings are quantifiable measures for validating change. Other benefits may be more difficult to quantify, especially in economic terms, yet these may be the most important justification for a program. These include:

- better knowledge of the awareness and impact of policies and programs;

- more effective delivery of health services that may not save money, but do improve client health, comfort, or convenience;

- better understanding of the factors affecting human health;

- development of ancillary information that places targeted research into a broader context and assists in prioritizing further research, surveillance, and policy initiatives;

- assurance that newly emerging health issues will be identified and acted upon in a timely manner;

- assurance that current programs are still appropriate and adequate; and

- greater ability to address the health needs of special subgroups.

Such diverse considerations ensure that "optimization" is not a simple algorithmic exercise, but more of a balancing act. Formulating a coherent surveillance strategy should hasten the evolutionary process of refining the surveillance system and ultimately reduce development costs.

Background and Overviews

US Behavioral Risk Factor Surveillance System (BRFSS)

The BRFSS is a state-based, ongoing survey that conducts a number of random-digit-dialed telephone interviews each month (see the BRFSS web site for extensive documentation).[11] Formally established in 1984 by the Centers for Disease Control and Prevention (CDC) and 15 participating states, the BRFSS grew to include all 50 states, the District of Columbia, and three territories by 1997. The target population consists of those 18+ years old living in households. (The US has a separate school-based Youth Risk Behavior Surveillance System, not treated here.)[12] State sample sizes in 1999 ranged from 1,248 to 7,543 per year, with a median of 2,939 (D. Nelson, personal communication). The CDC attempted to fund about 2,500 interviews per state in 1999; state discretion in spending CDC funds and state-funded additional interviews account for the variation in sample size.

Key BRFSS attributes are the nature of the federal-state collaboration, the practice of conducting surveys at monthly intervals, and flexibility. The CDC coordinates the BRFSS, designs a substantial part of the questionnaire, provides logistical support, and supplies core funding. The questionnaire has three parts: a core component, common for all states and prepared by the CDC, consisting of fixed, rotating, and emerging core questions; optional modules prepared by the CDC but selected for inclusion by each state; and questions that are developed or acquired by each state. State-added questions are rated highly for utility by BRFSS program directors.[13] Each state conducts its own survey. Implementation differs somewhat among states; for example, some use in-house units to conduct the interviews, while others contract them out. The state-based survey system is believed to make it easier to address local issues, such as the impact of a new state program.

Spreading the annual sample evenly over 12 months keeps the BRFSS ongoing, which in turn benefits data quality, data analysis, and the capacity to react to emerging issues. Employees can be kept on a continuing basis, and interviewers can be highly trained and can cost-effectively gain experience and consistency over time. It is also BRFSS experience that a bias shift (such as a change in the proportion underreporting a particular behaviour) is more readily detected with continuous monthly data if a question or methodology is modified (D. McQueen, personal communication).

The data sets generated potentially offer substantial added value for statistical analysis in comparison to annually aggregated data sets. The continuous time series with monthly intervals generated for fixed core questions have the following attributes:

- the data are better suited for detecting temporal changes;

- data are collected at frequent enough intervals that the effect of a program or policy can be tracked over time, a valuable property when the effect occurs quickly;

- data can be flexibly aggregated to larger time periods (e.g., annually, semi-annually, quarterly), or to make before-and-after comparisons when a new program is initiated or other potential impacts occur;

- seasonality components can be examined, both for their own sake and to de-seasonalize the monitoring of temporal trends; and

- statistical procedures analogous to quality control techniques (e.g., *p*-charts, CUSUM charts[5]) would allow fast response to statistically significant changes in time.

Rotating core questions generate 12 months of data cycling with a 12-month gap. Typically, such data are pooled by year and treated as equivalent to biennial surveys, but seasonality components can be examined with the monthly data. Rotating core modules are made available as optional modules in off-core years, so individual states can maintain ongoing surveillance on

these variables. Illinois splits the state sample and runs dual questionnaires (current year plus previous year), so that all "rotating core" modules are run continuously (B. Steiner, personal communication).

In practice, most data are rolled up annually for reporting and analysis.[14] Examples of monthly data use include an examination of seasonal patterns of leisure-time physical activity[15] and a time series analysis of trends in perceived cost as a barrier to medical care.[16]

Two other lessons have emerged from BRFSS experience. First, the use of surveillance data builds over time.[13] Second, concentrating resources on "front end" data collection at the expense of "back end" analysis can result in sub-optimal information yield.[9]

Canadian National Health Surveys to 1998

The Canadian Sickness Survey in 1950/51 initiated what is now a half-century tradition of national population-based health surveys.[17] These surveys have provided a wealth of health information. Repeating surveys typically provide the best basis for detecting temporal change. Current national surveys that are intended to be repeated on an ongoing basis include the National Population Health Survey (NPHS), General Social Survey (GSS), Tracking Nutrition Trends Survey, Health of Canada's Youth Survey (HCYS), National Longitudinal Survey of Children and Youth (NLSCY), Physical Activity Monitor, and National Vaccine Coverage Survey.[17]

Information on the same variable has also been collected over time under surveys of different names. Results from an eclectic collection of surveys can be compared if the surveyed population and the wording of the questions are closely comparable, and if survey methodologies do not introduce significant biases or if the biases are at least consistent.

Papers on tobacco use by Stephens[18–19] and Gilmore[20] illustrate various strengths and weaknesses in the set of national health surveys for monitoring temporal change prior to 1998. Stephens'[18] 1988 review of tobacco use, attitudes and knowledge used four different survey sources: the Labour Force Survey (LFS) for 11 different years between 1965 and 1986, the Canada Health Survey for July 1978 to March 1979, the Health Promotion Survey for June 1985, and the General Social Survey (GSS) for October 1985. Factors enhancing integration of the multiple survey sources were:

- similar sampling designs;

- almost identical wording for relevant questions;

- Statistics Canada collected all data; and

- well documented methods allowed informed judgments on variations in approach.

Consistency was hindered, however, by variations in methodologies (e.g., both household visits and telephone interviews were used) and particularly by the acceptance of proxy data in the LFS. Estimates of tobacco use in

youth by proxy data were shown to have a strong downward bias, and proxy report data could not be separated from self-report data prior to 1981.

Stephens[19] reported on the results of a workshop held to examine trends in smoking prevalence from 1991 to 1994. A landmark event during this period was the federal action on February 8, 1994 (with concordant actions by five provinces on or shortly thereafter) to sharply reduce tobacco taxes in order to combat cigarette smuggling. Key information on smoking prevalence was provided by the 1991 GSS and the 1994 Survey on Smoking in Canada, but no comparable information was available for smoking prevalence in 1992 and 1993. Instead, smoking prevalence for these two years had to be inferred from information gleaned from several smaller national and provincial surveys, including three commercial surveys with relatively small sample sizes but consistent time series, as well as industry consumption statistics.

Gilmore[20] analysed the statistical significance of changes in tobacco use prevalence during the period 1985–1999 using only Statistics Canada surveys. This study demonstrated the difficulties in detecting change with continually changing surveys. The GSS for 1991 and 1996, the NPHS for 1994/95 and 1996/97, and CTUMS for the first half of 1999 were judged to be comparable surveys. The National Alcohol and Drugs Survey of 1989, the Health Promotion Survey of 1990, the Canadian Alcohol and Drugs Survey of 1994, and the GSS for 1995 were judged as reasonably comparable to the other surveys for daily smoking rates, but current and non-daily rates were judged as not comparable due to question differences.

We say little about these surveys in the following sections for several reasons. The NPHS and NLSCY are longitudinal surveys, which have limitations as surveillance instruments.[5,21] The GSS now has limited health-related content. The topic of youth surveillance merits separate treatment, and the remaining surveys cover specialized topics. The role and design of these surveys for surveillance will need to be reappraised with a view to how they fit in with the surveillance capabilities of the CCHS and CTUMS. However, we expect the new surveys to have relatively little impact on most of the previously established surveys, which generally provide complementary health information.

Canadian Tobacco Use Monitoring Survey (CTUMS)

CTUMS conducts monthly computer-assisted telephone interview surveys.[22] The target population consists of those aged 15+ years, with residents of the territories and full-time residents of institutions excluded. The annual sample size is targeted at 20,000, partitioned equally among provinces and between the two age groups of 15–24 and 25+. The first questionnaire contained 35 questions related to tobacco use plus demographic questions. Analysis of the first wave of surveys, covering

February to June 1999, was released in mid-January 2000. Reporting is scheduled for each half calendar year.[22]

Canadian Community Health Survey (CCHS)

The CCHS started in September 2000.[23] The design (as of November 1, 2000) consists of two surveys alternating annually in a two-year cycle (briefly summarized here, the design is discussed in greater detail by Béland et al.[24]). The health region level survey in the first year of the cycle has a customized component to meet the individual priorities of the 136 health regions. The target population consists of household residents 12+ years old in all provinces and territories, excluding primarily those on native reserves, Canadian Forces bases and some remote areas. Youths (12–19) and seniors (65+) will be systematically oversampled. The aggregate sample of 130,750 will consist of 115,000 computer-assisted personal interviews (CAPI) and 15,750 computer-assisted telephone interviews (CATI); this breaks down into samples of 2,000–42,260 per province, 800–900 per territory and 280–3410 per health region.[23–24] The second-year provincial-level survey will consist of 30,000 computer-assisted personal interviews. The target population will differ somewhat from the health region-level survey; for example, the first provincial-level survey, focusing on mental health, will be restricted to those 15+ (B. Diverty, personal communication).

