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A population-level study of the mental health of siblings of children who have a developmental disability

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ABSTRACT

This study used population level administrative data for health service utilization from the Ministry of Health, British Columbia, Canada to assess the mental health of siblings of children who have a developmental disability. At a population level, the study found strong evidence that siblings of children who have a developmental disability experience higher odds of a depression or other mental health diagnosis compared to siblings of children who do not have a developmental disability. In addition, there was evidence that in families with a child with a developmental disability, siblings who are diagnosed with depression or another mental health problem use physician and/or hospital services for these conditions to a greater extent than siblings who are diagnosed with depression or a mental health problem but do not have a family member with a developmental disability. Evidence of increased depression and mental health problems existed across all income levels, indicating that other stressors may have an impact. These findings suggest that siblings of children who have a developmental disability are a vulnerable group in need of programs and services that support their mental health.

1. Introduction

Within the literature there is evidence that having a sibling with a developmental disability (DD) may affect siblings who do not have a DD (Marquis, Hayes, & McGrail, 2019). Studies have examined effects upon sibling wellbeing (Emerson & Giallo, 2014), stress (Nixon & Cummings, 1999), adjustment (Giallo & Gavidia-Payne, 2006), anxiety (Pollard, Barry, Freedman, & Kotchick, 2013), physical health (Hogan, Park, & Goldscheider, 2003), behavior (Platt, Roper, Mandleco, & Freeborn, 2014) and mental health (Giallo, Gavidia-Payne, Minett, & Kapoor, 2012; Goudie, Havercamp, Jamieson, & Sahr, 2013; Lovell & Wetherell, 2016). However, findings have often been conflicting (Marquis et al., 2019), with some studies reporting no deleterious effect upon siblings (Cuskelly & Gunn, 2006; Dempsey, Llorens, Brewton, Mulchandani, & Goin-Kochel, 2012; Tomeny, Barry, & Bader, 2012), some reporting a positive effect upon siblings (Connors & Stalker, 2003) and some reporting negative effects (Constantino et al., 2006; Hogan et al., 2003).

Conflicting findings may be due to methodological differences among studies (Marquis et al., 2019; Stoneman, 2009). Studies often have differing theoretical underpinnings and/or interests of the researchers (Hodapp, Glidden, & Kaiser, 2005; Stoneman, 1993, 2005, 2009); differ in outcome measures (Constantino et al., 2006; Dempsey et al., 2012; Griffith, Hastings, & Petalas, 2014; Hogan et al., 2003;

Nixon & Cummings, 1999; Stoneman, 1993, 2009); differ in the type of disability examined and how DD is defined (Marquis et al., 2019); and use differing measurement tools (Marquis et al., 2019). In addition, studies of siblings of children with a DD have often been limited by methodological issues of small sample size (Burke & Fujiura, 2013; Hodapp et al., 2005), the practice of combining different types of DD to increase sample size (Marquis et al., 2019), convenience sampling (Burke & Fujiura, 2013), availability and use of an appropriate control group (Hodapp et al., 2005; Stoneman, 2009), lack of accurate data on the incidence and prevalence of DD (Fujiura, Rutkowski-Kmitta, & Owen, 2010; Lin et al., 2013, 2014), reliance upon self-reports (Marquis et al., 2019), and whether or not the study included variables other than having/not having a sibling with a DD (Marquis et al., 2019; Rossiter & Sharpe, 2001).

There is evidence of complex interactions among social determinants of health, the characteristics of the child with the DD, characteristics of the non-DD sibling, and family factors which can be related to the mental health of siblings of children who have a DD (Marquis et al., 2019). Studies have included different variables, among these are: sex of the non-DD sibling (Walton & Ingersoll, 2015); sex of the child with the DD (Begum & Blacher, 2011; Petalas, Hastings, Nash, Lloyd, & Dowey, 2009); type of DD (Constantino et al., 2006; Fisman, Wolf, Ellison, & Freeman, 2000; Hastings, 2007; Pilowsky, Yirmiya,

Gross-Tsur, & Shalev, 2007; Pollard et al., 2013); birth order (Dyke, Mulroy, & Leonard, 2009; Tomeny, Barry, & Bader, 2014); number of children in the family (Mulroy, Robertson, Aiberti, Leonard, & Bower, 2008); family income measures (Emerson & Giallo, 2014; Giallo & Gavidia-Payne, 2006; Mulroy et al., 2008; Neely-Barnes & Graff, 2011; Platt et al., 2014); and neighborhood characteristics (Emerson & Giallo, 2014). However, there is little agreement in the literature on the effects of these variables and there are few studies using population level data to study the range of variables (Marquis et al., 2019).

