

Trends in Autism Prevalence: Diagnostic Substitution Revisited

Helen Coo · Hélène Ouellette-Kuntz · Jennifer E. V. Lloyd · Liza Kasmara ·
Jeanette J. A. Holden · M. E. Suzanne Lewis

Published online: 2 November 2007
© Springer Science+Business Media, LLC 2007

Abstract There has been little evidence to support the hypothesis that diagnostic substitution may contribute to increases in the administrative prevalence of autism. We examined trends in assignment of special education codes

to British Columbia (BC) school children who had an autism code in at least 1 year between 1996 and 2004, inclusive. The proportion of children with an autism code increased from 12.3/10,000 in 1996 to 43.1/10,000 in 2004; 51.9% of this increase was attributable to children switching from another special education classification to autism (16.0/10,000). Taking into account the reverse situation (children with an autism code switching to another special education category (5.9/10,000)), diagnostic substitution accounted for at least one-third of the increase in autism prevalence over the study period.

H. Coo · H. Ouellette-Kuntz (✉)
Department of Community Health and Epidemiology, Queen's University, c/o Ongwanada Resource Centre, 191 Portsmouth Avenue, Kingston, ON, Canada K7M 8A6
e-mail: oullette@post.queensu.ca

H. Ouellette-Kuntz · J. J. A. Holden
Department of Psychiatry, Queen's University, Ongwanada, Kingston, ON, Canada

J. E. V. Lloyd
Edudata Canada, Faculty of Education, University of British Columbia, Vancouver, BC, Canada

Present Address:
J. E. V. Lloyd
Human Early Learning Partnership (HELP), Vancouver, BC, Canada

L. Kasmara · M. E. S. Lewis
Department of Medical Genetics, University of British Columbia, Vancouver, BC, Canada

Present Address:
L. Kasmara
Department of Human Development, Teachers College, Columbia University, New York, NY, USA

J. J. A. Holden
Department of Physiology, Queen's University, Ongwanada, Kingston, ON, Canada

M. E. S. Lewis
Department of Medical Genetics, BC Children's and Women's Health Center, Child & Family Research Institute, Vancouver, BC, Canada

Keywords Autism · Autistic disorder · Diagnostic substitution · Prevalence · Edudata Canada · British Columbia Ministry of Education

The prevalence of autism spectrum disorders (ASDs)—which include autistic disorder, pervasive developmental disorder-not otherwise specified and Asperger disorder (American Psychiatric Association, 1994)—has increased markedly over the past few decades (Fombonne 2003; Newschaffer and Curran 2003). The reasons for this increase are a matter of debate: some argue that this group of disorders is becoming more common (Blaxill 2004), while others attribute the increase to broadening of diagnostic criteria, greater awareness among parents and professionals, and improved case-finding methods (Fombonne 2003; Charman 2002; Wing and Potter 2002).

Special education data provide a cost-effective way to examine changes over time in the prevalence of ASDs. However, such data often include only the child's primary diagnosis, and it has been suggested that diagnostic substitution—whereby children with multiple diagnoses are categorized differently over time (Caronna and Hall

2005)—may partially account for apparent increases in prevalence when using administrative data to study trends (Volkmar et al. 2004). Diagnostic substitution may also occur when children are given a different diagnosis in the present than they would have been given in the past, due to changing referral and diagnostic practices (Newschaffer 2006; Shattuck 2006).

There is currently little evidence to support the hypothesis that diagnostic substitution is contributing to recent increases in ASD prevalence. One group of investigators reported that the prevalence of “full-syndrome autism” among children receiving services through the California Department of Developmental Services increased from 5.8 to 14.9 per 10,000 over eight successive birth cohorts, while the prevalence of mental retardation decreased from 28.8 to 19.5 per 10,000 over the same time period. They concluded that part of the increase in prevalence was due to the reclassification of children from the mental retardation to the autism category (Croen et al. 2002). However, the authors retracted diagnostic substitution as a possible explanation for the increase (Croen and Grether 2003) after other researchers questioned their interpretation of the findings (Blaxill et al. 2003). Another group of investigators analyzed data from the Minnesota Department of Children, Families & Learning, which collects information each year from school districts on all children who receive state-funded special education services. They found that the prevalence of ASDs among children 6–11 years of age increased from 3 per 10,000 in 1991/1992 to 52 per 10,000 in 2001/2002. Over the same time period the prevalence of other major special education disability categories also increased, with the exception of severe mental handicap, which decreased slightly from 24 to 23 per 10,000 (Gurney et al. 2003). Similarly, a study that used special education data to examine national autism prevalence trends from 1992 to 2001 among American children 6–17 years of age found no evidence for diagnostic substitution (Newschaffer et al. 2005). In contrast, an analysis of changes in the administrative prevalence of autism and other special education classifications in the United States among children 6–11 years of age suggested that diagnostic substitution contributed to the increasing administrative prevalence of autism from 1994 to 2003 (Shattuck 2006).

All the studies cited above have relied on aggregate data, and have therefore been limited to group-level comparisons. Examining whether individual children switched special education classifications would provide more direct evidence for or against the hypothesis that diagnostic substitution is partially responsible for observed increases in autism prevalence. Using British Columbia Ministry of Education data provided to us by Edudata Canada,¹ we

explored trends in the assignment of special education codes to children 4–9 years of age who had an autism code in at least 1 year between 1996 and 2004, inclusive. Our main objective was to determine what proportion of children switched from other special education classifications to the autism category during the study period, and to quantify the contribution of diagnostic substitution to changes in the administrative prevalence of autism between 1996 and 2004.