The CCHS broadly covers health determinants, health status and health system utilization. The 45-minute health region-level survey has 30 minutes of common content, 10 minutes of optional content selected from a set of modules, and five minutes of socioeconomic and demographic content. The provincial-level survey will contain additional common content and one focus content topic (an in-depth treatment of a topical issue);[23] the first is expected to run at least one hour (B. Diverty, personal communication). Quarterly release for high-level population health indicators is planned.[23]

Comparing CTUMS and the CCHS to the BRFSS

CTUMS and the BRFSS fixed core component generate data sets with the same temporal structure: ongoing uniform time series at monthly intervals. CTUMS is restricted to tobacco use and has a richer tobacco content, but the BRFSS fixed core includes a broader range of health variables plus additional tobacco use content in the optional modules.

CCHS data sets generated for questions unique to either the health region-level survey or provincial-level survey that are repeated in subsequent cycles will have the same temporal structure as those generated by the BRFSS's rotating core: 12 monthly estimates alternating with 12 months without data. Some common content is expected to be carried over from the health region-level survey to the provincial-level survey. Since health region-level data can be rolled up to provide provincial-level estimates, continuous monthly-interval time series can be generated for common content questions. These time series would have the peculiar property of being generated by

biennially rotating surveys. Results otherwise obtainable through time series analysis could be obscured by differing methodologies, but options exist to minimize or eliminate such differences. For example, by excluding CATI data, CAPI-only estimates could be generated for health region-level survey years, essentially matching the methodology of the CAPI-only provincial-level survey years (G. Catlin, personal communication).

We believe that the impact of the 1994 tax reduction on cigarettes would have been more readily and clearly discerned if a monthly surveillance survey had been in place. With CTUMS now operational, it can be argued that the need has been met for generating uniform time series on tobacco use at national and provincial levels. But the case for maintaining a similar level of surveillance on certain other health variables is reasonable, and it would be prudent to have an instrument in place for close surveillance if other needs emerge, such as tracking the temporal and demographic dynamics of use and exposure to the drug Ecstasy. In the next section we briefly outline some scenarios for enhancing surveillance.

Options for Enhancing Surveillance

Adapt the BRFSS Model for Canada

It is useful to consider what a Canadian adaptation of the decentralized BRFSS survey model might look like. Under various allocation schemes, a national sample of roughly 20,000–30,000 would match information quality at the province-state level. Following the current BRFSS standard of 2,500 per state would result in a national annual sample of 25,000 for the provinces (the current design of CTUMS is similar to this); additional but likely smaller samples would be required to cover the territories. Since Ontario and Quebec carry such heavy weight in the national roll-up (much more so than any pair of states in the US), an allocation scheme designed to balance reliability requirements between national and provincial or territorial levels may be preferable. A scheme employed by the 1994 NPHS resulted in a core national sample of 22,000, with 1,200 (a set minimum) to 1,996 sampled per province or territory except for Ontario (4,817) and Quebec (3,584).[25] Some provinces or territories could choose to share a single survey unit yet administer separate questionnaires.

Advantages of this option include the close involvement of the provincial health systems with the national surveillance system, the flexibility that individual provinces and territories would have on questionnaire content, and BRFSS experience and expertise. We would expect separate Canadian-designed questionnaires, but any questions duplicated with the US survey would allow nation-to-nation or province-to-state comparisons. These comparisons could be of interest given the very different US health care model.

Reconfigure the CCHS

As noted previously, there is at least the potential for the CCHS to generate continuous time series, but issues of data uniformity and content capacity remain to be resolved. If the CCHS had been designed so that the health region-level survey was spread evenly over 24 rather than 12 months, an instrument would already exist for conducting ongoing uniform surveys at monthly intervals for a wide range of health variables. The annual national sample of 65,000 would generally provide information for provincial roll-ups comparable to or exceeding the usual BRFSS state standards; annual samples would exceed 2,000 for all provinces except PEI (1,000). Even the larger health regions would have quite reasonable annual samples. Idaho is one of the few states that stratifies BRFSS sampling by health district, with each targeted for 700 interviews annually (J. Aydelotte, personal communication).

While it may have been operationally simpler to spread the very large health region survey over two years, the current CCHS design appears to have been driven by a requirement to deliver health region-level information quickly. Concentrating the smaller samples for the sparsely populated health regions also has analytic benefits; for example, there were concerns about the validity of estimates for smaller health regions if sampling was spread over 24 months. However, if the health region-level survey was operated continuously, moving averages may have advantages as a way of reporting health region information.

It is hard to fault the design tradeoffs made – the CCHS can deliver health data at an unprecedented level of geographic resolution with the same temporal structure as the BRFSS rotating core, and there is flexibility for selecting content to meet provincial and health region priorities. It may very well be that little extra value is to be gained from closer surveillance on most variables. But we do not believe this is true for tobacco use, and it cannot be assumed true of all other variables. How desirable or necessary it is to reconfigure the CCHS to meet these needs will depend, in part, on the yet-to-be-determined surveillance capability of the common content component, and possibly through insights gained during the first survey cycle. Even if the CCHS is capable of meeting needs for generating continuous monthly time series, the question of whether it is the most efficient instrument for doing so will need to be addressed.

Expand CTUMS to Include Other Variables

Since CTUMS is already operating as an ongoing monthly telephone survey, it would seem relatively simple to expand its scope to include some other variables. Oversampling of youth and young adults is a useful design feature for surveillance of other risk factors. Even if the survey was renamed, the value of existing data would remain intact.

Certainly there is room to add questions while keeping the questionnaire size reasonable. By way of illustration, adding the entire BRFSS fixed core of health-related

non-tobacco questions to the CTUMS questionnaire would still leave it with under 90 questions.

This option is likely to be resisted by CTUMS stakeholders if they perceive that expanding the scope of the survey would result in reduced tobacco use content and/or in a more cumbersome survey that delivers less timely information. But we see no reason why these concerns cannot be met, and the assurance of stable funding and possibly increased sample sizes and extended demographic coverage would be powerful inducements to change.

Discussion

A health information system will continually evolve as it responds to changes in demographics, spending priorities, medical technologies, health care practices, and information technologies. A surveillance system needs both flexibility, to respond to new information and priorities, and stability, to facilitate the detection and evaluation of temporal change. Canada already has several national health surveys in place that primarily require stable funding to continue to act as surveillance instruments.

Populations, needs, resources, institutions and traditions differ between countries. We see both considerable merit and substantial obstacles to adapting the BRFSS model. The decentralized survey structure of the BRFSS generates significant benefits by motivating interest in developing and using health information at state and regional levels. The argument that Statistics Canada should be able to achieve more uniform methodology for better comparisons among provinces and territories seems sound, yet numerous studies have rated BRFSS data highly for reliability and validity.[16] The specialized expertise required to extract greater value from surveillance data with more sophisticated statistical analyses is likely more efficiently provided at the national level.

Among the options presented for generating uniform time series at monthly intervals, the simplest is to expand CTUMS to include other variables. If the CCHS retains its current design, one of the two surveys would be able to generate a data set with the same temporal structure as any BRFSS variable. The common content component of the CCHS could potentially generate satisfactory, if not completely uniform, continuous time series. The CCHS could potentially subsume CTUMS if matters of content depth and timeliness were resolved, but this may not be a realistic alternative. An expanded CTUMS would take some content pressure off the CCHS, which appears to be already saturated for questionnaire content. We note here that the BRFSS, despite having a somewhat narrower focus than the CCHS, may implement dual questionnaires in order to cope with demands for increased content (D. Nelson, personal communication).

Ferrence and Stephens[6] see CTUMS and the CCHS as being complementary rather than competing surveys for the surveillance of tobacco use. CTUMS is expected to be more timely (8–9 months estimated total turnaround time versus 2–2.5+ years for the CCHS; main difference is in time to introduce new topics), has richer tobacco use content, monitors tobacco use only, and generates continuous uniform time series at monthly intervals, while the CCHS has lower age coverage (12+ years versus 15+ years for CTUMS) and superior small area coverage in the health region level survey. This complementary relationship should remain intact if close surveillance needs for non-tobacco variables are met with an expanded CTUMS or through the CCHS common content core.

The question of which variables merit closer surveillance requires further consideration. The dynamic nature of tobacco use and its premier status as a preventable health risk likely makes it the best choice,[26] but it is unlikely to be the only good one. The 2000 BRFSS fixed core was composed of four questions on health status, seven on health care access, one on diabetes, five on tobacco use, 11 on women's health, and 11 on HIV/AIDS. Is this set necessary and sufficient? In planning for the next decade, proposed criteria for selecting BRFSS core measures are based on public health impact, scientific validity, data utility, and implementation considerations.[27] However, placement between the fixed and rotating core is still an open question. Theoretically, close surveillance of any variable is warranted if sufficient extra value is expected. We suggest that factors that can be heavily impacted by government policy, especially through legislation or taxation, merit inclusion. For such variables the greatest value of surveillance data may come through a clearer picture of the impact (or lack of impact) by retrospective analysis of the time series overlapping the time of change.

Viewed from the perspective of data set temporal structure, the set of Canadian national health surveys is close to matching all BRFSS capabilities. The BRFSS still has some flexibility that the Canadian national health surveys do not, and in a coordinating role the CDC is perhaps better situated than Statistics Canada for ensuring that survey content is best designed to meet public health information needs. Statistics Canada held a broad consultation process with users of health information to determine content for the CCHS,[28] but we consider the onus to be on the Canadian health community to ensure content quality. The CCHS survey provides considerable flexibility and an additional level of geographic resolution, but we believe it is best to ensure the capability of conducting close surveillance on variables other than tobacco use. Key issues and questions to address in the further evolution of the national health surveys relate to:

- what content to include and what levels of temporal and demographic resolution are in some sense optimal for each variable;

- adequacy of coverage for youth and other special populations;

- extracting greater value from data through increased application of specialized statistical methods;

- integrating information from survey and non-survey sources;

- ensuring appropriate support and flexibility in assisting provinces/territories and health regions to meet their information needs; and

- obtaining maximal information transferral to health policy and practice.