This study used population level administrative data from the Ministry of Health in British Columbia, Canada. The data are collected from physicians for billing purposes and contains information on the reason for each visit made by a patient to a physician or hospital expressed as a diagnostic code (ICD-9 or ICD-10). These data were used to examine the relationship between having a sibling with a DD and the mental health of siblings who do not have a DD. Administrative data have many benefits for studying disability related issues and address several of the weaknesses of previous studies. The most significant advantage of using administrative data is the size of the data set (Burke & Fujiura, 2013; Glasson & Hussain, 2008; Jutte, Roos, & Brownell, 2011). Large data set size can improve the generalizability of the findings, reduce problems with selection bias (Burke & Fujiura, 2013), enhance the capacity to study low prevalence associations and relatively small population (Andresen, 2011; Hogan, Msall, & Drew, 2006; Jutte et al., 2011), and provide the ability to select varying control groups (Glasson & Hussain, 2008). In addition, administrative data provide information on the prevalence of DD (Lin et al., 2014; Marquis, McGrail, Hayes, & Tasker, 2018), include data on demographics, reduce recruitment problems (Andresen, 2011), maintain privacy (Glasson & Hussain, 2008), and do not rely on self-reports. By linking administrative data with other data sources additional variables which may have an association with outcomes can be included in analyses. At the time this study was initiated no other literature examining the mental health of siblings was found that used large samples and population-based measures that are not based upon self-reports.

This study adds to the body of research by using population level data to ask the question:

In British Columbia (B.C.), Canada, is the mental health (as measured by use of health services) of siblings of children who have a developmental disability different from the mental health of siblings of children who do not have a developmental disability?

2. Materials and methods

2.1. Data source

Data for this study were obtained from Population Data B.C. (PopData B.C.), a service that provides access to and linkages among a number of different administrative databases in the province of B.C. Linkages are formed between multiple data sets and identifier data is separated from records of service use. Each individual is then given a unique identification number. PopData provides data for the entire population of B.C., with the exception of data from people who have never used provincial healthcare services; data from alternative funding arrangements for health professionals (e.g. salaried physicians and nurse practitioners); and data from on-reserve First Nations health services.

Three databases held by PopData B.C. were linked.

- A *central consolidation file* providing demographic information on all individuals in B.C. The file contains information on birth date and sex of each individual within the province as well as neighborhood income quintiles from census data. ("British Columbia Ministry of Health [creator] (1986–2014): Consolidation File (MSP Registration and Premium Billing). Population Data B.C. [publisher]. Data Extract. MOH.," 2016; "Statistics Canada [creator] (1994, 2002, 2006): Statistics Canada Income Band Data. Population Data B.C. [publisher]. Data Extract. Population Data B.C.," 2016).

- The *Medical Services Plan* (MSP) payment file that contains administrative information for all fee-for-service care provided by physicians in B.C. The file includes the date of each visit to a physician, the diagnostic code (ICD-9), the *Health Authority* and *Health Service Delivery Areas* where the visit occurred, the subsidy code indicating whether payments to the physician were subsidized through provincial programs and the amount of the subsidy ("Subsidy for MSP Premiums in B.C.," 2014) ("British Columbia Ministry of Health [creator] (1985–2014): Medical Services Plan (MSP) Payment Information File. Population Data B.C. [publisher]. Data Extract. MOH.," 2016).

- The *hospital separation file* containing information on all hospitalizations (but not including emergency room visits), the date of admission and discharge and related diagnostic codes (ICD-9 and ICD-10) ("Canadian Institute for Health Information [creator] (1985–2014): Discharge Abstract Database (Hospital Separations). Population Data B.C. [publisher]. Data Extract. MOH.," 2016). The file does not include emergency room visists.

For this study data were obtained for the years 1990–2014. The B.C. Ministry of Health approved use of the data. Ethics approval was granted by the University of Victoria Human Research Ethics Board (#15–043).

2.2. Study groups

Study groups consisted of children (aged 0–19) who had siblings with a DD. Four cohorts of siblings were used: siblings of children who have a DD and were born between 1990 and 1995 (siblings of children with a DD (SDD1990)); siblings of children who have a DD and who were born between 2000 and 2005 (siblings of children with a DD (SDD2000)); siblings of children born between 1990 and 1995 who did not have a DD (siblings of typical children (STC1990)); and siblings of children born between 2000 and 2005 who did not have a DD (siblings of typical children STC2000)). For each of the groups data were obtained up to the year 2014 (data were obtained from 1990 to 2014 for cohorts in which the reference children were born 1990–95, data were obtained from 2000 to 2014 for cohorts in which the reference children were born in 2000–05). Stratification of the data by time was done in order to reduce cohort effect and to counteract large sample size fallacy. The time frames chosen represent a large proportion of the population, but still provide numbers of observations that were manageable for data manipulation.

