Status report

National Epidemiologic Database for the Study of Autism in Canada (NEDSAC)

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In 2001, the Autism Spectrum Disorders—Canadian-American Research Consortium (ASD-CARC) launched a program of research on autism spectrum disorders (ASD). As part of that undertaking, and in response to concerns about the growing proportion of children diagnosed with ASD,[1,2] the National Epidemiologic Database for the Study of Autism in Canada (NEDSAC; www.nedsac.ca) was created as a multi-site ASD surveillance program. Government departments, clinicians and researchers* collaborated to establish regional teams to collect information (see Table 1) on children with ASD in British Columbia, Calgary (Alberta), Manitoba, southeastern Ontario,† Prince Edward Island, and Newfoundland and Labrador. NEDSAC provides estimates of the prevalence of ASD in Canadian children and a profile of those who are affected,[3] and allows researchers to monitor trends in age at diagnosis.[4] These data can help the health, education and social services sectors with planning and allocation of resources.[5,6]

Estimating the prevalence of ASD

Different approaches are used to estimate the prevalence of ASD. One is to conduct population screening to identify suspected or diagnosed cases, followed by an assessment process to confirm or rule out the diagnosis. This approach is exemplified by a study done in Karlstad, Sweden: investigators used a combination of procedures to screen 826 children born there in 1985 and still living there in 1992.[7] The children were observed in

TABLE 1
Data collected in NEDSAC

Demographics of children with ASD
• Date of birth and sex
• Number of biological siblings
• Number of biological siblings with confirmed or suspected ASD
• Mother's place of residence during pregnancy
• Parental ages at birth of child
• Ethnocultural identity
Diagnosis
• Type of professional(s) who made the diagnosis
• Tests or tools that were used
• Diagnostic subgroup
• Date of diagnosis
• Child's place of residence when diagnosed

Abbreviations: ASD, autism spectrum disorders; NEDSAC, National Epidemiologic Database for the Study of Autism in Canada

* A list of researchers and collaborators can be viewed at www.nedsac.ca under "Who we Are."

† Includes the six counties of Hastings, Prince Edward, Lennox & Addington, Frontenac, Leeds & Grenville and Lanark.

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various settings, and their parents and teachers were interviewed to assign a diagnosis. This approach has the potential to capture undiagnosed cases and allows for direct assessment to verify case status. However, low response rates, with potentially biased estimates, are a concern.[8] Moreover, it is too costly to use such an approach for ongoing surveillance in large populations.

A records-review approach is used by the Autism and Developmental Disabilities Monitoring Network in the United States.[9,10] Using standardized procedures and a common case definition, data are abstracted from health and education[‡] records and reviewed by clinicians to assign case status.[9,10] This surveillance approach was deemed unfeasible in Canada: privacy legislation makes access to school records difficult, if not impossible, for researchers, and identifying cases through health sources alone could miss substantial numbers of children with ASD.[9,10]

A third approach relies on survey data to estimate the prevalence of ASD. For example, investigators in the United States analyzed responses to the 2007 National Survey of Children's Health; a child was considered to have ASD if the caregiver reported that the child had been diagnosed by a physician or other health care provider and still had the diagnosis.[11] Statistics Canada's quinquennial Participation and Activity Limitation Survey (PALS) contains a question specific to the occurrence of autism.[12] The PALS is not, however, a general population survey; its target population comprises individuals who responded "yes" to either of two questions on the Census concerning limitations of activity. Moreover, the Federal Government recently announced that it will no longer be conducting the PALS (Statistics Canada, personal communication, 30 June 2011).

A question on whether a health professional has ever diagnosed the child with autism was added to Cycle 8 (2008–2009) of the biennial National Longitudinal Survey of Children and Youth (NLSCY).[13]

Statistics Canada cautions that the NLSCY is a general-population survey and is not designed for the analysis of relatively rare subpopulations, which could yield small samples and high sampling error. The initial sample for Cycle 8 of the NLSCY consisted of 35 795 children aged 0 to 7 years and youth aged 14 to 25 years. Cross-sectional weights, which are used to make inferences at the population level for the survey time period, are only available for the 0- to 7-year age group. It is unclear, therefore, how valid these survey data would be for estimating the prevalence of autism in Canada, particularly if they are stratified by age group and region.

Using administrative data is another option for surveillance, although these data demonstrate imperfect sensitivity and specificity.[14] This approach is only feasible in areas where the appropriate datasets can be linked for research purposes. In 2001, few provinces and territories had an administrative dataset infrastructure that could be used for ASD surveillance. (One notable exception was Manitoba: the Manitoba Centre for Health Policy houses health and education datasets that can be linked to estimate ASD prevalence. See the section at the end of this paper titled "NEDSAC now and in the future.")