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Completeness and Accuracy of the Birth Registry Data on Congenital Anomalies in Alberta, Canada

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Abstract

Vital statistics and other administrative data are becoming an increasingly important source for epidemiologic research and surveillance. This study, the first in Canada, evaluated the usefulness of birth registry data on congenital anomalies in Alberta. We compared the number of birth defects recorded in the birth registry with the number collected through the Alberta Congenital Anomalies Surveillance System (ACASS) between 1985 and 1996. In addition, records of 3,881 (99.9%) babies with a birth defect(s) from the ACASS during 1994–1996 were matched to the birth registry by deterministic linkage. Of these, 2,969 babies had single anomalies that were used for validity analysis. The anomalies were grouped by those within International Classification of Disease (ICD) ICD-9 Section XIV (ICD-9=740.0-759.9) and those outside Section XIV. For those within Section XIV, 24 summary diagnostic categories were examined. As shown, the total case count from the birth registry was on average about 3% lower than that from the ACASS between 1985 and 1996. The validity of diagnostic categories is high for the 24 categories examined, with an overall agreement of between 80% and 100%. The sensitivity, positive predictive value, and kappa are also high for all these anomalies combined during 1994 and 1996, showing 95.7%, 99.8, and 0.81 respectively.

Key words: *birth registry; congenital anomalies; reliability; validity*

Introduction

Secondary data, such as vital statistics birth/death registries, physician claims, and hospital records, have become increasingly important sources for epidemiological research and surveillance.[1-5] The birth registry, for instance, may be an important source of information for studies of congenital anomalies and other health events.[2,6] The province of Alberta birth registry has been readily available in a standard format for years and records some 40,000 births each year. Each record also contains infant health and reproductive health-related information such as birth weight, mother's age at delivery, and the presence of congenital anomalies. The major variables available in the registry are listed in Appendix 1. To date, no study has evaluated the usefulness of birth registry data on the study of congenital anomalies in Canada.

Only a few studies have evaluated the adequacy and completeness of the congenital anomalies data. Knox and his colleagues evaluated a national congenital anomaly surveillance system in the United Kingdom (UK)[7] and concluded that the system was inadequate when a case is ascertained from a single source, mainly the birth notice. Another study examined the completeness and accuracy of diagnosis by comparing data from several birth defect registries in the UK and found a high degree of validity in diagnosis.[2] A Canadian study discussed the limitations of the Canadian Congenital Anomalies Surveillance System (CCASS), focusing on data collected from hospital stays.[8]

The Alberta Congenital Anomalies Surveillance System (ACASS), one of the earliest congenital anomalies registries in Canada, routinely collects data on congenital anomalies in the province of Alberta from multiple sources,[9-10] making it a valuable reference for evaluation studies on congenital anomalies in the province. Using data from the ACASS as a "gold standard", this study evaluates the completeness and accuracy of congenital anomalies data available in the

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Alberta birth registry. The total number of cases in the birth registry and the ACASS between 1985 and 1996 were compared for completeness. Like previous studies,[2,11-12] the overall percent agreement, estimated kappa, and sensitivity were used for validity assessment. Regional variations in the overall quality of congenital anomalies data in the province were also compared.

Materials and Methods

Description of Data Sources

Data on congenital anomalies were made computer accessible from the ACASS (1980–1996) and from the birth registry (1985–1996). The ACASS started in 1966 and has the best information on congenital anomalies available in the province. Like surveillance systems in other countries, the ACASS collects data on selected malformations for infants up to one year of age.[9] Cases are obtained from a variety of independent sources, such as the Congenital Anomalies Reporting Form, the Medical Certificate of Stillbirth, the Notice of Birth form, the Medical Certificate of Death, acute care hospitals, and some agencies, outpatient specialty clinics and laboratories. For suspected cases, the ACASS verifies the diagnosis with the relevant physicians/laboratories. Quality measures are taken to ensure the accuracy of diagnosis. The methodology in case ascertainment and evaluation has been detailed elsewhere.[9-10] The British Paediatric Association Classification of Diseases (BPA) code, an adaptation of the International Classification of Disease 9th version (ICD-9), is used in the ACASS for congenital anomalies.

After receiving data on congenital anomalies (in hard copy) from the ACASS, the birth registry staff will, whenever possible, enter the data into the birth registry database. Data on congenital anomalies from the Alberta birth registry are expected to be of a reasonably good quality. The anomalies in the birth registry are also coded according to the BPA scheme. After receiving data from the birth registry, Alberta Health and Wellness determines the geographic location of the mother's residence according to Regional Health Authority (RHA). Data are grouped into five geographic regions for analysis: Southern (RHA 1–3, 5), Central (RHA 6–9), Northern (RHA 11–17), Calgary (RHA 4), and Edmonton (RHA 10).

Case Definition and Assessment of Completeness of Registry

In this study, a case refers to an individual live or stillborn infant who is coded as having one or more anomaly(ies). The diagnosis may be made at birth (in the birth registry) or during infancy (in the ACASS). An anomaly includes structural defects, chromosomal and monogenic syndromes, inborn errors of metabolism, and other related inborn disorders. An individual can have more than one birth defect. Because only the first defect, the major anomaly of a newborn, is available in the birth registry, the number of cases rather than the number of defects is used in this study.

Similar to the ACASS and other studies,[13] the birth case prevalence for this study refers to the number of individual live and stillborn infants with at least one birth defect per 1,000 total births.

It is estimated as the following:

$$\text{Prevalence} = \frac{B_{CASE}}{B_{TOTAL}} \times 1000$$

where:

B_{CASE} = still births ($\geq$ 20 weeks) and live births with a birth defect; and

B_{TOTAL} = total still births ($\geq$ 20 weeks) and live births

The completeness of the registry of congenital anomalies in the birth registry is evaluated by monitoring:

1. the difference in the total number of cases (the birth registry minus the ACASS), and

2. the prevalence ratio (the birth registry divided by the ACASS). The implication of the difference and ratio is:

- difference = 0 or ratio = 1:
  Equal reporting in the birth registry

- difference > 0 or ratio > 1:
  Overreporting in the birth registry

- difference < 0 or ratio < 1:
  Underreporting in the birth registry

Data Linkage and Validity Analysis

Validity is the extent to which the study measures what it is intended to measure. In Stone's work,[2] it was said "Validity has been defined as 'an expression of whether a response or measure actually represents what it purports to; essentially a measure of truth within the terms of reference'.[14] Since the truth is seldom at hand, a more pragmatic definition is 'the extent to which the results of a method agree with an independent external criterion.'"[15] In this study, validity refers to the agreement in each specific diagnostic category/section of congenital anomalies between the birth registry and the ACASS. The diagnosis from the ACASS was used as a "gold standard" for comparisons.

Data from the ACASS during 1994 to 1996 (incomplete at the time of study) were linked to the birth registry file of the same period by:

- BRN (birth registry number), and

- DOB (date of birth of the baby).

During the three-year period, 3,886 babies with congenital anomalies were recorded in the ACASS database. Of these, 3,881 (99.9%) were matched to the birth registry. Five cases that did not match were excluded from further analysis. For those matched cases, 2,969 (76.5%) had single anomalies. These single anomalies were used for validity analysis. The anomalies were grouped by those within the adaptation of ICD-9 Section XIV (ICD-9 = 740.0–759.9) and those outside

TABLE 1
Number of cases and birth prevalence between the birth registry and the ACASS, 1985–1996

Year	Birth registry		ACASS		Birth – ACASS difference	Birth/ACASS prevalence ratio
	Number[a]	Prevalence[b]	Number[a]	Prevalence[b]		
1985	1,602	36.8	1,644	37.7	–42	0.97
1986	1,743	40.0	1,752	40.2	–9	0.99
1987	1,672	39.8	1,705	40.6	–33	0.98
1988	1,812	43.2	1,857	44.2	–45	0.98
1989	1,864	43.1	1,910	44.2	–46	0.98
1990	1,882	43.8	1,945	45.3	–63	0.97
1991	1,631	38.2	1,731	40.6	–100	0.94
1992	1,665	39.7	1,689	40.3	–24	0.99
1993	1,417	35.3	1,416	35.2	1	1.00
1994	1,312	33.0	1,377	34.7	–65	0.95
1995	1,068	27.5	1,150	29.6	–82	0.93
1996	1,084	28.7	1,120	29.7	–36	0.97
Total	18,752	37.6	19,296	38.7	–544	0.97

[a] Only anomalies within the ICD-9 Section XIV are included.
[b] The birth prevalence is expressed as the number of cases per 1,000 total births.

Section XIV. The latter include over 20 diagnostic categories, such as umbilical hernia (ICD-9 = 553.10), inguinal hernia (ICD-9 = 550.90), anomalies of jaw size (ICD-9 = 524.00), hereditary hemolytic anemia (ICD-9 = 282.00), cystic fibrosis (ICD-9 = 277.00), disorders of carbohydrate transport and metabolism (ICD-9 = 271.00), etc. Twenty-four summary diagnostic categories were examined for those within Section XIV.