Methods

Data Source and Study Population

British Columbia (BC) is located on the west coast of Canada. With a population of 4.3 million in 2006, it is Canada’s third most populous province. Officials from all BC public and independent (private) schools are required to provide their school district offices with enrollment and demographic information for each child in their respective schools (including home-schooled students) as of September 30 each year. The school district offices then forward this information to the BC Ministry of Education for tracking and funding purposes. As part of this effort, the Ministry required schools to begin assigning and reporting special education codes in September 1996. Children can be assigned only one special education code per school year.

Schools receive different funding amounts per student, depending on the child’s special education needs. A standard amount is allocated for each child enrolled. This allocation covers the costs of providing learning supports for students who are identified under the special education categories of “Learning Disability”, “Mild Intellectual Disability”, “Moderate Behaviour Disorder” or “Gifted”. Additional supplementary funding is provided for children who are identified with other special education needs. Level 1 supplementary funding includes students who are classified as “Physically Dependent” or “Deaf and Blind”. These students are funded at the highest level. Level 2 supplementary funding includes students who are classified under the categories “Moderate to Severe/Profound Intellectual Disability”, “Physical Disability or Chronic Health Impairment”, “Visual Impairment”, “Deaf or Hard of Hearing”, or “Autism”. Level 3 students—which include those requiring intensive behaviour interventions or with a serious mental illness—receive the lowest amount of supplementary funding (British Columbia Ministry of Education 2002). As only one special education code can be assigned each year, in cases where a child meets the requirements for two or more special education categories, the school generally assigns the code that will yield the most funding.

Edudata Canada is a research centre at the University of British Columbia. Funded by the various levels of

¹ <http://www.edudata.ca>

government in Canada, it supports education research in BC and other regions of the country. The BC Ministry of Education makes its datasets available to Edudata Canada, which handles all Ministry data brokering. As part of a national study examining the prevalence of ASDs, we entered into a research agreement with the Ministry of Education/Edudata Canada to obtain information on children born on or after January 1, 1987 who had a special education code of “G” (=“autism code”) as of September 30 in at least 1 year between 1996 and 2004, inclusive (the study period). In 1996, the year in which special education codes were first collected by the BC Ministry of Education, children born in 1987 would have been 9 years old. Very few children enter the school system before age 4 years. Accordingly, we restricted the current analysis to children 4–9 years of age. The dataset included date of birth, sex and year in which the child was first assigned an autism code, as well as the child’s special education code for each year of the study period. We were also provided counts of children 4–9 years of age who were enrolled in the school system on September 30 of each year of the study period.

According to BC Ministry of Education guidelines, “autism” refers to a diagnosis of autistic disorder, thus excluding other ASDs (British Columbia Ministry of Education Special Programs Branch 2000). A diagnosis by a qualified professional (which includes a paediatrician, psychiatrist or registered psychologist, or a developmental assessment team at one of three centres in the province) is required for a child to be assigned an autism code. On January 1, 2004, the BC government introduced standards and guidelines for the assessment and diagnosis of children with ASDs (British Columbia Ministry of Education, n.d.). Prior to this, assessment and diagnostic practices varied across the province. During the time period covered by the current analysis, children with an ASD who did not meet the criteria for autistic disorder may have been identified under the special education category that most closely matched the type and intensity of educational intervention required, including one of the behaviour disorder or intellectual disability categories, “Learning Disability” or “Physical Disability or Chronic Health Impairment” (British Columbia Ministry of Education Special Programs Branch 2000). In October 2006, the BC Minister of Education announced that funding would be provided for all students diagnosed with an ASD (British Columbia Ministry of Education 2006a), and the special education category of “Autism” was changed to “Autism Spectrum Disorder”.

Analysis

The follow-up period comprised all years from 1996 to 2004 that a child was between 4 and 9 years of age and

enrolled in the BC school system. Children were divided into three groups, depending on their pattern of code assignment over the follow-up period. Group 1 comprised children who had an autism code for each year of follow-up. Group 2 included children who had autism as the only special education classification during the follow-up period, but who, in one or more years, also had no special education code assigned. This may have occurred before an autism code was initially assigned, subsequent to assignment of an autism code, or in a few instances a child may have been assigned an autism code in 1 year, followed by no special education code, and then at some point during the follow-up period he or she may have again been assigned an autism code. Group 3 consisted of children who had, in addition to an autism code in one or more years, a special education code other than autism in at least 1 year during the follow-up period. We calculated the proportion of children in each of these groups, as well as the median years of follow-up.

We estimated the point prevalence of autism for each year of the study period by including in the numerator any child 4–9 years of age with an autism code on September 30 (the prevalence date), and using the total school population of 4- to 9-year-olds on the prevalence date as the denominator. We then quantified the contribution of various factors, defined below, to the change in prevalence per 10,000 from year to year (see Fig. 1).

Diagnostic Substitution

The number of children with a special education code other than autism in the previous year who had an autism code on the following year’s prevalence date, divided by the total school population of 4- to 9-year-olds on the prevalence date, and multiplied by 10,000.