For the reasons described above, none of the aforementioned approaches was considered feasible for ASD surveillance in Canada when NEDSAC was established. Instead, we formed partnerships with agencies that provide services to children with ASD in order to identify cases in the most cost-effective manner possible. This approach, similar to the one that uses administrative datasets to estimate prevalence, is apt to result in under-detection of cases for a number of reasons. First, the methods used by agencies to identify cases may mean that some children with ASD are missed. For example, in many education databases only one special education code is assigned in a given year, and so a child with ASD who also falls under another special education category may be coded under that category instead.[15] Service-based data also fail to

capture children who meet research criteria for ASD but who have not been diagnosed. Across all Autism and Developmental Disabilities Monitoring Network sites, the proportion of children with a previously documented classification of ASD was lower than prevalence estimates obtained using the Network's surveillance methodology and case definition.[9,10] Similarly, findings from a population-based study in Olmsted County, Minnesota, revealed that only 46.8% of children who met research criteria for having ASD had a previous diagnosis.[16] Thus, the findings from NEDSAC should be interpreted as minimum prevalence estimates.

In contrast to relying on administrative datasets to estimate prevalence, however, identifying children with ASD through agencies that provide services to this population provides greater assurance that cases are "true positives" in the sense of having received a clinical diagnosis of ASD. It also enables the research team to contact families in order to directly assess and confirm the diagnosis in a subset of individuals.

Identifying children with ASD for NEDSAC

A considerable challenge with our surveillance approach is the use of provincial services such as health, education and publicly funded intensive behavioural intervention programs to identify cases. The delivery of these services, the ease of accessing data on children with ASD (i.e. whether information on these children is readily retrievable through service providers' databases or whether they have to review files to identify cases), and internal policies regarding data sharing vary widely across provinces and service providers. Accordingly, it proved impossible to use the same case ascertainment and data collection method in every region. Instead, regional protocols were designed to capture diagnosed cases of ASD in the most efficient way possible and to meet the information needs of agencies that provide the data (the latter explains the different age cut-offs among regions, as described below). Surveillance

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[‡] In 10 of 14 surveillance sites in 2002[9] and 6 of 11 surveillance sites in 2006[10].

was rolled out in 2002 and 2003 in six regions, and it continues in Manitoba, southeastern Ontario, Prince Edward Island, and Newfoundland and Labrador. Data collection was terminated in Calgary in 2006 and British Columbia in 2007. In the following sections, we describe the case ascertainment and data collection protocols in the surveillance regions.

Table 2 summarizes these for the four regions where surveillance is ongoing.

In Prince Edward Island, the Department of Education and Early Childhood Development provides minimal data[§] to NEDSAC on preschoolers and school children aged less than 18 years diagnosed with ASD, including those who are home-schooled or attend private school. In Newfoundland and Labrador, the Department of Education and the Department of Health and Community Services provide minimal data[§] to NEDSAC on children aged less than 15 years diagnosed with ASD.[**] Children who are home-schooled or attend private school are not captured unless they are identified through the regional diagnostic teams. In

TABLE 2
Agencies in Prince Edward Island, Newfoundland and Labrador, southeastern Ontario and Manitoba that identify children with an autism spectrum disorder (ASD) for the National Epidemiologic Database for the Study of Autism in Canada

Region	Agency	Population served	Case ascertainment and data collection
Prince Edward Island	Department of Education and Early Childhood Development	Pre-school and school-aged children (includes those who are home-schooled or attend private school)	The Department of Education and Early Childhood Development provides the research team with the date of birth, sex, prevalence year[a] and diagnostic subgroup for children < 18 years with an ASD, and sends information letters and consent forms to the parents or legal guardians. If a signed consent form is sent back to the research team, more detailed demographic and diagnostic information is collected by telephone interview with the parent or legal guardian.
Newfoundland and Labrador	Department of Education	School-aged children (excludes those who are home-schooled or attend private school)	Similar to above, except limited to children < 15 years
	Department of Health and Community Services (Regional diagnostic teams operating in the four Regional Health Authorities, Intervention Services)	Collectively, all ages	
Southeastern Ontario[b]	Limestone District School Board	School-aged children (includes those who are home-schooled)	Same as for Newfoundland and Labrador
	Upper Canada District School Board		
	Hastings & Prince Edward District School Board		
	Algonquin & Lakeshore Catholic District School Board		
	Catholic District School Board of Eastern Ontario		
	Conseil des écoles publiques de l'Est de l'Ontario		
	Conseil des écoles catholiques de langue française du Centre-Est		
	Child Development Centre, Hotel Dieu Hospital[c]	Children < 18 years	
	Pathways for Children & Youth, Autism Intervention Program	Children < 18 years who have been diagnosed with autistic disorder or considered to be on the moderate to severe end of the autism spectrum	
Manitoba	Children's Special Services, Manitoba Department of Family Services and Consumer Affairs	Children < 18 years, with the exception of those living on reserves	Agency staff review files and complete data collection forms for children < 18 years with ASD