The diagnostic categories recorded in the birth registry were compared with those in the ACASS. The overall percent agreement, kappa estimate, sensitivity and positive predicative value were estimated according to Fleiss[16] and Sorensen et al.[4] The definition and calculation of each measure are illustrated in Appendix 2. The criteria used to judge the level of percent agreement are greater than 80% for excellent, 61–80% for good, 41–60% for moderate, and less than 40% for poor.[12] The clinical significance of the estimated kappa, which excludes the chance-induced agreement (when positive), was interpreted according to the "benchmarks" suggested by Landis and Koch[17] as the following:

Kappa statistics	Strength of agreement
< 0.00	Poor
0.00–0.20	Slight
0.21–0.40	Fair
0.41–0.60	Moderate
0.61–0.80	Substantial
0.81–1.00	Almost perfect

A chi-square test was applied as appropriate for regional variations and for trend analysis.[18]

Results

Completeness of the Registry

Table 1 summarizes the annual birth case prevalence of congenital anomalies (ICD-9 XIV) from the birth registry and ACASS in Alberta from 1985–1996. The difference in the total number of cases and the prevalence ratio are presented for comparison. As shown, the number of cases from the birth registry is on average about 3% less than that from the ACASS during the 12-year study period. The number of non-captured cases by the birth registry varies by year, from nine in 1986 to 100 in 1991, with a total of 544 under-reported during the 12 years. It is interesting to note that the difference is larger in recent years, especially for 1995, and that in 1993 the birth registry had one case of overreporting.

Accuracy of the Information – Validity of Diagnosis

Table 2 presents the data on single anomalies from the two sources for years 1994, 1995, and 1996. Of a total of 2,969 cases recorded in the ACASS, 2,662 (89.7%) are within the adaptation of ICD-9 Section XIV (7400–7599), and 307 cases are outside ICD-9 XIV. Overall, 93.4% of diagnoses during the three-year period from the two sources are in the same anomaly section. The percentage is relatively higher for anomalies within ICD-9 Section XIV (94.3%) and lower for anomalies outside this section (89.3%). From 1994 to 1996 the agreement was decreasing for anomalies within ICD-9 Section XIV (p< 0.001). It was higher in 1994 (96.5%), dropped slightly in 1995 (95.3%), and was lower in 1996 (90.3%). For those outside ICD-9 Section XIV, the agreement was 90.6%, 88.3%, and 88.9% for the years 1994, 1995, and 1996 respectively.

TABLE 2
Agreement in diagnosis of single anomalies between the birth registry and the ACASS, 1994–1996

Year	Defects within ICD9-XIV		Defects outside ICD9-XIV		Total defects	
	No. of cases	Agree (%)	No. of cases	Agree (%)	No. of Cases	Agree (%)
1994	1,046	96.5	106	90.6	1,152	95.9
1995	830	95.3	120	88.3	950	94.2
1996	786	90.3	81	88.9	867	90.2
Total	**2,662**	**94.3**	**307**	**89.3**	**2,969**	**93.4**
Chi-square test for trend	p = 0.001		p = 0.689		p = 0.001	

TABLE 3
Validity of diagnosis on single birth defects within ICD-9 Section XIV recorded in the birth registry by year, Alberta, 1994–1996

Year	Cases from the birth registry	Cases from the ACASS		Measures of validity			
		Yes	No	Sensitivity	Predictive value	Kappa	Overall (%)
1994	Yes	1,018	4	97.3	99.6	0.9	97.2
	No	28	102				
	Total	**1,046**	**106**				
1995	Yes	807	1	97.2	99.9	0.9	97.5
	No	23	119				
	Total	**830**	**120**				
1996	Yes	722	0	91.9	100.0	0.7	92.6
	No	64	81				
	Total	**786**	**81**				
1994–1996	Yes	2,547	5	95.7	99.8	0.8	96.0
	No	115	302				
	Total	**2,662**	**307**				

Table 3 presents the sensitivity, positive predictive value, kappa and the overall agreement on single birth defects within ICD-9-XIV by the birth registry. As shown, the sensitivity ranges from 97.3% in 1994 to 91.9% in 1996, with an average of 95.7% for the study period. This suggests that during 1994 to 1996, on average about 96% of single anomalies (within ICD-9-XIV) recorded in the ACASS were identified by the birth registry. The positive predictive value was high, from 100.0% in 1996 to 99.6% in 1994. The high predictive value suggests that almost all congenital anomalies within ICD-9-XIV recorded in the birth registry are in the same broad categories as those recorded in the ACASS. The kappa statistic was also high, an average of 0.81 (81%) over the study period. Even the lowest kappa (0.68) in 1996 is still substantial. The overall agreement was high, ranging from 92.6% in 1996 to 97.2% in 1994, with an average of 96.0%.

Further to this analysis, Table 4 presents the percentage of agreement for the major diagnostic categories of congenital anomalies between the birth registry and the ACASS. The agreement ranges from a high of 100% for "Other Anomalies of the Nervous

System" to a low of 80.0% for "Limb Reductions". On average, the agreement is 93.8% for all congenital anomalies. As noted, the agreement is greater than 93% for the majority of diagnostic categories, but it is relatively lower for Down Syndrome (86.6%), "Other and Unspecified Anomalies" (85.7%), "Microcephaly and Hydrocephaly" (90.0%), and "Other Chromosome Anomalies" (90.2%).

Data Quality across Geographic Regions

Health surveillance and studies of congenital anomalies often require comparisons of regional differences. Understanding the accuracy of diagnosis and completeness of data across regions is crucial for appropriate interpretation.

Table 5 presents the overall agreement of diagnoses on congenital anomalies between the birth registry and the ACASS, 1994–1996. As shown, there is no appreciable difference in the agreement across the five geographic regions for all congenital anomalies combined, and anomalies within or outside the ICD-9 Section XIV. The agreement is slightly higher for the anomalies within ICD-9 Section XIV regardless of the region.

TABLE 4
Agreement in diagnostic category of single anomalies
between the birth registry and the ACASS, Alberta, 1994–1996

Diagnostic category of congenital anomalies	ICD-9/BPA[a] code	Total cases	Number of agreements	Agreement (percentage)
All anomalies combined	**740–759[b]**	**2,969**	**2,784**	**93.8**
Neural tube defects	740.0–742.0	55	49	89.1
Microcephaly/hydrocephaly	742.1–742.3	40	36	90.0
Other anomalies of the nervous system	742.4–742.9	17	17	100.0
Eye anomalies	743.0–743.9	33	31	93.9
Anomalies of ear, face, and neck	744.0–744.9	185	178	96.2
Cardiac septal defects	745.0–745.9	185	174	94.1
Valve atresia/stenosis etc.	746.0–746.9	60	57	95.0
Vessel & other CV defects	747.0–747.9	167	160	95.8
Anomalies of the respiratory system	748.0–748.9	24	23	95.8
Facial clefts	749.0–750.2	165	154	93.3
Anomalies of the digestive system	750.3–751.9	119	108	90.8
Anomalies of major genital organs	752.0–752.5	220	213	96.8
Hypospadias & epispadias	752.6	172	166	96.5
Anomalies of other genital organs	752.7–752.9	13	12	92.3
Anomalies of the urinary system	753.0–753.9	128	121	94.5
Musculoskeletal deformities	754.0–754.4	207	201	97.1
Deformities of feet	754.5–755.1	439	418	95.2
Limb reductions	755.2–755.4	20	16	80.0
Other limb anomalies	755.5–755.9	44	41	93.2
Anomalies of bone/spine/ribs	756.0–756.9	109	101	92.7
Anomalies of the integument	757.0–757.9	81	77	95.1
Down syndrome	758.0	97	84	86.6
Other chromosome anomalies	758.1–758.9	61	55	90.2
Other & unspecified anomalies	759.0–759.9	21	18	85.7
Anomalies outside section XIV	Outside_XIV	307	274	89.3

[a] BPA = British Paediatric Association Classification of Diseases (adaptation of ICD-9)
[b] Include diagnostic codes of ICD-9 XIV (740-759) and outside ICD-9 XIV (90 codes).

TABLE 5
Agreement in diagnosis of birth defects between the birth registry and
the ACASS by region, 1994–1996

Geographic region		Defects within ICD9-XIV		Defects outside ICD9-XIV		Total defects	
		No. of cases	Agree (%)	No. of cases	Agree (%)	No. of cases	Agree (%)
Southern	(RHAs 1–3,5)	424	95.8	76	93.4	500	94.0
Central	(RHAs 6–9)	386	95.3	58	91.4	444	93.7
Northern	(RHAs 11–17)	354	95.5	34	94.1	388	94.3
Calgary	(RHA 4)	870	95.3	70	91.4	940	93.9
Edmonton	(RHA 10)	627	94.7	67	88.1	694	93.5
Chi-square test for difference		p > 0.05		p > 0.05		p > 0.05	

Note: Three cases with unknown RHA were excluded.

TABLE 6
Validity of diagnosis on single birth defect recorded in the birth registry by region, Alberta, 1994–1996

Geographic region	Cases from the birth registry	Cases from the ACASS		Measures of validity			
		Yes	No	Sensitivity	Predictive value	Kappa	Overall (%)
Southern (RHAs 1–3,5)	Yes	408	1	96.2	99.8	0.9	96.6
	No	16	75				
	Total	424	76				
Central (RHAs 6–9)	Yes	369	1	95.6	99.7	0.8	95.9
	No	17	57				
	Total	386	58				
Northern (RHAs 11–17)	Yes	340	0	96.1	100.0	0.8	96.3
	No	14	34				
	Total	354	34				
Calgary (RHA 4)	Yes	835	2	96.0	99.8	0.8	96.0
	No	35	68				
	Total	870	70				
Edmonton (RHA 10)	Yes	595	1	94.9	99.8	0.8	95.2
	No	32	66				
	Total	627	67				

Note: Three cases with unknown RHA were excluded.