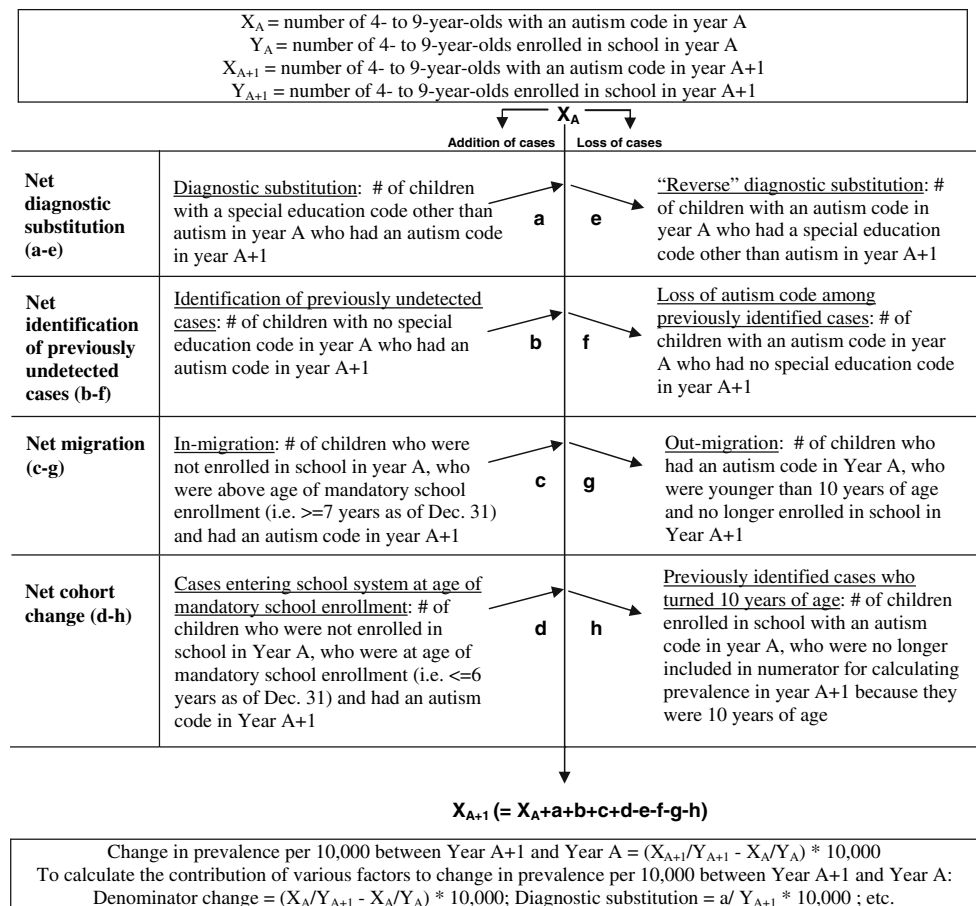
“Reverse” Diagnostic Substitution

The number of children with an autism code in the previous year who had a different special education code recorded on the following year’s prevalence date, divided by the total school population of 4- to 9-year-olds on the prevalence date, and multiplied by 10,000.

Identification of Previously Undetected Cases

The number of children enrolled in school with no special education code in the previous year who had an autism code on the following year’s prevalence date, divided by the total school population of 4- to 9-year-olds on the prevalence date, and multiplied by 10,000.

Fig. 1 Diagram illustrating method used to quantify the contribution of various factors to changes in the administrative prevalence of autism among British Columbia school children 4–9 years of age



Loss of Autism Code among Previously Identified Cases

The number of children with an autism code in the previous year who had no special education code recorded on the following year’s prevalence date, divided by the total school population of 4- to 9-year-olds on the prevalence date, and multiplied by 10,000.

In-Migration

The number of children who were not registered in school in the previous year who, on the following year’s prevalence date, were above the age of mandatory school enrollment² and had an autism code, divided by the total school population of 4- to 9-year-olds on the prevalence date, and multiplied by 10,000.

² Generally, children in British Columbia are required to enroll in an educational program “...on the first day of a school year if, on or before December 31 of that school year, the person will have reached the age of 5 years.” However, parents may opt to defer enrollment of their child until the first school day of the next school year (British Columbia Ministry of Education 2006b). Accordingly, we considered a child to be at the age of mandatory school enrollment if he or she was 6 years of age or under as of December 31, and above the age of mandatory school enrollment if he or she was 7 years of age or over as of December 31 of a given year.

Out-Migration

The number of children with an autism code in the previous year who, on the following year’s prevalence date, were under 10 years of age but no longer enrolled in school, divided by the total school population of 4- to 9-year-olds on the prevalence date, and multiplied by 10,000.

Cases Entering School System at Age of Mandatory School Enrollment (See footnote 2)

The number of children who were not registered in school in the previous year who, in the following year, were 6 years of age or younger as of December 31 and had an autism code on the prevalence date, divided by the total school population of 4- to 9-year-olds on the prevalence date, and multiplied by 10,000.

Previously Identified Cases who Turned 10 Years of Age

The number of children enrolled in school in the previous year with an autism code, who were no longer included in the numerator on the following year’s prevalence date because they were 10 years of age, divided by the total

school population of 4- to 9-year-olds on the prevalence date, and multiplied by 10,000.

Denominator Change

The change in prevalence that results if the number of children with an autism code in the previous year remains the same in the following year, but the total number of children enrolled in school changes (i.e., the numerator used to calculate prevalence is the same in both years, but the denominator is different). Calculated by dividing the number of children with an autism code in the previous year by the total school population of 4- to 9-year-olds on the following year's prevalence date, minus the prevalence of autism in the previous year, and multiplied by 10,000.

We summed the results across the years to determine each factor's contribution to the change in autism prevalence over the study period. We then calculated the net contribution of diagnostic substitution to the change in prevalence by subtracting the total results for "reverse" diagnostic substitution from those for diagnostic substitution. We repeated this for identification of previously undetected cases (identification of previously undetected cases minus loss of autism code among previously identified cases), migration (in-migration minus out-migration) and cohort change (cases entering school system at the age of mandatory school enrollment minus previously identified cases who turned 10 years of age).