Identification of children with a DD was done using the algorithm developed by Lin, Balogh, et al. (2013). Using this algorithm, children aged 0–19 were identified by ICD-9 codes in MSP files and by ICD-9 and ICD-10 codes in hospital separations files (see Appendix A). Identification required at least two occurrences of the ICD-9 codes identifying DD in MSP data or at least one occurrence of DD identified by ICD-9 or ICD-10 codes in hospital separation data.

Linking siblings who do not have a DD and children with a DD was done using Medical Services Plan (MSP) contract numbers which are shared within families. Linkages can include non-biological children such as adopted children and step-siblings as well as biological siblings. Therefore children may be linked with more than one family if parents have divorced, remarried and become associated with a new MSP contract number. At the time this study was initiated there were no other studies which used MSP contract numbers to link siblings.

2.3. Outcome measures

Diagnoses of depression or another mental health problem were used as outcome measures. Diagnoses could occur for the sibling any time between the birth of the child with the DD and 2014. Diagnoses were identified using ICD-9 codes in MSP data and ICD-9 and ICD-10 codes in hospital separations data. Diagnoses used are listed in Appendix B. Diagnoses from MSP and hospital files were combined for each person. For older siblings, only diagnoses that occurred after the birth of the reference child were used, regardless of whether or not the sibling had a previous mental health problem.

2.4. Variables

The primary exposure variable for the study was having or not having a sibling who has a DD. For both the logistic and negative binomial regressions, other variables used in the model were selected based upon findings in the literature (Marquis et al., 2019), the variables available in administrative data and evidence of a significant association in the initial descriptive statistics. The administrative data does not include information on potentially important variables such as race/ethnicity, parent's marital status, child behavior problems, or severity of the DD. Variables that are available include: number of children in the family, birth order, age of the older sibling at the time of birth of the child who has the DD, nearness in age of younger siblings to the child who has the DD, sex of the siblings, and health authority where the child resides. Two measures of socioeconomic status were also used - neighborhood income quintile (a population level measure of socioeconomic status) and receipt of an MSP subsidy (an individual level indication of socioeconomic status). Eligibility for a MSP subsidy is based upon annual net income calculated using family size, type of disability, child care subsidies and related socioeconomic characteristics.

Neighborhood income quintile was determined initially at the time of the birth of the child with the DD. These figures were used for the initial descriptive statistics. Subsequently the last neighborhood income quintile value available for each individual in the data and corresponding MSP subsidy value were used for regression analyses. The last (most recent) values were used as this maximized completeness of the variable and increased the probability that the value reflected circumstances after the births of all children within the family.

Location (health authority) was used as a variable in this study as health services can vary across B.C., with fewer services located in northern areas. In the Class statements of the analyses the reference levels were formed as male for the variable of child sex, Vancouver Coastal for the variable of health authority, no subsidy for the variable of MSP subsidy, and highest income quintile for the variable of neighborhood income quintile.

Analyses were also stratified by sex of the child who did not have the DD and by birth order. There is evidence in the literature that both of these variables can be associated with the mental health of siblings of children who have a DD but again the findings are conflicting. In a study of 52 siblings of children who had a variety of different disabilities, Giallo et al. (2012) reported that the sex of the non-disabled sibling was not a significant predictor of self-reported mental health. In contrast, Walton and Ingersoll (2015) found that older male siblings (average age 11 years) of children with ASD were more likely to demonstrate difficulties with behavioural/emotional adjustment than female siblings. Mulroy et al. (2008) found that in families with a child with Down syndrome, later birth order of the child with Down syndrome was associated with less likelihood of parents reporting disadvantages to non-disabled siblings and more likelihood of parents

reporting advantages to non-disabled siblings. Tomeny et al. (2014) reported that siblings of children with ASD exhibited increased behavior problems only when the child with ASD was older than the sibling. However, Giallo et al. (2012) reported that birth order was not a significant predictor of sibling mental health.

2.5. Analyses

Siblings were first examined to obtain descriptive parameters. Data were then analyzed using *logistic regression* to determine whether or not there was a diagnosis of depression or a mental health problem. The data for those who were diagnosed (e.g. with depression) were analyzed using negative binomial regression to determine use of the health care system for that particular diagnosis.

SAS proc logistic ("SAS/STAT 14.1 User's Guide:: SAS/STAT(R) 14.1 User's Guide," n.d.) was used for all logistic regression analyses. Proc logistic fitted logistic regression models and estimated parameters by *maximum likelihood* using a *Fisher's Scoring* algorithm. Individuals with any missing observations were automatically deleted during model formation by proc logistic. Missing data were primarily for the variable neighborhood income quintile and secondly for health authority. Depending upon the time period and the age of the sibling, between 88% and 97% of the data were complete.