TABLE 7
Birth defects not captured by the birth registry by year, Alberta, 1994–1996

Year	Defects within ICD9-XIV		Defects outside ICD9-XIV		Total defects	
	Number	Percent (%)	Number	Percent (%)	Number	Percent (%)
1994	28	2.7	5	4.7	33	2.9
1995	21	2.5	9	7.5	30	3.2
1996	63	8.0	9	11.1	72	8.3
Total	112	4.2	23	7.5	135	4.6

Table 6 presents the sensitivity, positive predictive value, kappa, and overall agreement of the diagnosis on single birth defects within ICD-9-XIV by the birth registry for each geographic region. As shown, the sensitivity is fairly close across the five regions during the study period, from 96.2% for Southern Alberta to 94.9% for Edmonton, as is the positive predictive value, from 100.0% for Northern Alberta to 99.7% for Central Alberta. As noted, the kappa value appears slightly higher in Southern Alberta (0.88) but lower in the Calgary health region (0.76). The overall agreement does not show much difference across the regions. These findings suggest that data on the diagnosis of single congenital anomalies within ICD-9 Section XIV from the birth registry are fairly good for all geographic regions.

Congenital Anomalies Not Captured by the Birth Registry

Table 7 shows the number of babies with a single birth defect who were not captured by the birth registry during the 3-year period. Overall, 135 babies (4.6%) with a single anomaly were not captured by the birth registry. The proportion of these non-captured anomalies increased, from 2.9% (33/1152) in 1994 to 3.2% (30/950) in 1995, and 8.3% (72/869) in 1996 ($p < 0.05$). When looking at the anomalies subgroups, the proportion of the not-captured cases was higher for those outside the adaptation of ICD-9 Section XIV. The majority of the anomalies outside ICD-9-XIV are of less clinical significance, such as umbilical hernia (ICD-9 = 553.10), inguinal hernia (ICD-9 = 550.90), anomalies of jaw size (ICD-9 = 524.00), etc. However, some important genetic diseases, such as hereditary hemolytic anemia (ICD-9 = 282.00), cystic fibrosis (ICD-9 = 277.00), and disorders of carbohydrate transport and metabolism (ICD-9=27100) were missing from the birth registry.

TABLE 8
Birth defects not captured by the birth registry by region, Alberta, 1994–1996

Geographic region		Defects within ICD9-XIV		Defects outside ICD9-XIV		Total defects	
		Number	Percent (%)	Number	Percent (%)	Number	Percent (%)
Southern	(RHAs 1–3,5)	16	3.8	4	5.3	20	4.0
Central	(RHAs 6–9)	17	4.4	4	6.9	21	4.7
Northern	(RHAs 11–17)	12	3.4	2	5.9	14	3.6
Calgary	(RHA 4)	35	4.0	4	5.7	39	4.2
Edmonton	(RHA 10)	31	4.9	7	10.5	38	5.5
Province		**112**	**4.2**	**23**	**7.5**	**135**	**4.6**

Note: Three cases with RHA unknown are included in the provincial total.

Table 8 summarizes the number and proportion of single anomalies non-captured by the birth registry for the five geographic regions from 1994–1996. As shown, the proportion is fairly close across the five geographic regions, although it appears slightly higher in Edmonton.

Discussion

Using administrative data sources for surveillance and research is becoming increasingly important in the health setting.[4] Understanding the completeness and accuracy of this information will assist data analysis and interpretation. This study, using the birth registry as an example, examined the completeness and accuracy of such data on congenital anomalies in Alberta, Canada.

It was found that total case counts recorded in the birth registry were on average 3% lower than those recorded in the ACASS between 1985 and 1996. The difference is fairly close for most of the years under study but is larger in 1991 (6% lower), 1994 (5% lower) and 1995 (8% lower). This difference should be taken into account in interpretation when the birth registry data are used. It is important to emphasize that the data on congenital anomalies from the two sources are expected to be the same. The observed differences can probably be attributed to the "lag period" between data collection, verification, and entry into the system, and perhaps to other sources (i.e., transcription error in updating, data and data entry errors). Staff shortages and staff change over time are likely reasons for the difference. There is also a 6–10 month delay, depending on the year, in accessing the birth registry data that may also account for some of the differences. The larger differences in the most recent two years may, in part, be attributed to the improved case ascertainment from the ACASS and a decrease in the length of hospital stay for obstetric cases.

The validity of diagnostic categories (single anomalies from the birth registry during 1994–1996) is excellent and clinically almost perfect for the 24 categories examined as well as for the overall anomalies. There is no evidence suggesting regional variations in data quality with respect to both data validity and completeness. These findings suggest the data on congenital anomalies from the birth registry in Alberta are fairly useful.

It must be emphasized that the usefulness of vital statistics for surveillance of a particular health event depends on the characteristics of that health event, as well as on the procedures used to collect, code and summarize relevant information. In general, vital statistics will be more useful for conditions that can be ascertained easily at the time of birth or death. Likewise, mortality rates derived from death certificate data will more closely approximate true incidence for conditions with a short clinical course that are easy to diagnose, are easily identified as initiating a chain of events leading to death, and are usually fatal.

The majority of birth defects can be easily identified at birth. The Physician Notice of Livebirth or Stillbirth used in a birth registry may serve as an important source for case identification. If the data on congenital anomalies in the Alberta birth registry have reached such a high quality, it is because they are all obtained from the ACASS, demonstrating that a close tie between the birth registry and the ACASS is required to maintain this level. Without the use of Physician Notice of Livebirth or Stillbirth in the birth registry, some cases may be not initially identified and may miss attention from the ACASS. However, without a well-established mechanism of case identification, verification, confirmation, coding, and entry into a data system from the ACASS, which then sends the data to the birth registry, the case registration in the province may not be as complete and accurate. The rich information and the high quality of birth defect data collected by the Alberta birth registry (Appendix 1), and perhaps of other provinces with similar mechanisms of case ascertainment, would suggest its potential value for surveillance and research of congenital anomalies. This study also found that some factors such as the lag period, errors in data entry and transcription – common in administration of all birth registries – appear to have little impact on the completeness and accuracy of case registration in a birth registry. Of note, although the birth and death certificates are filed shortly after the event occurs, the process of producing final vital statistics at a provincial level from these data sources can take at least eight months or longer.

This exercise has demonstrated an approach to evaluating an administrative database, and may help future evaluation studies of secondary data. In fact, a similar approach is being used in another study comparing congenital anomalies data between physician claims, hospital morbidity and the ACASS.

It is important to keep in mind, however, that this study also has important limitations. Although comparing data from several sources has been used and recommended by others,[1-4] the data from the reference source are assumed to be valid. Data errors in the reference group may lead to some variations in the level of the validity measures, though this error is likely very small for the ACASS data. There are hundreds of specific anomalies and the present study evaluated only 24 diagnostic categories, so the potential differences among specific anomalies within each diagnostic category are not revealed by the present study. Many fetal anomalies, especially those of less than 20 weeks' gestation, were not captured by the birth registry/ACASS in the past, and about 20% of cases with multiple anomalies were not included in this evaluation.

Nonetheless, findings from this study lead to the following conclusions:

- The data on congenital anomalies from the birth registry represent about 97% of total infants with a single birth defect during 1985–1996. For infants with multiple anomalies, the "minor" defects were not available in the birth registry during this period. Thus, the total number of defects from the birth registry was likely underreported by about 20%.

- Most of the diagnostic categories of single congenital anomalies (within ICD-9 Section XIV) from the birth registry agree with those from the ACASS during 1994–1996, suggesting accuracy of diagnostic information for the 24 categories examined. Overall, 99.8% of single anomalies in the birth registry during 1994 to 1996 were likely "true" cases. During the same period, the probability of correctly classifying the single anomalies within ICD-9 Section XIV was 95.7%. Clinically, the validity of the diagnosis for single anomalies within ICD-9 Section XIV was almost perfect during the three-year period.

- No significant regional variations were found with respect to the completeness of information, sensitivity, predictability, kappa, and overall agreement of diagnosis on single anomalies within ICD-9 XIV as a whole during 1994–1996.

- The ACASS has played an essential role in surveillance of congenital anomalies in the province. Further collaboration between Alberta Health and Wellness, the ACASS, the birth registry and other agencies is required for the surveillance, prevention and control of congenital anomalies in the province.

It is recommended that data on congenital anomalies from the birth registry, in combination with data from the ACASS and other sources, such as Alberta Health Care Insurance Plan (AHCIP), physician claims, and hospital morbidity, be used for monitoring/surveillance purposes such as:

- monitoring long-term trends of birth prevalence of selected congenital anomalies;

- identifying differences in infant health status within ethnic (Aboriginal vs. Non-Aboriginal) or other subgroups of the population;

- assessing differences in congenital anomalies, stillbirths, low birth weight by geographic area (spatial pattern) or by maternal age or other factors;

- monitoring congenital anomalies that are generally considered preventable (such as neural tube defect);

- generating hypotheses regarding possible causes or correlations of congenital anomalies or other infant health indicators;

- conducting health-planning activities related to infant health; and

- monitoring progress toward achieving improved health status of the infant or child population.