Results

A total of 2,198 children 4–9 years of age were assigned an autism code in at least 1 year during the follow-up period. Table 1 shows what proportion of these children were included in each of the three groups described in the Analysis section, as well as the median length of follow-up. These statistics are also reported for subgroupings of the Group 2 and Group 3 children. Among the subgroupings of children who had autism as the only special education code, but who were not assigned any special education code in at least 1 year of the follow-up period (Group 2), the autism code was assigned for 785 (52.6%) of the 1,492 person-years of total follow-up, while no special education code was recorded for 707 person-years (47.4%).

Among the subgroupings of children who had a special education code other than autism recorded in at least 1 year of follow-up (Group 3), no special education code was recorded for 295 person-years (12.8%), an autism code was recorded for 1,005 person-years (43.5%), and a special education code other than autism was recorded for 1,010 person-years (43.7%). Two special education classifications ("Moderate to Severe/Profound Intellectual Disability" and "Physical Disability or Chronic Health Impairment")

accounted for slightly more than one-half of the person-years of follow-up during which a special education code other than autism was assigned, while "Severe Behaviour" accounted for 16.8% (Table 2).

Table 3 shows counts and proportions of children with an autism code in each year of the study period, as well as counts and proportions of children categorized under a variety of factors (described in the Analysis section) that contributed to changes in the administrative prevalence of autism from year to year, and over the study period. The point prevalence of autism increased by 30.8 per 10,000 over the study period, from 12.3 per 10,000 in 1996 to 43.1 per 10,000 in 2004. Children with another special education code who were then assigned an autism code in the following year accounted for 16.0 per 10,000 (51.9%) of the total increase in prevalence over the study period. In contrast, children with an autism code in 1 year who were then assigned another special education code in the following year accounted for a decrease in autism prevalence of 5.9 per 10,000. Accordingly, the net contribution of diagnostic substitution to the increase in autism prevalence from 1996 to 2004 was 10.1 per 10,000, or 32.8%. The net impact of identification of previously undetected cases, migration, cohort change and denominator change on the overall change in autism prevalence was 10.7 per 10,000 (34.7%), -1.2 per 10,000 (-3.9%), 8.0 per 10,000 (26.0%) and 3.2 per 10,000 (10.4%), respectively (Table 3).

Discussion

Since autistic disorder is a chronic, nonfatal condition that is diagnosable by 3 years of age (Lord and Risi 2001), the prevalence should theoretically remain stable throughout the school-age years if: a) all cases are identified at an early age; b) there is no differential migration to or from the area (i.e., the numbers of children with and without autism migrating to the area is equal to the numbers with and without autism leaving the area); c) the incidence of autism remains unchanged; and d) the diagnostic criteria and their implementation remain unchanged. In BC however, the prevalence of autism among school children 4–9 years of age increased 3.5-fold—an absolute increase of 30.8 per 10,000—over a 9-year period.

The BC data reveal that not all children with autism are identified upon entering school and assigned an autism code at that time. A large percentage (45.5%) of the total increase in prevalence over the study period was due to identification of what we have termed "previously undetected cases", or children who had no special education code in the previous year who were assigned an autism code in the following year. We were unable to assess the reasons for this, but it is possible that these children were diagnosed only after they had started school. Although autistic disorder is diagnosable at

Table 1 Pattern of special education code assignment among British Columbia school children 4–9 years of age who had an autism code in at least one year of the study period (1996–2004)

	<i>n</i> (%)	Median length of follow-up, ^a person-years (IQ range)
Group 1 (autism code during each year of follow-up)	1,322 (60.1)	3 (3)
Group 2 (autism code only, but no special education code assigned in at least one year of follow-up)	359 (16.3)	4 (2)
No special education code initially; once autism code assigned, it was retained for remainder of follow-up period	289 (80.5)	4 (2)
Autism code initially, then lost code in a subsequent year and no special education code assigned for remainder of follow-up period	49 (13.6)	5 (1)
Pattern of code assignment not straightforward ^b	21 (5.8)	5 (1.5)
Group 3 (autism code in at least one year of follow-up, and other special education code in at least one year of follow-up)	517 (23.5)	5 (1)
Special education code other than autism assigned prior to autism code (may also have had no special education code in one or more years); once autism code assigned, it was retained for remainder of follow-up period	348 (67.3)	5 (1)
Autism code assigned prior to another special education code (may also have had no special education code in one or more years); once another special education code assigned, it was retained for remainder of follow-up period	98 (19.0)	4 (2)
Pattern of code assignment not straightforward ^c	71 (13.7)	5 (0)

IQ: Interquartile

Bolded values are percentages based on within-group totals

^a The follow-up period comprised all years between 1996 and 2004, inclusive, during which a child was 4–9 years of age and enrolled in school in British Columbia

^b Pattern of code assignment does not fall into either of the other two categories listed for the group (e.g., child may have had an autism code in Year X, no special education code in Year Y, then an autism code in Year Z, etc.)