Abbreviations: ASD, autism spectrum disorder.

[a] Year in which the child was first known to have an ASD diagnosis and reside in the surveillance region.

[b] Includes the six counties of Hastings, Prince Edward, Lennox & Addington, Frontenac, Leeds & Grenville and Lanark.

[c] Referral and assessment centre for children in southeastern Ontario with suspected developmental problems.

[§] Date of birth, sex, prevalence year (year in which the child was first known to have an ASD diagnosis and reside in the surveillance region), diagnostic subgroup.

[**] At the time of initial contact with the family or at the time of diagnosis, the regional diagnostic teams in Newfoundland and Labrador advise families of the NEDSAC study and the types of data provided to the researchers. In 2009, school principals in Newfoundland and Labrador sent a letter to parents and legal guardians of all school age children with ASD describing the NEDSAC project and the data that are collected. Parents and legal guardians were informed that they could contact the Department of Education directly to opt out of having any information on their child provided to the research team. To date, five parents/legal guardians have opted out.

Ontario, it is not possible to access special education data through the Ministry of Education, making province-wide surveillance unfeasible. The catchment area is therefore restricted to southeastern Ontario[††] where the coordinating centre for NEDSAC is located. The seven public and Catholic school boards in that region provide minimal data[‡‡] to NEDSAC on children aged less than 15 years diagnosed with ASD, including home-schooled children. The agency that delivers the provincially funded intensive behavioural intervention program in southeastern Ontario also participates in NEDSAC. However, only children on the moderate to severe end of the autism spectrum are eligible for this program. To ensure that most preschoolers with an ASD diagnosis are captured, the main referral and assessment centre in southeastern Ontario for children who have a suspected developmental condition also identifies cases to NEDSAC.

In these three regions, research ethics boards and agency policies require us to obtain consent from parents or legal guardians to contact them directly for more detailed information on their child's condition. The participating agencies mail information packages about the study to families. Parents or legal guardians who want to provide more detailed demographic and diagnostic information to NEDSAC return a consent form to the research team. A researcher then collects the data by telephone. If no consent form is returned, the minimal information provided by the agencies (i.e. date of birth, sex, prevalence year, diagnostic subgroup) allows us to include all cases in our prevalence estimates.

In Manitoba, Children's Special Services, a government program that supports children with special needs throughout the province (excluding those on native reserves, which fall under federal jurisdiction), identifies cases of ASD among children

aged less than 18 years to NEDSAC. Data collection in Manitoba differs from the three other regions: agency staff at Children's Special Services provide the demographic and diagnostic information that is collected from parents or legal guardians who return a consent form in southeastern Ontario, Prince Edward Island, and Newfoundland and Labrador.

All children identified through the agencies mentioned above would have been diagnosed by a qualified health professional (e.g. developmental pediatrician, psychologist, psychiatrist). We confirmed the diagnosis in a sample of children within the four regions, using examiners who had obtained research reliability in administering the Autism Diagnostic Interview-Revised[17] (ADI-R) and the Autism Diagnostic Observation Schedule-Generic[18] (ADOS-G), two reference-standard tools for diagnosing ASD. Of 145 children assessed, 96.6% met the "autism" cut-off on the ADI-R or the "ASD" cut-off on the ADOS-G. The Metropolitan Atlanta Developmental Disabilities Surveillance Program reported a similarly high proportion of children with a previous ASD diagnosis who met surveillance criteria for having ASD (98%).[1]

In 2002, three tertiary provincial referral and assessment agencies in Vancouver[§§] undertook a chart review to identify children with ASD aged less than 15 years. Information on diagnosed cases continued to be collected this way until 2007, at which time data collection for British Columbia ceased. From 2003 to 2006, the Developmental Clinic at Alberta Children's Hospital in Calgary and various community sources in that city also provided data to NEDSAC on children with ASD aged less than 15 years. Although the British Columbia and Calgary data are not used to estimate prevalence because of incomplete case capture in those regions, they contribute

to our understanding of the epidemiology of ASD in Canada (see *Correlates of age at diagnosis of autism spectrum disorders in six Canadian regions* in this issue).[19]