Acknowledgements

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APPENDIX 1
List of selected variables available in the birth registry

Information	Variable description	Comments
Personal identification	Mother's name (full)	Complete, assist data linkage
	Mother's date of birth	Complete, assist maternal age and birth cohort calculation, and data linkage
	Mother's AHCIP/PHN	Incomplete in the past, assist data linkage
	Father's name (full)	Incomplete, assist data linkage, etc.
	Father's date of birth	Incomplete, paternal age estimation
	Name of newborn (full)	Complete and inaccurate, assist linkage
	Newborn's date of birth	Complete and accurate for age calculation, the birth cohort development, data linkage, etc.
	Birth registration number	Complete and accurate for linkage
	Newborn's sex	Complete and accurate, data linkage and report
Demographic and socioeconomic information	Mother's residence PC[a]	Incomplete, assist linkage and spatial comparison
	Mother's residence SGC[b]	Complete, assist linkage and spatial comparison
	Mother's marital status	Incomplete, SES indicator
	Mother's residence RHA[c]	Complete, assist regional comparison
Reproductive history	History of stillbirth	Complete and accurate, risk indicator
	History of abortion	Complete and accurate, risk indicator
	Number of live birth	Complete and accurate, risk indicator
	Single or multiple births	Complete and accurate, risk indicator
	Birth order	Complete and accurate, risk indicator
Behavioral risk factors	Maternal smoking	Useful for prevalence and risk estimation
	Alcohol use	Useful for prevalence and risk estimation
	Street drug use	Useful for prevalence and risk estimation
Medical interventions	Prenatal visit	Useful for percent and frequency estimation
	Hospital of delivery	Complete, assist linkage and analysis
	Assisted labour	Incomplete, proportion of assistance by type
Selected pregnancy outcomes	Stillbirth	Complete, perinatal health indicator
	Cause of stillbirth/death	Complete, perinatal health indicator
	Congenital anomalies	Incomplete, perinatal health indicator
	Birth weight	Complete, perinatal health indicator
	Gestational age (weeks)	Incomplete, perinatal health indicator
	Length of newborn	Complete, risk indicator
	Apgar score	Complete, risk indicator

[a] PC = postal code
[b] SGC = standard geographic code
[c] RHA = regional health authority

Sensitivity is a measure of the probability of correctly diagnosing/classifying a case or event (Syn: true positive rate).

Specificity is a measure of the probability of correctly identifying/classifying a non-case or non-event (Syn: true negative rate).

Positive Predictive Value: In screening and diagnostic tests, the probability that a person/event with a positive test is a true positive.

Overall agreement is a measure of the probability of correctly diagnosing/classifying a case or event plus a non-case or non-event.

Kappa is a measure of the degree of non-random agreement between observers or measurements of the same categorical variable.

These measures have been widely used in screening tests and interclass or intraclass correlations. The calculations are illustrated by the following two by two table:

Table layout for sensitivity, specificity, predictive value, and agreement analysis

		Cases or events from the standard		
		Yes	**No**	**Total**
Cases or events being classified from the study	Yes	a (90)	b (15)	**a + b**
	No	c (10)	d (25)	**c + d**
Total		**a + c**	**b + d**	**a + b + c + d = N**

Sensitivity = a / (a + c) * 100 = 90 / (90 + 10) * 100 = 90.0%

Positive Predicative Value (PV) = a / (a + b) * 100 = 90 / (90 + 15) * 100 = 85.7%

Specificity = d / (b + d) * 100 = 25 / (15 + 25) * 100 = 40.0%

Overall Agreement = (a + d) / N * 100 = (90 + 25) / 140 * 100 = 82.1%

Kappa = $P_0 - P_e$ / 1 – P_e = [2(ad – bc)] / {[(a + b)(b + d)] + [(a + c)(c + d)]}

= 2(90*25 – 15*10) / (105*40 + 100*35) = 4200 / 7700 = 0.54

where P_0 is the observed agreement and P_e is the expected agreement.

Status Report

The Development of the National Diabetes Surveillance System (NDSS) in Canada

Clarence Clottey, Frank Mo, Barbara LeBrun, Phillip Mickelson, Jeff Niles and Glenn Robbins

Introduction

In Canada, health care is primarily the constitutional responsibility of the provinces and territories. Consequently, considerable information related to the provision of health services is available at that level of government. At the present time, only limited use is being made of these data to assess the impact of chronic diseases such as diabetes on the population of Canada.[1] This article describes an innovative means of using these administrative data banks to gather local and national epidemiological data on diabetes.

Background

In this information era and in light of a prevailing commitment to support evidence-based decision making in health, the Government of Canada is investing resources in the improvement of the national capacity to systematically collect, analyze and disseminate accurate health information.

Canada does have some rudimentary data on diabetes from disparate sources, including investigations relying on self-reported diabetes in surveys, and on mortality and hospitalization data. At present, it is not possible to consistently track diabetes-associated complications and deaths in Canada. Research has shown that current morbidity and mortality records underestimate the burden of diabetes.[2] Although diabetes kills and maims primarily through its complications, diabetes complications data are not linked to other relevant databases. This lack of systematic prevalence and incidence data limits our ability to plan and evaluate prevention and control programs for this disease. The availability of accurate, comprehensive data is an important requirement of good public health policy: accurate data on diabetes will assist in projections on its burden to the health care system.

The National Forum on Health[1] encouraged the use of existing administrative databases to further chronic diseases research in Canada. One such example is the National Diabetes Surveillance System (NDSS), which is being funded as part of the Canadian Diabetes Strategy (CDS). The CDS is a Government of Canada initiative launched in November 1999, funded for $115 million over 5 years. Of this, $10.8 million has been directed to the development of the NDSS.

Governance of the National Diabetes Surveillance System

The NDSS is the result of five years of collaboration between government and non-government bodies, and this is reflected in its governance. This partnership involves Health Canada, other federal agencies, including Statistics Canada, all provincial and territorial governments, national Aboriginal groups, academic researchers, and non-government health organizations (NGOs). Of the NGOs, the Canadian Diabetes Association has been an active partner in the development of the NDSS from the very beginning, and has mobilized private-sector sponsorship, most notably that of SmithKline Beecham. Other national NGOs with a significant interest in diabetes are represented through their membership in the multi-sectoral Diabetes Council of Canada (DCC), which promotes and participates in the NDSS.
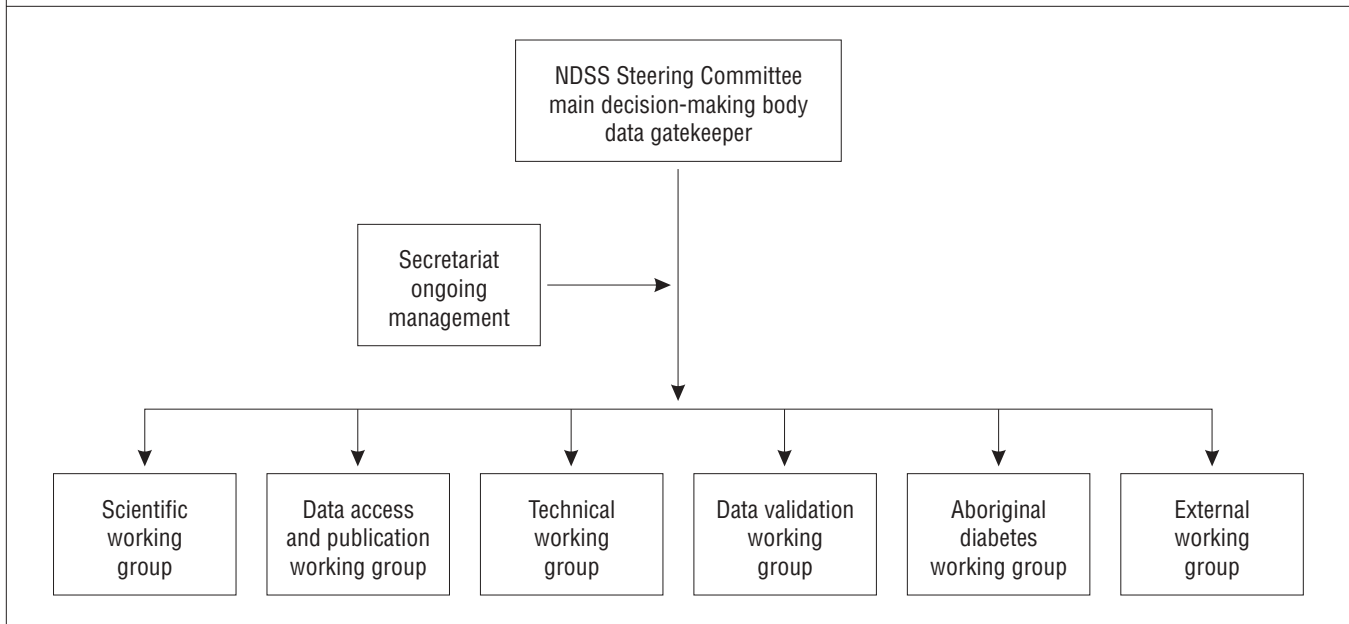
The NDSS Steering Committee is headed by the Chair of the DCC and represents all partners. It meets twice a year to monitor progress and to plan strategic directions. A secretariat is responsible for administrative and operational matters. Working groups address such key issues as data validation, data quality, database access and publication, review of the scientific direction and Aboriginal issues. Health Canada coordinates and funds the activities of the working groups.

FIGURE 1
Multi-stakeholder governance in the NDSS

```
                    ┌─────────────────────────────┐
                    │   NDSS Steering Committee    │
                    │   main decision-making body  │
                    │       data gatekeeper        │
                    └─────────────────────────────┘
                                  │
        ┌──────────────┐          │
        │  Secretariat │          │
        │   ongoing    │─────────▶│
        │  management  │          │
        └──────────────┘          │
                                  ▼
   ┌──────┬──────────┬──────────┬──────────┬──────────┬──────┐
   ▼      ▼          ▼          ▼          ▼          ▼
```

Scientific working group	Data access and publication working group	Technical working group	Data validation working group	Aboriginal diabetes working group	External working group

Goal of the National Diabetes Surveillance System

The NDSS will develop provincial and territorial surveillance capacities to establish a national standardized database for disseminating comparative data on rates of diabetes and its complications in Canada. This will ultimately lead to a better understanding of the magnitude of the diabetes burden in Canada.