^c Pattern of code assignment does not fall into either of the other two categories listed for the group, but 32 (6.2%) ended the period of follow-up with an autism code; 30 (5.8%) with a special education code other than autism; and 9 (1.7%) with no special education code

Table 2 Frequency of special education code assignment (excluding autism) among British Columbia school children 4–9 years of age who had an autism code and at least one other special education code assigned between 1996 and 2004 (Group 3; *n* = 517)

Special education classification	Person-years of follow-up ^a during which code was assigned <i>N</i> (%)
Moderate to severe/profound intellectual disability	300 (29.7)
Physical disability or chronic health impairment	275 (27.2)
Severe behaviour	170 (16.8)
Learning disability	103 (10.2)
Behaviour disorder—Moderate and rehabilitation	77 (7.6)
Mild intellectual disability	53 (5.2)
Physically dependent	23 (2.3)
Deaf or hard of hearing	5 (0.5)
Gifted	3 (0.3)
Visual impairment	1 (0.1)
Total	1010 (100.0)

^a The follow-up period comprised all years between 1996 and 2004, inclusive, during which a child was 4–9 years of age and enrolled in school in British Columbia

3 years, in practice children may not be identified until later (Howlin and Asgharian 1999; Lingam et al. 2003; Mag-nússon and Sæmundsen 2001; Mandell et al. 2002; Wiggins et al. 2006). Under-identification in past years has been suggested as one reason why the prevalence of autism appears to be increasing (Gurney et al. 2003; Ouellette-Kuntz et al. 2007).

The proportion of the prevalence that was accounted for by children being assigned an autism code upon entering the school system at the age of mandatory school enrollment did, however, generally increase over the study period (from 4.1 per 10,000 in 1997 to 6.6 per 10,000 in 2004). Net cohort change, based on the difference between this proportion and the proportion of children who were

Table 3 Contribution of various factors to changes in the administrative prevalence of autism among British Columbia school children 4–9 years of age (1996–2004)

	1996	1997	1998	1999	2000	2001	2002	2003	2004	Total (1996–2004)
Number of 4- to 9-year-olds with autism code	316	409	472	590	696	777	853	933	1,003	-
Number of 4- to 9-year-olds enrolled in school	257,089	260,182	258,075	255,838	251,790	247,987	242,643	238,488	232,945	-
Prevalence of autism per 10,000	12.3	15.7	18.3	23.1	27.6	31.3	35.2	39.1	43.1	-
Change in prevalence per 10,000 (current year's minus previous year's prevalence per 10,000)	-	3.4	2.6	4.8	4.6	3.7	3.8	4.0	3.9	30.8
<i>Count of 4- to 9-year-olds with autism code in current year who:</i>										
Had an autism code in previous year	-	242	292	372	456	530	569	647	703	-
Had a special education code other than autism in previous year	-	21	28	24	66	54	71	51	78	-
Had no special education code in previous year	-	33	40	52	31	37	44	53	56	-
Were not in school in previous year, above age of mandatory school enrollment ^a in current year	-	7	10	8	11	7	9	16	12	-
Were not in school in previous year, age of mandatory school enrollment ^a in current year	-	106	102	134	132	149	160	166	154	-
<i>Count of 4- to 9-year-olds with autism code in previous year who:</i>										
Have a special education code other than autism in current year	-	15	15	21	20	15	11	14	35	-
Have no special education code in current year	-	7	10	5	12	10	14	17	7	-
Are younger than 10 years of age but no longer enrolled in school in current year	-	7	16	13	10	16	23	12	14	-
Are 10 years of age, and therefore no longer included in current year's prevalence estimate	-	45	76	61	92	125	160	163	174	-
<i>Contribution of various factors to change in autism prevalence per 10,000</i>										
Diagnostic substitution (children with a special education code other than autism in previous year who have an autism code in current year)	-	0.8	1.1	0.9	2.6	2.2	2.9	2.1	3.3	16.0
"Reverse" diagnostic substitution (children with an autism code in previous year who have a special education code other than autism in current year)	-	-0.6	-0.6	-0.8	-0.8	-0.6	-0.5	-0.6	-1.5	-5.9
Net diagnostic substitution (diagnostic substitution minus "reverse" diagnostic substitution)	-	0.2	0.5	0.1	1.8	1.6	2.5	1.6	1.8	10.1
Identification of previously undetected cases (children with no special education code in previous year who have an autism code in current year)	-	1.3	1.5	2.0	1.2	1.5	1.8	2.2	2.4	14.0
Loss of autism code among previously identified cases (children with autism code in previous year who have no special education code in current year)	-	-0.3	-0.4	-0.2	-0.5	-0.4	-0.6	-0.7	-0.3	-3.3
Net identification of previously undetected cases (identification of previously undetected cases minus loss of autism code among previously identified cases)	-	1.0	1.2	1.8	0.8	1.1	1.2	1.5	2.1	10.7
In-migration (children not registered in school in previous year who are above age of mandatory school enrollment ^a and have an autism code in current year)	-	0.3	0.4	0.3	0.4	0.3	0.4	0.7	0.5	3.2
Out-migration (children with an autism code in previous year who are younger than 10 years of age in current year, but no longer registered in school)	-	-0.3	-0.6	-0.5	-0.4	-0.6	-0.9	-0.5	-0.6	-4.5
Net migration (in-migration minus out-migration)	-	0.0	-0.2	-0.2	0.0	-0.4	-0.6	0.2	-0.1	-1.2

Table 3 Contribution of various factors to changes in the administrative prevalence of autism among British Columbia school children 4–9 years of age (1996–2004)