In both Prince Edward Island and Newfoundland and Labrador, the potential for identifying an individual more than once (which could result in duplicate records in the database and hence an overestimation of the number of children with ASD) is minimal. In Prince Edward Island, only one agency provides data to NEDSAC. In Newfoundland and Labrador, representatives from the two participating agencies meet to resolve potential duplicates prior to sending the information to the NEDSAC coordinating centre.[***] In Manitoba, although only one agency participates in NEDSAC, seven regional offices complete data collection forms. Thus, if a family moves within the province, two regional offices could potentially submit data collection forms for the same child. The chance that a child is identified more than once is highest in southeastern Ontario, where multiple agencies participate in NEDSAC.

Regardless of the region, when information on a new case is entered in the database, an algorithm searches for records from the same region with matching date of birth (including reversed day and month of birth) and first two letters of the child's surname and first name[†††] (including transposed letters; i.e. if "abcd" is entered, the algorithm searches for exact matches and "cdab"). If potential duplicates are detected at this stage, the reporting agencies are contacted to ascertain whether the information they provided is accurate. If the reporting agencies confirm no errors in the information provided and it is reasonable to assume that the information refers to the same child,[‡‡‡] no new record is created and the existing record is tagged as a suspected duplicate case.

[††] Includes the six counties of Hastings, Prince Edward, Lennox & Addington, Frontenac, Leeds & Grenville and Lanark.

[‡‡] Date of birth, sex, prevalence year (year in which the child was first known to have an ASD diagnosis and reside in the surveillance region), diagnostic subgroup.

[§§] The Provincial Autism Resource Centre, Department of Pediatrics, Sunnyhill Health Centre for Children; The Provincial Programme in Medical Genetics, Department of Medical Genetics; and The Division of Child and Adolescent Psychiatry, Department of Psychiatry, BC Children's and Women's Health Centre, University of British Columbia.

[***] Individuals' names are not shared between departments during this process.

[†††] "xxxx" is entered for cases from southeastern Ontario, Prince Edward Island, and Newfoundland and Labrador whose parents did not return a consent form.

[‡‡‡] For example, the search algorithm finds an existing record with the same date of birth and first two letters of the surname and first name, the data are provided by two school boards in southeastern Ontario, and one of these reports that the child is no longer with that board.

Thus, when there is the possibility of a potential duplicate, only one record is created in NEDSAC.

NEDSAC now and in the future

Although data collection has ceased in British Columbia and Calgary, we continue to collect information on children with ASD in Manitoba, southeastern Ontario, Prince Edward Island, and Newfoundland and Labrador. We are currently analyzing the 2003–2008 data for these four regions. While inter-regional variations need to be interpreted with caution, given the different case ascertainment protocols, the findings will comprise the first population-based Canadian data on changes in ASD prevalence within a particular region over a six-year period. Agencies that provide services to individuals with ASD can use these data for planning and resource allocation.

In 2009, our group received funding from the Public Health Agency of Canada to evaluate the feasibility of linking datasets housed at the Manitoba Centre for Health Policy to supplement or replace our current surveillance protocol in that province. We are also collaborating with researchers in Quebec to explore the potential for linking datasets in that province to identify children with ASD. Various other initiatives, such as Population Data BC and the Child and Youth Data Lab in Alberta, could make population-based surveillance of ASD using administrative data a viable option in other areas of the country. The use of administrative data for surveillance purposes does present challenges in terms of data quality, as noted earlier. However, there are ways to deal with such challenges. For example, case definitions based on administrative data have been validated for conditions such as inflammatory bowel disease[20] and diabetes.[21] A group of investigators in Nova Scotia recently compared how accurately various combinations of diagnostic codes in three administrative health datasets identified children with ASD.[14] Using diagnoses made by the Autism Team at the IWK Health Centre in Halifax as the reference standard, the sensitivity ranged from 11.9% to 69.3% and the specificity from 77.3% to 97.7%. Linking education datasets with health ones, rather than relying on health sources alone, would likely improve detection rates (i.e. sensitivity).[9,10,14]

Conclusion

Linking administrative datasets is a cost-effective option that could allow us to expand ASD surveillance to more regions of the country. In light of the public health importance of this group of disorders,[5] NEDSAC will continue to evolve and to provide information for policy makers, families and advocates on the occurrence of ASD in Canada.

Acknowledgments

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