Rationale for the National Diabetes Surveillance System

To date, there is no national diabetes surveillance system in Canada. There have been some smaller investigations that relied on self-reported diabetes in surveys and on mortality and hospitalization data. This has, however, led to an underestimation of the burden of diabetes for various reasons, including the unreliability of self-reporting methods, bias in the small sample sizes in surveys and the inability to track diabetes-associated complications and deaths in Canada. The use of provincial and territorial administrative health databases will improve our ability to measure the incidence and prevalence of diabetes and establish an ongoing diabetes surveillance system in Canada.

The National Diabetes Surveillance System Model

To maximize the use of existing data sources before creating new ones, the NDSS initially focuses on using provincial and territorial administrative databases, such as health insurance registry, medical claim and hospital discharge data for standardized diabetes surveillance.

No other national level health surveillance initiative has been based on these data sources to date, but the rationale for this approach in the study of diabetes should be apparent.

The clinical path of diabetes, from detection to treatment and comorbidity management, makes it possible for the diabetes burden to be tracked through various client interactions with the provincial and territorial health care systems. Tracking can begin with physician visits (where diabetes is typically diagnosed and treated in the early stages), and followed, using the health insurance registry and/or vital statistics, through hospitalizations for diabetes-associated conditions (complications), procedures such as amputations and, ultimately, death. This system provides estimates of rates and rate ratio for populations with and without diabetes. Epidemiological and statistical models will be used in the comparative analysis of health services utilization for populations who have, and do not have, diabetes. Standardized methods also will be used in the determination of diabetes prevalence, incidence and mortality across provinces and territories.

There are well-recognized precedents in parts of Canada for utilizing administrative data to provide diabetes-related information. Manitoba Health, for example, has published a number of articles in peer-reviewed journals that report on diabetes incidence and prevalence rates in the province using the health insurance registry, physician claims and hospital services.[3]

Demonstration Project

The feasibility of the NDSS model and its implement-ation has been explored through a demonstration project undertaken in Manitoba, Saskatchewan and Alberta, in partnership with the University of Alberta. Matching funds for the project were awarded through Health Canada's Health Infostructure Support Program. This demonstration project documented some early deliver-ables for the NDSS in the areas of methodological innovation and standardization of diabetes surveillance across the three provinces.

Challenges

In exploring the use of provincial and territorial data for national diabetes surveillance, issues of data privacy and ownership are constant challenges. Implementation of the NDSS for the continuing surveillance of diabetes will involve linking already existing provincial and ter-ritorial administrative databases to develop longitudinal profiles of individual cases identified by unique health insurance numbers. For the purpose of privacy, these newly created longitudinal records will not contain names of people – only non-nominal case identifiers. Any person-identifying information remains in the province or territory where it is protected by corresponding data privacy legislation. Provincial or territorial data sent to Health Canada or any other NDSS partner will be non-nominal and will be in grouped, or aggregate, format.

Next Steps

As the capacity to use administrative data for diabetes surveillance becomes well established across the country over the next few years, the NDSS will likely broaden the reach of the surveillance by integrating or coordinating with other complementary sources of health information.

Some of these sources could include national and com-munity health risk-factor surveys. It is envisioned that the NDSS will need to identify some important areas for in-depth community-level investigation when existing administrative data does not provide sufficient information.

Conclusion

The NDSS has taken on the dual challenges of address-ing the critical information gaps in our knowledge of the frequency and effects of diabetes in Canada and respond-ing to the very real and important concerns related to an individual's right to privacy and an institution's right to control the use of data it has collected. It must also address provincial and territorial concerns about the transfer of data beyond jurisdictional boundaries.

This project represents an important advance in chronic disease epidemiology and health service research in Canada. Health Canada is confident that the country-wide capacity building, the maintenance of partnerships and the improvement in the comprehensiveness of diabetes information represented by the NDSS initiative will ensure cautious but steady progress over the next few years towards an era of truly evidence-based decision-making.

References

1. National Forum on Health (Canada). *Canadian Health Action: building on the legacy*. Ottawa, 1997.
2. Health Canada. *Diabetes in Canada. National statistics and opportunities for improved surveillance, prevention, and control*. Ottawa, 1999; 14–15.
3. Blanchard JF, Ludwig S, Wajda A, Dean H, and Anderson K. Incidence and prevalence of diabetes in Manitoba, 1986–1991. *Diabetes Care* 1996; 19: 807–11. ∎

Book Review
Peer Review in Health Sciences

Edited by Fiona Godlee and Tom Jefferson
London (England): BMJ Publishing Group, 1999;
271 pp; ISBN 0-7279-1181-3; $88.95 (CDN) through the Canadian Medical Association

The main message of *Peer Review in Health Sciences* is based on the observation that despite its critical importance to science, the peer review process is flawed and has remained "remarkably untouched by the rigours of science". The book is a call to action to improve peer review, consider alternatives and increase research into the review process.

The two editors, Fiona Godlee, a scientific editor from the *British Medical Journal*, and Tom Jefferson, from the Surrey and Cochrane Centre in the United Kingdom, have brought together data and the views of many editors and researchers interested in peer review to give a description of the current state of peer review and reflections on its future directions.

In the first section, Drummond Rennie, a scientific editor at the *Journal of the American Medical Association*, co-director of a Cochrane Centre and organizer of three international congresses on peer review, masterfully identifies the main problems with peer review.

It is unreliable, unfair and fails to validate or authenticate; it is unstandardized and open to bias; blinded peer review invites malice, either from ad hominem attacks on the author or by facilitating plagiarism; it stifles innovation; it lends spurious authority to reviewers; reviewers knowledgeable enough to review a study are often competitors, and therefore have a conflict of interest; and it causes unnecessary delays in publication.

To this sobering list is added, in a chapter on the peer review of grant applications, the observation that "the most important question to be asked ... is whether or not [peer review] assists scientists in making important discoveries that stand the test of time. We do not know."

Some may wonder – in light of the depth and breadth of these difficulties – why there is peer review at all. A chapter on the state of the evidence for journal peer review concludes that "It is the only [viable] system we currently have" and as other authors point out, the alternatives, either a free-for-all or audits, look worse.

Some optimists may be tempted to say "Peer review may not be perfect. So what?" noting that while these problems exist, they do not appear to be rampant, so in general the system continues to work. The answer to this is in the chapter on bias, subjectivity, chance and conflict of interest: if the studies that get funding and the studies that are published are subject to a flawed review system, this leads to publication bias. They then note: "publication bias is perhaps one of the more important practical and ethical issues currently facing biomedical journals."

In a much shorter section on the future of peer review, the editors agree that peer review is here to stay, but could be improved. Clinical trial registries would help to stem the tendency to publish only positive trials, but this will not improve the review process. Electronic communications can, and likely will, affect future peer review. In some disciplines, such as physics, authors post electronic drafts of their manuscripts, called "eprints", for comment prior to formal publication. The *Medical Journal of Australia* experimented with electronic posting and the authors were given the opportunity to make revisions prior to the print publication. This journal's editors have also tested online peer review and note that one of its advantages is that it allows interactivity among reviewers, authors and the editor.

A central issue in peer review is exactly how the evaluation is carried out. This has remained largely discretionary. Although most journals and granting agencies use checklists, it is the reviewer comments that carry much more weight. Teaching the peer review process is a surprisingly new concept. In a tongue-in-cheek interview near the end of the book, "Socrates" is engaged in a discussion on the problems of peer review, then poses this rhetorical question regarding some research results on the subject: "You asked untrained people to do what you concede is a difficult job. And then you went to the trouble of carrying out a study which showed that they weren't very good at it?"

In an attempt to address this problem, the book also includes a "how to" section. This includes a researchers' guide to the peer review of grant applications, an authors' guide to editorial peer review, a reviewers' guide to peer review, and an editors' guide to setting up a peer review system. Although this is a useful section, the changing target audience for each chapter, as well as the contrast in the target audience for this section (mostly neophytes) with respect to the rest of the book, which targets seasoned editors and researchers, may seem

somewhat disjointed to readers who read the book from cover to cover.

Peer Review in Health Sciences is a credible, fairly expensive, paperback reference text that identifies the major issues in peer review today and summarizes the problems and dearth of evidence to support this critical practice. Its biggest contribution is in highlighting the humbling fact that the peer review process is far from perfect and that continued efforts are needed to improve it.

Overall rating: Good

Strengths: This book is an excellent overview, bringing together the views of prominent editors, researchers and funders to summarize the current problems and potential for peer review in the health sciences.

Weaknesses: Its inclusiveness is both a strength and a weakness. It covers such very basic topics as what a neophyte author needs to know about peer review as well as such advanced topics as what seasoned editors, funders and researchers need to know on the debates that drive research on peer review. It is stronger in identifying current issues than in recommending future directions.

Audience: There are many audiences for this book, identified as "those involved in peer review and those who have been judged by it".