Initially variables were added manually to the model. This was tested against the three automatic selection methods offered by proc logistic. Very little difference was found between the outcomes of the manual selection and the three automatic methods. Also, there was no difference in the outcomes between the three methods (forward, backward or stepwise). Stepwise selection was ultimately used for all of the proc logistic procedures. The *probability level* (*alpha*) was set at 0.01 for entry of the variables into the model and as a criterion for the variable to stay in the model. *Odds ratios* were produced for each variable that had a significant relationship with the outcome of interest. SAS proc logistic automatically dropped any variables that did not improve the fit of the regression model; therefore these variables do not appear in the final results tables.

Negative binomial regression was conducted using data from siblings who had at least one diagnosis of depression or of a mental health problem other than depression. Visits to physician offices or to a hospital were combined. Therefore the outcome was the number of physician/hospital visits which involved a diagnosis of depression or a diagnosis of a mental health problem excluding depression.

Proc countreg in SAS ("SAS/STAT 14.1 User's Guide:: SAS/STAT(R) 14.1 User's Guide," n.d.) was used. Proc countreg uses *maximum likelihood estimation* and a *Newton Raphson optimization technique.* Missing observations were automatically deleted during model formation. Fit statistics include Akaike's Information Criteria (AIC), the Schwarz's Bayesian Information Criterion (SBC), and the *log likelihood* for the final model. AIC and SBC were used to compare different models.

3. Results

3.1. Descriptive results

Table 1 shows descriptive parameters of the four groups of siblings. The groups were similar in percentage of sisters and brothers and family size. There was a higher percentage of siblings of children with a DD in the lowest income quintile at birth of the reference child compared to the comparison groups. There was a slightly lower percentage of siblings of children with a DD in the highest income quintile compared to siblings of the comparison groups. Both sisters and brothers of children who have a DD had a higher percentage of diagnoses of a mental health

Table 1
Descriptive statistics.

Variable	Siblings of children who have a DD who were born 1990–95 (SDD1990)	Siblings of typical children who were born 1990–95 (STC1990)	Siblings of children who have a DD who were born 2000–05 (SDD2000)	Siblings of typical children who were born 2000–2005 (STC2000)
Total number of siblings	9,807	565,033	8,488	315,939
Number of sisters (percent of total siblings)	5,056 (51.56%)	277,184 (49.06%)	4,140 (48.77%)	156,901 (49.66%)
Number of brothers	4,751	287,849	4,348	159,038
Mean number of children in the family	1.06 (S.D. 0.24)	1.12 (S.D. 0.41)	1.32 (S.D. 0.47)	1.22 (S.D. 0.46)
	Range 1-3	Range 1-7	Range 1-3	Range 1-5
Income quintile of the reference child at birth				
Lowest	1,169 (27.10%)	57,527 (22.08%)	1,193 (25.16%)	49,542 (21.77%)
2nd	914 (21.19%)	55,449 (21.28%)	1,017 (21.47%)	48,413 (21.28%)
3rd	812 (18.82%)	53,388 (20.49%)	957 (20.21%)	46,452 (20.42%)
4th	759 (17.59%)	50,919 (19.54%)	849 (17.93%)	44,057 (19.36%)
Highest	660 (15.30%)	43,264 (16.61%)	720 (15.20%)	39,074 (17.17%)
Sisters with a diagnosis of depression (number and %)	2,363 (46.74%)	65,340 (23.57%)	882 (21.49%)	14,442 (9.20%)
Sisters with a diagnosis of a mental health problem (other than depression) (number and %)	2,443 (48.32%)	63,004 (22.73%)	1,110 (27.05%)	19,171 (12.22%)
Brothers with a diagnosis of depression (number and %)	1,620 (34.16%)	47,783 (16.60%)	658 (15.13%)	12,731 (8.01%)
Brothers with a diagnosis of a mental health problem (other than depression) (number and %)	2,338 (49.21%)	66,304 (23.03%)	1,462 (33.62%)	26,455 (16.64%)

problem and depression compared to siblings in the other two groups.

3.2. Logistic regression of odds of having a depression or another mental health diagnosis

Tables 2–5 show the results of the logistic regression analyses comparing younger sisters, older sisters, younger brothers and older brothers of siblings of children with a DD to siblings of children without a DD. Two time periods were compared: siblings of children who were born 1990–1995 and who had a DD to siblings of children born 1990–1995 who did not have a DD; and siblings of children born 2000–2005 who had a DD and siblings of children born 2000–2005 who did not have a DD. In all cases, siblings of children with a DD had greater odds of a diagnosis of depression or of another mental health problem excluding depression compared to siblings of children without

a DD. Odds were greatest for younger sisters and younger brothers of children with a DD.

Increasing age of older siblings (both sisters and brothers) at birth of the reference child was positively associated with odds of either a depression or mental health problem diagnosis. Although the findings were significant, age at birth had a relatively small association and may be due to the large size of the data set. The closer in age younger siblings (both sisters and brothers) were to the reference child, the higher their odds of either a depression or mental health diagnosis. These findings may primarily be due to time, as older siblings have more time in which to have a diagnosis. Sex of the reference child had no significant association with odds of a depression or mental health diagnosis. Increasing numbers of children in the family was associated with lower odds of a diagnosis of depression or a mental health problem in some cases.