	1996	1997	1998	1999	2000	2001	2002	2003	2004	Total (1996–2004)
Cases entering school system at age of mandatory school enrollment ^a (children not registered in school in previous year who are at age of mandatory school enrollment ^a and have an autism code in current year)	-	4.1	4.0	5.2	5.2	6.0	6.6	7.0	6.6	44.7
Previously identified cases who turned 10 years of age (children registered in school in previous year with an autism code who are no longer included in current year's prevalence because they are 10 years of age)	-	-1.7	-2.9	-2.4	-3.7	-5.0	-6.6	-6.8	-7.5	-36.7
Net cohort change (cases entering school system at age of mandatory school enrollment ^a minus previously identified cases who are 10 years of age)	-	2.3	1.0	2.9	1.6	1.0	0.0	0.1	-0.9	8.0
Denominator change	-	-0.1	0.1	0.2	0.4	0.4	0.7	0.6	0.9	3.2

^a Generally, children in British Columbia are required to enroll in an educational program "...on the first day of a school year if, on or before December 31 of that school year, the person will have reached the age of 5 years." However, parents may opt to defer enrollment of their child until the first school day of the next school year (British Columbia Ministry of Education 2006b). Accordingly, we considered a child to be at the age of mandatory school enrollment if he or she was 6 years of age or under as of December 31, and above the age of mandatory school enrollment if he or she was 7 years of age or over as of December 31 of a given year

previously included in the numerator for prevalence but were no longer included in the following year because they turned 10 years of age, accounted for 26.0% of the increase in prevalence over the study period. Differential migration of preschool children with autism into BC over the study period could be one explanation for the increase attributable to cohort change. It is also possible that alterations in diagnostic and referral patterns, or an increase in the real risk of autism, may underlie the increase attributable to this factor. We cannot examine any of these hypotheses with the data available. One interesting finding, however, is that from 1997 through 2001 the proportion of cases entering school at the age of mandatory school enrollment was higher than the proportion of cases who turned 10 and hence were no longer included in the numerator for prevalence. In contrast, in 2002 and 2003 these proportions were the same or very similar, which is what is expected if the prevalence remains stable throughout the school-age years due to early identification, no differential patterns of migration, no change in the incidence rate and no change in diagnostic criteria or practices. In 2004 the trend was reversed, and the proportion of cases entering school at the age of mandatory school enrollment was lower than the proportion of cases who turned 10 and hence were no longer included in the numerator for prevalence.

We were unable to fully assess the impact of migration on changes in the administrative prevalence of autism, as we had no information on where children were born. However, for each year we were able to calculate the number of children with an autism code who were first registered in the BC school system when they were above the age of mandatory school enrollment, as well as the number of children with an autism code who left the BC school system before the age of 10 years. We assumed that these numbers were an accurate representation of how many children moved to or from BC, as the dataset contained information on all school-age children, including those enrolled in independent schools or being home-schooled. When these numbers were converted to proportions and compared, the overall impact on change in the administrative prevalence of autism over the study period was a decrease of 1.2 per 10,000.

We were unable to quantify the full extent to which diagnostic substitution, broadly defined, contributed to the increase in autism prevalence over the study period, as it is impossible to know how many of the children diagnosed with autism in this dataset would not have received a similar diagnosis in the past due to differences in referral and diagnostic practices. Accordingly, our findings regarding diagnostic substitution should be taken as minimum estimates. Based on the number of children switching from a special education code other than autism

in a previous year to an autism code in the following year, diagnostic substitution accounted for a large percentage (51.9%) of the increase in autism prevalence over the study period. Even when “reverse” diagnostic substitution was factored in, the net contribution of diagnostic substitution to the increase in autism prevalence was 10.1 per 10,000, or 32.9% of the total increase. This finding contradicts results from several studies where there was no evidence of diagnostic substitution (Gurney et al. 2003; Newschaffer et al. 2005). One explanation for this may be that we used individual- rather than group-level data. A major limitation with the latter is that it is impossible to determine whether individual children switched classifications (Newschaffer 2006; Shattuck 2006). It may have been difficult for us to detect diagnostic substitution if we had analyzed data at the aggregate level, since, as shown in Table 2, there was no one classification that accounted for most of the non-autism special education codes assigned. Shattuck (2006) found significant decreases in the prevalence of two special education categories in the US—learning disabilities and mental retardation—around the same time that autism prevalence was increasing rapidly, and concluded that these categories were the most likely candidates for a possible diagnostic substitution effect. In the BC data, “Moderate to Severe/Profound Intellectual Disability”, which most closely corresponds to the mental retardation category in the US classification system, accounted for the largest percent (29.7%) of the total person-years of follow-up during which a special education code other than autism was assigned. In contrast, “Learning Disability” accounted for only 10.2% of this total.

We cannot state with certainty why some children with another special education code were switched to an autism code over the study period. Higher funding for some special education categories may have played a role. For example, children previously classified under “Learning Disability” or “Moderate Behaviour Disorder” may have been assigned an autism code in order to receive supplementary funding. However, the “Moderate to Severe/Profound Intellectual Disability” and “Physical Disability or Chronic Health Impairment” categories accounted for more than half the years of follow-up during which other special education codes were assigned. Since these categories are funded at the same level as the “Autism” one, it is unlikely that funding issues were behind a major portion of the switching between special education codes observed in this dataset. It is also unlikely that changes in diagnostic criteria played a major role in children switching classifications during the study period, since the DSM-IV criteria (American Psychiatric Association 1994) were in effect throughout.