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Health Products and Food Branch
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Calendar of Events

September 4–7, 2001 **Atlanta, Georgia, USA**	"Using Science to Build Comprehensive Cancer Programs: A 2001 Odyssey" US Department of Health and Human Services Centers for Disease Control and Prevention 2001 Cancer Conference	Kathleen Carey Conference Co-chair E-mail: kcarey@cdc.gov <www.cdc.gov/cancer/conference2001>
September 22–25, 2001 **Sydney, Australia**	4th International Conference on the Scientific Basis of Health Services Sydney Convention and Exhibition Centre – Darling Harbour	<www.icsbhs.org>
October 18–21, 2001 **Saskatoon, Saskatchewan**	"Health Research in Rural and Remote Canada: Taking the Next Steps" Second Scientific Conference and Annual Meeting of The Rural Health Research Consortium	The Rural Health Research Consortium 103 Hospital Drive PO Box 120, RUH Saskatoon, SK S7N 0W8 Tel.: (306) 966-7888 Fax: (306) 966-8378 E-mail: bizetto@sask.usask.ca <www.usask.ca/medicine/agmedicine>
October 28–31, 2001 **Niagara Falls, Ontario**	"New Frontiers in Health Information" First joint US/Canadian Case Mix and Quality Assurance Conference	Jeannie Lasquinha Event Manager Tel.: (416) 979-7474 Fax: (416) 979-1144 E-mail: info@casemix2001.com <www.casemix2001.com>
November 5–7, 2001 **Kitchener, Ontario**	"Harvesting Health: Embracing Tradition and Change" 52nd Annual Ontario Public Health Association Conference	Marc Xuereb Conference Co-ordinator 99 Regina Street South Waterloo, ON N2J 4V3 Tel.: (519) 883-2004 x 5765 Fax: (519) 883-2241 E-mail: xmarc@region.waterloo.on.ca <www.opha.on.ca/conference/index.html>
November 18–21, 2001 **Ottawa, Ontario**	"Child and Youth Health: Action, Research and Advocacy" 8th Canadian Conference on International Health	CCIH Secretariat Tel.: 1-877-722-4140 (Outside Canada: 1-613-722-4140) x 224 E-mail: ccih@ag-cdn.com <www.csih.org/index_e.html>
November 29– **December 1, 2001** **Toronto, Ontario**	Canada's Fifth National Conference on Asthma and Education (ASED 5)	A. Les McDonald, Executive Director Canadian Network for Asthma Care (CNAC) 1607 – 6 Forest Laneway North York, ON M2N 5X9 Tel.: (416) 224-9221 Fax: (416) 224-9220 E-mail: ased@cnac.net <www.cnac.net>

12–14 December, 2001 Paris, France	"Dynamic Management of Health and Safety in the Construction Industry: Practicable Solutions" XXVIth International Symposium of the International Section for the Prevention of Occupational Risks in the Construction Industry	Jacques Tonner CRAMIF – Secrétariat du Colloque AISS-BTP 17-19, place de l'Argonne F-75019 Paris France Tel.: (33) 1 40 05 38 02 Fax: (33) 1 40 05 38 84 E-mail: construction.issa@ cramif.cnamts.fr <www.cramif.fr>
February 27–March 1, 2002 Atlanta, Georgia, USA	"Cultivating Healthier Communities Through Research, Policy and Practice" 16th National Conference on Chronic Disease Prevention and Control	Terrye Hornsby Tel.: (301) 588-6000 x 270 Fax: (301) 588-2106 E-mail: thornsby@kevric.com <www.cdc.gov/nccdphp/conference>

In Memoriam

Andrés Petrasovits

February 11, 1937 – July 24, 2001

We mourn the untimely passing of our friend and colleague, Dr. Andrés Petrasovits. His 30-year career in Health Canada spanned the history of thinking and methods for health promotion. Since 1990 he had been Senior Advisor of the Heart Health/WHO Collaborating Centre within Health Canada, and Co-Director of Countrywide Integrated Noncommunicable Diseases Intervention (CINDI) Canada, in which capacity he pioneered and guided the Canadian Heart Health Initiative through its various phases. He was recognized internationally for his tireless commitment and substantive contributions to heart health and chronic disease prevention.

Besides being an outstanding professional and a learned scholar, Andrés was a truly gentle man who treated everyone he met with great kindness and respect. The loss to his friends and colleagues of his deep knowledge, experience and wisdom, and of his gentle presence, are incalculable.

To inquire about donating to a research fund in his name, please contact the Heart and Stroke Foundation of Canada:

222 Queen St., suite 1402
Ottawa, Ontario K1P 5V9
Telephone: (613) 569-4361
Fax: (613) 569-3278

Occupational Surveillance in Canada: Cause-Specific Mortality Among Workers, 1965–1991 (Aronson KJ, Howe GR, Carpenter M, Fair ME) (Catalogue No. 84-546-XCB; $500)

Statistics Canada has released an easy-to-use bilingual CD-ROM entitled *Occupational Surveillance in Canada: Cause-Specific Mortality Among Workers, 1965–1991*. This product describes the impact of 670 different occupations on 70 different causes of death for women and men from a 27 year follow-up of 700,000 workers (approximately 10% of the labour force between 1965 and 1971).

This valuable tool provides the information you need to:

- detect previously unsuspected potential associations between occupation and death;

- target areas for future studies on occupational mortality risks among women and men;

- adjust employment conditions to protect employees from potential danger; and

- carry out a wide variety of research projects.

The data can be reviewed and used in prepared tables (in EXCEL) or can be manipulated from an ASCII file into the spreadsheet of your choice.

Order information

Available for $500 plus applicable taxes in Canada or US$500 plus shipping charges outside Canada. Order toll-free **by phone** at 1 800 267-6677, **by fax** at 1 877 287-4369, **by mail** from Statistics Canada, Dissemination Division, Circulation Management, 120 Parkdale Avenue, Ottawa, Ontario K1A 0T6 or **by e-mail** at order@statcan.ca.

For further information, see http://www.statcan.ca/english/IPS/Data/84-546-XCB.htm

CDIC: Information for Authors

Chronic Diseases in Canada (CDIC) is a peer-reviewed scientific journal published four times a year. Contributions are welcomed from outside of Health Canada as well as from within this federal department. The journal's focus is the prevention and control of non-communicable diseases and injuries in Canada. This may include research from such fields as epidemiology, public/community health, biostatistics, behavioural sciences and health services. CDIC endeavours to foster communication about chronic diseases and injuries among public health practitioners, epidemiologists and researchers, health policy planners and health educators. Submissions are selected based on scientific quality, public health relevance, clarity, concise-ness and technical accuracy. Although CDIC is a Health Canada publication, authors retain responsibility for the contents of their papers, and opinions expressed are not necessarily those of the CDIC Editorial Committee or of Health Canada.

FEATURE ARTICLES

Regular Feature Articles: Maximum 4,000 words for main text body (excluding abstract, tables, figures, references) in the form of original research, surveillance reports, meta-analyses, methodological papers, literature reviews or commentaries

Short Reports: Maximum 1,200 words (as above)

Status Reports: Describe ongoing national programs, studies or information systems at Health Canada (maximum 3,000 words)

Workshop/Conference Reports: Summarize workshops, etc. organized or sponsored by Health Canada (maximum 3,000 words)

Cross-country Forum: For authors outside of Health Canada to exchange information from research or surveillance findings, programs under development or program evaluations (maximum 3,000 words)

ADDITIONAL ARTICLE TYPES

Letters to the Editor: Comments on articles recently published in CDIC will be considered for publication (maximum 500 words)

Book/Software Reviews: Usually solicited by the editors (500–1,300 words), but requests to review are welcomed

SUBMITTING MANUSCRIPTS

Submit manuscripts to the Editor-in-Chief, *Chronic Diseases in Canada*, Population and Public Health Branch, Health Canada, Tunney's Pasture, CDIC Address Locator: 0602C3, Ottawa, Ontario K1A 0L2.

Since CDIC adheres in general (section on illustrations not applicable) to the **"Uniform Requirements for Manuscripts Submitted to Biomedical Journals"** as approved by the International Committee of Medical Journal Editors, authors should refer to this document for complete details before submitting a manuscript to CDIC (see **<www.cma.ca/publications/mwc/uniform.htm>** or *Can Med Assoc J* 1997;156(2):270–7).

Checklist for Submitting Manuscripts

☐ **Cover letter:** Signed by all authors, stating that all have seen and approved the final manuscript and have met the authorship criteria of the Uniform Requirements and including a full statement regarding any prior or duplicate publication or submission for publication

☐ **First title page:** Concise title; full names of all authors and institutional affiliations; name, postal and e-mail addresses, telephone and fax numbers for corresponding author; separate word counts for abstract and text

☐ **Second title page:** Title only; start page numbering here as page 1

☐ **Abstract:** Unstructured (one paragraph, no headings), maximum 175 words (100 for short reports); include 3–8 **key words** (preferably from the Medical Subject Headings (MeSH) of *Index Medicus*)

☐ **Text:** Double-spaced, 1 inch (25 mm) margins, 12 point font size

☐ **Acknowledgements:** Include disclosure of finan-cial and material support in acknowledgements; if anyone is credited in acknowledgements with substantive scientific contributions, authors should state in cover letter that they have obtained written permission

☐ **References:** In "Vancouver style" (consult Uniform Requirements and a recent CDIC issue for examples); numbered in superscript (or within parentheses) in the order cited in text, tables and figures; listing up to 6 authors (first 3 and "et al." if more); *without any automatic reference numbering feature used in word processing*; any unpublished observations/ data or personal communications used (discouraged) to be cited in the text in parentheses (authors responsible for obtaining written permission); authors are responsible for verifying accuracy of references

☐ **Tables and Figures:** Each on a separate page and in electronic file(s) separate from the text *(not imported into the text body)*; as self-explanatory and succinct as possible; not duplicating the text, but illuminating and supplementing it; not too numerous; numbered in the order that they are mentioned in the text; explanatory material for tables in footnotes, identified by lower-case superscript letters in alphabetical order; figures limited to graphs or flow charts/templates (no photographs), with software used specified and titles/footnotes on a separate page

☐ **Number of copies:** Four complete copies, including tables and figures; 2 copies of any related supplementary material