Table 2
Predictors of a Depression Diagnosis for Siblings of DD Children Born 1990–1995, Odds Ratios (CI).

Variable	Younger Sisters	Older Sisters	Younger Brothers	Older Brothers
Cohort (siblings of children with a DD vs siblings of children without a DD)	1.96 (1.77–2.17)	1.56 (1.43–1.69)	1.96 (1.75–2.19)	1.48 (1.36–1.61)
Age at birth of or nearness of age to the child with the DD	1.18 (1.17–1.19)	1.11 (1.10–1.11)	1.14 (1.13–1.15)	1.06 (1.05–1.06)
Number of children in the family	N.S.	0.81 (0.78–0.84)	N.S.	0.84 (0.81–0.88)
Income quintile				
Lowest vs highest	N.S.	1.09 (1.04–1.14)	N.S.	N.S.
2nd vs highest	N.S.	1.07 (1.02–1.12)	N.S.	N.S.
3rd vs highest	N.S.	1.06 (1.01–1.11)	N.S.	N.S.
4th vs highest	N.S.	1.08 (1.03–1.13)	N.S.	N.S.
Receipt of an MSP subsidy	1.09 (1.04–1.13)	N.S.	1.15 (1.10–1.20)	1.17 (1.14–1.21)
Health authority				
Interior vs Vancouver Coastal	1.43 (1.34–1.52)	1.52 (1.45–1.59)	1.18 (1.10–1.26)	1.15 (1.10–1.21)
Fraser vs Vancouver Coastal	1.22 (1.15–1.28)	1.30 (1.25–1.35)	1.05 1.00–1.11)	1.13 (1.08–1.17)
Island vs Vancouver Coastal	1.56 (1.47–1.67)	1.66 (1.58–1.74)	1.15 (1.08–1.23)	1.21 (1.15–1.27)
Northern vs Vancouver Coastal	1.20 (1.12–1.30)	1.33 (1.26–1.40)	0.98 (0.90–1.06)	0.96 (0.90–1.01)

Table 3
Predictors of a Depression Diagnosis for Siblings of DD Children Born 2000–2005, Odds Ratios (CI).

Variable	Younger Sisters	Older Sisters	Younger Brothers	Older Brothers
Cohort (siblings of children with a DD vs siblings of children without a DD)	2.24 (1.64–3.06)	1.63 (1.41–1.88)	1.92 (1.75–2.19)	1.50 (1.28–1.76
Age at birth of or nearness of age to the child with the DD	1.11 (1.10–1.15)	1.10 (1.09–1.12)	1.11 (1.07–1.14)	1.09 (1.07–1.11)
Number of children in the family	N.S.	0.79 (0.74–0.86)	N.S.	0.79 (0.73–0.86)
Income quintile				
Lowest vs highest	N.S.	N.S.	N.S.	N.S.
2nd vs highest	N.S.	N.S.	N.S.	N.S.
3rd vs highest	N.S.	N.S.	N.S.	N.S.
4th vs highest	N.S.	N.S.	N.S.	N.S.
Receipt of an MSP subsidy	N.S.	N.S.	N.S.	N.S.
Health authority				
Interior vs Vancouver Coastal	0.65 (0.50–0.83)	1.44 (1.32–1.57)	N.S.	1.24 (1.13–1.37)
Fraser vs Vancouver Coastal	0.84 (0.69–1.00)	1.03 (0.96–1.11)	N.S.	1.04 (0.97–1.13)
Island vs Vancouver Coastal	0.93 (0.74–1.18)	1.55 (1.42–1.69)	N.S.	1.28 (1.16–1.41)
Northern vs Vancouver Coastal	0.70 (0.52–0.95)	1.26 (1.13–1.41)	N.S.	0.99 (0.88–1.12)

The findings for neighborhood income quintile varied depending upon the age and sex of the sibling and the time period when the reference child was born. Generally, income quintile had a small association with odds, with the lowest quintile compared to the highest quintile having the most effect. Receiving an MSP subsidy slightly increased odds of a depression or mental health diagnosis in most of the analyses conducted. Health authority had variable effects upon odds of a depression or mental health problem diagnosis depending upon the health authority, the diagnosis and the sex and age of the sibling.

3.3. Negative binomial analyses of number of physician/hospital visits

Very similar results were found in the negative binomial regression analyses conducted on the study groups (Tables 6–9). Siblings (both brothers and sisters) of children who had a DD and who had a diagnosis of either depression or a mental health problem had significantly greater numbers of visits to a physician/hospital for their diagnosis when compared to siblings of children who did not have a DD.