It is possible that some children classified under the “Moderate to Severe/Profound Intellectual Disability”

category were switched to “Autism” because the latter may be perceived as a less stigmatizing label. It is also possible that some of the switching between classifications occurred because educators felt that a certain special education category would provide the supports that most closely matched a child’s needs in a certain year, while another category subsequently proved more beneficial for this purpose. One notable government policy that was implemented during the study period was the provision of direct funding to families of children with an ASD, to allow them to purchase intensive early behavioural treatment and intervention. This occurred in June 2002 for families of children under 6 years of age, and was extended to cover the needs of children 6–18 years of age in April 2003. This may have provided the impetus for some families to seek an ASD diagnosis for their child, which, if the diagnosis was autistic disorder, could have impacted a number of the factors that contributed to changes in the prevalence of autism, including diagnostic substitution. However, this would only have taken place towards the end of the study period.

There were some limitations to the data available to us for this study. Although the BC Ministry of Education Guidelines state that “autism” refers to a DSM-IV diagnosis of autistic disorder (British Columbia Ministry of Education Special Programs Branch 2000), standards and guidelines for the assessment and diagnosis of ASD were only introduced in 2004, and these only applied to children under the age of 6 years (British Columbia Ministry of Education n.d.). Accordingly, we have no way of knowing how many of these children were diagnosed using standardized tools. However, the main purpose of this analysis was not to estimate the prevalence of autism, but rather, to determine whether diagnostic substitution occurred over the period of the study, thus enabling us to better interpret trends that were observed in this, and perhaps other, administrative datasets. We also had no information regarding the rate of coding errors in the data. Thus, for the 4.2% of cases in which the pattern of code assignment was not straightforward, we do not know whether this was actually the pattern of code assignment, or whether there was an error in recording the child’s special education code in one or more years. Moreover, because of this apparently non-straightforward pattern of code assignment in some cases, caution is necessary when interpreting the total changes over the study period. For example, children whose records showed a pattern of coding such as “autism code in Year X, other special education code in Year Y, autism code in Year Z” would have been included in the diagnostic substitution effect, even though there was an autism code initially, and such records would also have contributed to “reverse” diagnostic substitution. Similarly, if in one year a child had an

autism code, followed by no special education code in the following year and then an autism code in the subsequent year, the record would be included under “identification of previously undetected cases” when the code assignment changed from no special education code to an autism code, even though that child had had an autism code in an earlier year. Thus, even with individual-level data it is difficult to measure precisely the impact of diagnostic substitution and identification of previously undetected cases on changes in autism prevalence. Furthermore, as the BC School Act allows parents to defer enrollment of their child for a year, it is also possible that some cases who were 6 years of age as of December 31 and who were not registered in school in the previous year may have been misclassified under “cases entering school system at the age of mandatory school enrollment” rather than “in-migration”.

A number of researchers have highlighted the complexity of interpreting changes in autism prevalence when relying on administrative data, particularly educational data (Volkmar et al. 2004; Newschaffer et al. 2005; Mandell and Palmer 2005; Laidler 2005). This is true for the BC data, where a number of factors complicate the interpretation of changes in the proportion of children assigned the special education code for autism. These factors include the apparently non-straightforward nature of code assignment in a small percent of cases; uncertainty as to whether children who were initially assigned an autism code but were then assigned another special education code actually have autism, were misdiagnosed, or were assigned a different code because that category more closely matched their special education needs; and evidence of diagnostic substitution and under-detection of cases. Our analysis reveals, however, that while diagnostic substitution and identification of previously undetected cases each accounted for about one-third of the increase in the administrative prevalence of autism from 1996 to 2004 based on BC special education data, there was a substantial percentage of the increase that could not be explained by either of these factors. With the data that were available to us, we cannot say whether this unexplained increase was an artefact due to differential migration patterns or better detection of autism in the later years of the study period, or whether some portion may represent a true increase in the occurrence of autism.

Acknowledgments We wish to thank Ms. Nancy Walt, Ms. Caroline Ponsford and Ms. Claudia Roch of the British Columbia Ministry of Education, and Dr. Victor Glickman, Director of Edudata Canada, for providing us with access to the information resources of the British Columbia Ministry of Education through Edudata Canada that were integral to this study. We also wish to thank Ms. Sarah Moffat, a Psychology student at Queen’s University, for her assistance with the preparation of this manuscript. We are grateful to the members of the

Autism Spectrum Disorders-Canadian-American Research Consortium (ASD-CARC; Jeanette J.A. Holden, Principal Investigator) for their work on the grant proposal that led to the funding of this and other ASD-CARC projects, and to other members of the ASD-CARC Epidemiology Project team for their contribution to the overall project. Members of these groups are listed on the ASD-CARC’s website at <http://www.autismresearch.ca>. This work was supported by a CIHR Interdisciplinary Health Research Team grant (RT-43820) to the ASD-CARC. Dr. Suzanne Lewis sincerely appreciates the support provided by the CIHR Institute of Genetics Clinician Investigator (2003–2005) and the Michael Smith Foundation for Health Research Scholar Programs (2005–2010). The findings and interpretations expressed herein are those of the authors, and do not necessarily reflect the opinions of the British Columbia Ministry of Education or of Edudata Canada.