4. Discussion

There is evidence in the literature that having a sibling with a DD is associated with poor mental health of siblings without a DD. However, much of the evidence has been conflicting (Marquis et al., 2019). The purpose of this study was to add to the population health perspective of the relationships between the presence of a child with a DD and the mental health of siblings by using data available through administrative health data in B.C. This is the first time in Canada that administrative health data have been used to study the health of siblings of children who have a DD.

At the population level in B.C. there were significantly higher odds of a diagnosis of depression or another mental health problem in sisters and brothers of children who have a DD compared to the siblings of children who do not have a DD. In addition, siblings who have a diagnosis of depression or a mental health problem and who have a sibling with a DD, use the health care system more often for their diagnosed depression or mental health problem compared to siblings of children who do not have a DD and who have a diagnosis of depression or a mental health problem.

Table 4
Predictors of a Mental Health Problem Diagnosis for Siblings of DD Children Born 1990–1995, Odds Ratios (CI).

Variable	Younger Sisters	Older Sisters	Younger Brothers	Older Brothers
Cohort (siblings of children with a DD vs siblings of children without a DD)	2.20 (2.00–2.42)	1.79 (1.65–1.94)	2.35 (2.14–2.58)	1.77 (1.63–1.93)
Age at birth of or nearness of age to the child with the DD	1.11 (1.10–1.11)	1.08 (1.07–1.08)	1.07 (1.06–1.07)	1.03 (1.02–1.04)
Number of children in the family	0.90 (0.86–0.95)	0.86 (0.83–090)	0.89 (0.85–0.93)	0.87 (0.83–0.91)
Income quintile				
Lowest vs highest	1.11 (1.05–1.17)	1.08 (1.03–1.13)	1.11 (1.06–1.17)	N.S.
2nd vs highest	1.06 (0.99–1.12)	1.08 (1.03–1.13)	1.08 (1.02–1.13)	N.S.
3rd vs highest	1.11 (1.05–1.17)	1.02 (0.98–1.07)	1.03 (0.98–1.09)	N.S.
4th vs highest	1.07 (1.01–1.13)	1.04 (0.99–1.09)	1.04 (0.99–1.09)	N.S.
Receipt of an MSP subsidy	1.13 (1.09–1.17)	1.04 (1.01–1,07)	1.13 (1.09–1.17)	1.20 (1.16–1.23)
Health authority				
Interior vs Vancouver Coastal	1.17 (1.10–1.23)	1.24 (1.19–1.30)	1.18 (1.12–1.24)	1.20 (1.15–1.26)
Fraser vs Vancouver Coastal	1.17 (1.11–1.22)	1.30 (1.25–1.35)	1.10 (1.06–1.15)	1.28 (1.23–1.33)
Island vs Vancouver Coastal	1.36 (1.28–1.44)	1.50 (1.43–1.57)	1.22 (1.16–1.29)	1.28 (1.22–1.34)
Northern vs Vancouver Coastal	0.77 (0.71–0.83)	0.943 (0.89–0.99)	0.87 (0.82–0.93)	0.93 (0.89–0.99)

Table 5

Predictors of a Mental Health Problem Diagnosis for Siblings of DD Children Born 2000–2005, Odds Ratios (CI).

Variable	Younger Sisters	Older Sisters	Younger Brothers	Older Brothers
Cohort (siblings of children with a DD vs siblings of children without a DD)	2.26 (1.88–2.72)	1.80 (1.57–2.07)	2.43 (2.10–2.81)	1.84 (1.61–2.09)
Age at birth of or nearness of age to the child with the DD	1.08 (1.06–1.11)	1.05 (1.03–1.06)	1.11 (1.09–1.13)	N.S.
Number of children in the family	N.S.	0.80 (0.75–0.86)	N.S.	0.84 (0.79–0.90)
Income quintile				
Lowest vs highest	1.11 (1.05–1.17)	N.S.	1.24 (1.12–1.38)	N.S.
2nd vs highest	1.09 (0.95–1.26)	N.S.	1.16 (1.05–1.29)	N.S.
3rd vs highest	1.26 (1.10–1.44)	N.S.	1.11 (0.99–1.23)	N.S.
4th vs highest	1.11 (0.97–1.18)	N.S.	1.09 (0.99–1.21)	N.S.
Receipt of an MSP subsidy	1.14 (1.12–1.37)	N.S.	1.20 (1.11–1.30)	1.16 (1.09–1.22)
Health authority				
Interior vs Vancouver Coastal	1.01 (0.88–1.16)	1.25 (1.15–1.36)	1.01 (0.91–1.13)	1.22 (1.13–1.32)
Fraser vs Vancouver Coastal	0.97 (0.86–1.08)	1.07 (0.99–1.14)	1.01 (0.92–1.10)	1.06 (0.99–1.13)
Island vs Vancouver Coastal	0.99 (0.86–1.14)	1.39 (1.28–1.52)	1.04 (0.94–1.16)	1.25 (1.16–1.35)
Northern vs Vancouver Coastal	0.46 (0.37–0.58)	0.84 (0.75–0.95)	0.60 (0.51–0.70)	0.84 (0.75–0.93)

In addition, this study added to the research on associations between income, the sex of the sibling without the DD, birth order and mental health outcomes. These are important additions to the literature as previous studies have often had conflicting results.