References

- American Psychiatric Association. (1994). *Diagnostic and Statistical Manual of Mental Disorders (DSM-IV)* (4th ed.) Washington: American Psychiatric Association.
- Blaxill, M. F. (2004). What’s going on? The question of time trends in autism. *Public Health Reports*, *119*, 536–551.
- Blaxill, M. F., Baskin, D. S., & Spitzer, W. O. (2003). Commentary: Blaxill, Baskin, and Spitzer on Croen et al. (2002) The changing prevalence of autism in California. *Journal of Autism and Developmental Disorders*, *33*, 4920.
- British Columbia Ministry of Education. (2002). Ministry policy site. Policy document: K-12 funding—Special needs. Retrieved April 23, 2007 from http://www.bced.gov.bc.ca/policy/policies/funding_special_needs.htm
- British Columbia Ministry of Education. (n.d.). Special education services: A manual of policies, procedures and guidelines. Retrieved April 30, 2007 from http://www.bced.gov.bc.ca/specialed/ppandg/planning_11.htm
- British Columbia Ministry of Education. (2006a). Province funds students with autism spectrum disorder. Retrieved April 23, 2007 from http://www2.news.gov.bc.ca/news_releases_2005–2009/2006EDU0104-001205.htm
- British Columbia Ministry of Education. (2006b). Manual of school law K to 12 (online). Retrieved November 5, 2006 from <http://www.bced.gov.bc.ca/legislation/schoollaw/>
- British Columbia Ministry of Education Special Programs Branch. (2000). Teaching students with autism. A resource guide for schools. [RB0102]. Retrieved December 4, 2006 from <http://www.bced.gov.bc.ca/specialed/docs/autism.pdf>
- Caronna, E. B., & Hall, A. J. (2005). Dipping deeper into the reservoir of autistic spectrum disorder. *Archives of Pediatric and Adolescent Medicine*, *157*, 619–621.
- Charman, T. (2002). The prevalence of autism spectrum disorders: Recent evidence and future challenges. *European Child and Adolescent Psychiatry*, *11*, 249–256.
- Croen, L. A., & Grether, J. K. (2003). Response: A response to Blaxill, Baskin, and Spitzer on Croen et al. (2002), “The changing prevalence of autism in California”. *Journal of Autism and Developmental Disorders*, *33*, 227–229.
- Croen, L. A., Grether, J. K., Hoogstrate, J., & Selvin, S. (2002). The changing prevalence of autism in California. *Journal of Autism and Developmental Disorders*, *32*, 207–215.
- Fombonne, E. (2003). The prevalence of autism. *The Journal of the American Medical Association*, *289*, 87–89.
- Gurney, J. G., Fritz, M. S., Ness, K. K., Sievers, P., Newschaffer, C. J., & Shapiro, E. G. (2003). Analysis of prevalence trends of

- autism spectrum disorder in Minnesota. *Archives of Pediatric and Adolescent Medicine*, 157, 622–627.
- Howlin, P., & Asgharian, A. (1999). The diagnosis of autism and Asperger syndrome: Findings from a survey of 770 families. *Developmental Medicine & Child Neurology*, 41, 834–839.
- Laidler, J. R. (2005). US Department of Education data on “autism” are not reliable for tracking autism prevalence. *Pediatrics*, 116, e120–e124.
- Lingam, R., Simmons, A., Andrews, N., Miller, E., Stowe, J., & Taylor, B. (2003). Prevalence of autism and parentally reported triggers in a north east London population. *Archives of Disease in Childhood*, 88, 666–670.
- Lord, C., & Risi, S. (2001). Diagnosis of autism spectrum disorders in young children. In A. M. Wetherby & B. M. Prizant (Eds.), *Autism spectrum disorders: A transactional developmental perspective* (pp. 11–30). London: Paul H Brookes.
- Magnússon, P., & Sæmundsen, E. (2001). Prevalence of autism in Iceland. *Journal of Autism and Developmental Disorders*, 31, 153–163.
- Mandell, D. S., Listerud, J., Levy, S. E., & Pinto-Martin, J. A. (2002). Race differences in the age at diagnosis among Medicaid-eligible children with autism. *Journal of the American Academy of Child & Adolescent Psychiatry*, 41, 1447–1453.
- Mandell, D. S., & Palmer, R. (2005). Differences among states in the identification of autistic spectrum disorders. *Archives of Pediatric and Adolescent Medicine*, 159, 266–269.
- Newschaffer, C. J. (2006). Investigating diagnostic substitution and autism prevalence trends [commentary]. *Pediatrics*, 117, 1436–1437.
- Newschaffer, C. J., & Curran, L. K. (2003). Autism: An emerging public health problem. *Public Health Reports*, 118, 393–399.
- Newschaffer, C. J., Falb, M. D., & Gurney, J. G. (2005). National autism prevalence trends from United States special education data. *Pediatrics*, 115, e277–e282.
- Ouellette-Kuntz, H., Coe, H., Lloyd, J. E. V., Kasmara, L., Holden, J. J. A., & Lewis, M. E. S. (2007). Trends in special education code assignment for autism: Implications for prevalence estimates. *Journal of Autism and Developmental Disorders*. doi: 10.1007/s10803-007-0478-x.
- Shattuck, P. T. (2006). The contribution of diagnostic substitution to the growing administrative prevalence of autism in US special education. *Pediatrics*, 117, 1028–1037.
- Volkmar, F. R., Lord, C., Bailey, A., Schultz, R. T., & Klin, A. (2004). Autism and pervasive developmental disorders. *Journal of Child Psychology and Psychiatry*, 45, 135–170.
- Wiggins, L. D., Baio, J., & Rice, C. (2006). Examination of the time between first evaluation and first autism spectrum disorder diagnosis in a population-based sample. *Developmental and Behavioral Pediatrics*, 27, S79–S87.
- Wing, L., & Potter, D. (2002). The epidemiology of autistic spectrum disorders: Is the prevalence rising? *Mental Retardation and Developmental Disabilities Research Reviews*, 8, 151–161.

Copyright of *Journal of Autism & Developmental Disorders* is the property of Springer Science & Business Media B.V. and its content may not be copied or emailed to multiple sites or posted to a listserv without the copyright holder's express written permission. However, users may print, download, or email articles for individual use.