Differing results between this study and previous studies may in part be due to differing methodology and sample size. Many previous studies used poorly defined populations of disabled children or poorly defined comparison groups, were done over short time periods, did not differentiate between sisters and brothers, and/or relied solely upon self-reported health data (Stoneman, 2009). This study expanded the research by using population level administrative data, a variety of defined variables, diagnoses provided by physicians, definitions of DD according to International Classification of Disease codes, and several large comparison populations.

Differing results may also be due to the complexity of studying families, many previous studies have not accounted for the complexity inherent in studies of families. By stratifying the analyses according to gender (sister/brother) and age (younger and older siblings) and by including variables such as socioeconomic measures, complex inter-relationships were found in this study that may not have been apparent in previous studies.

Similar to studies that found lower incomes among families with a child with a DD (Parish, Rose, Grinstein-Weiss, Richman, & Andrews, 2008; Parish, Seltzer, Greenberg, & Floyd, 2004), in this study, there was a greater proportion of siblings of a child with a DD in the lowest neighborhood income quintiles compared to siblings of children without a DD. This study found some evidence of increased odds of depression or a mental health diagnosis in lower income groups. However, this study found that the association between having a sibling with a DD and the odds of depression or another mental health problem remained when income measures were held constant, indicating that although income plays a role in the outcomes of depression and mental health problems, that other factors also play a role.

Limitations of this study include limitations specific to the algorithm

Table 6

Predictors of the Number of Hospital/Physician Visits for a Depression Diagnosis for Siblings of DD Children Born 1990–1995, (exp)β (probability).

Variable	Younger Sisters	Older Sisters	Younger Brothers	Older Brothers
Cohort (siblings of children with a DD vs siblings of children without a DD)	1.17 ($p = 0.0084$)	1.12 ($p = 0.0140$)	1.66 ($p < 0.0001$)	1.26 ($p < 0.0001$)
Age at birth of or nearness of age to the child with the DD	1.03 ($p < 0.0001$)	1.03 ($p < 0.0001$)	1.04 ($p < 0.0001$)	1.02 ($p < 0.0001$)
Sex of the child with the DD (female vs male)	N.S.	N.S.	1.120 ($p < 0.0001$)	N.S.
Number of children in the family	N.S.	0.88 ($p < 0.0001$)	N.S.	0.93 ($p = 0.0131$)
Income quintile				
Lowest vs highest	0.89 ($p = 0.0009$)	N.S.	0.86 ($p = 0.0002$)	0.88 ($p < 0.0001$)
2nd vs highest	0.90 ($p = 0.0026$)	N.S.	0.76 ($p < 0.0001$)	0.88 ($p < 0.0001$)
3rd vs highest	0.89 ($p = 0.0009$)	N.S.	0.92 ($p = 0.0264$)	0.90 ($p = 0.0005$)
4th vs highest	1.03 ($p = 0.3868$)	N.S.	0.89 ($p = 0.0027$)	0.93 ($p = 0.0189$)
Receipt of an MSP subsidy	N.S.	N.S.	1.08 ($p = 0.0082$)	1.14 ($p < 0.0001$)
Health authority				
Interior vs Vancouver Coastal	0.75 ($p < 0.0001$)	0.99 ($p = 0.7715$)	0.77 ($p < 0.0001$)	0.83 ($p < 0.0001$)
Fraser vs Vancouver Coastal	0.72 ($p < 0.0001$)	0.94 ($p = 0.0056$)	0.81 ($p < 0.0001$)	0.84 ($p < 0.0001$)
Island vs Vancouver Coastal	0.86 ($p < 0.0001$)	0.99 ($p = 0.9320$)	0.78 ($p < 0.0001$)	0.82 ($p < 0.0001$)
Northern vs Vancouver Coastal	0.64 ($p < 0.0001$)	1.10 ($p = 0.0023$)	0.67 ($p < 0.0001$)	0.68 ($p < 0.0001$)

Table 7

Predictors of the Number of Hospital/Physician Visits for a Depression.

Diagnosis for Siblings of DD Children Born 2000–2005, (exp)β (probability).

[Note: there were insufficient numbers of younger sisters and younger brothers who had multiple diagnoses of depression in the DD groups to conduct analyses for those groups].

Variable	Older Sisters	Older Brothers
Cohort (siblings of children with a DD vs siblings of children without a DD)	1.29 ($p = 0.0002$)	1.32 ($p = 0.0004$)
Age at birth of or nearness of age to the child with the DD	1.06 ($p < 0.0001$)	1.04 ($p < 0.0001$)
Sex of the child with the DD (female vs male)	N.S.	N.S.
Number of children in the family	N.S.	N.S.
Income quintile		
Lowest vs highest	N.S.	0.77 ($p < 0.0001$)
2nd vs highest	N.S.	0.83 ($p < 0.0001$)
3rd vs highest	N.S.	0.79 ($p < 0.0001$)
4th vs highest	N.S.	0.84 ($p = 0.0002$)
Receipt of an MSP subsidy	N.S.	N.S.
Health authority		
Interior vs Vancouver Coastal	0.75 ($p < 0.0001$)	0.67 ($p < 0.0001$)
Fraser vs Vancouver Coastal	0.81 ($p < 0.0001$)	0.74 ($p < 0.0001$)
Island vs Vancouver Coastal	0.88 ($p = 0.0025$)	0.81 ($p < 0.0001$)
Northern vs Vancouver Coastal	0.68 ($p < 0.0001$)	0.72 ($p < 0.0001$)

and codes used to identify children with a DD. As Lin, Balogh, et al. (2013) found, the number of people identified with a DD in administrative data is affected by the algorithm used. In this study children with a DD were identified as those with at least two occurrences of the ICD-9 codes identifying DD in MSP data or at least one occurrence of DD identified by ICD-9 or ICD-10 codes in hospital separation data. There is also evidence that the quality and completeness of physician coding is greater for three digit ICD-9 codes than for four and five digit

ICD-9 codes (Hu, 1996). This study used some four and five digit ICD-9 codes to identify particular disabilities. Therefore, there may have been under-representation of some disabilities including FAS and some of the rarer disabilities such as Prader-Willi syndrome.

However, the estimate of prevalence of children who have a DD in B.C. found in this study and reported elsewhere (Marquis et al., 2018) was equal to or higher than prevalence reported in the literature. The prevalence in 1990 was 0.68% and the prevalence in 2000 was 1.45% (Marquis et al., 2018), compared to prevalence estimates of 0.72–1.10% reported for children in the province of Manitoba (Ouellette-Kuntz et al., 2009). The prevalence findings in this study therefore appear to be in line with rates previously reported in the literature.

This study used two measures of income, neighborhood income quintiles provided by Statistics Canada, and MSP subsidy levels. The neighborhood divisions used in the neighborhood income quintiles include the variety of incomes which may occur within a geographic area. This obscuring of the heterogeneity of individual incomes may be particularly problematic in rural areas where the "neighborhood" may be a very large geographic area. In addition, because the last available neighborhood income quintile and MSP subsidy values were used results regarding the relationship with socioeconomic status must be interpreted with caution.

This study was unable to account for all variables which may affect sibling mental health; particularly it could not account for the effects of potentially important individual variables such as ethnicity, marital status of parents, severity of the disability, child behavior, stigma experienced, and parenting styles. However, this study did incorporate a range of both individual level and population level variables.

This is the first study to link family members in administrative data for the purpose of studying the effects of developmental disability. This is both a strength and a limitation of the research. The unique character of the research adds significantly to the literature, however, there has been no independent validation of the methodological approach.

Lastly, large data sets can have problems associated with large sample size fallacy (Lantz, 2013; Lin, Lucas, & Shmueli, 2013; Veldhuizen, Pasker-De Jong, & Atsma, 2012). As recommended in the literature (Lin, Lucas, et al., 2013; Veldhuizen et al., 2012), this issue was addressed in this study by stratifying the samples, reporting odds

Table 8

Predictors of the Number of Hospital/Physician Visits for a Mental Health Problem Diagnosis for Siblings of DD Children Born 1990–1995, (exp)β (probability).

Variable	Younger Sisters	Older Sisters	Younger Brothers	Older Brothers
Cohort (siblings of children with a DD vs siblings of children without a DD)	1.32 ($p < 0.0001$)	1.25 ($p < 0.0001$)	1.44 ($p < 0.0001$)	1.36 ($p < 0.0001$)
Age at birth of or nearness of age to the child with the DD	1.03 ($p < 0.0001$)	1.05 ($p < 0.0001$)	N.S.	1.02 ($p < 0.0001$)
Sex of the child with the DD (female vs male)	1.07 ($p = 0.0019$)	N.S.	N.S.	N.S.
Number of children in the family	N.S.	0.86 ($p < 0.0001$)	0.92 ($p = 0.0120$)	N.S.
Income quintile				
Lowest vs highest	0.94 ($p = 0.1118$)	1.09 ($p = 0.0054$)	1.18 ($p < 0.0001$)	0.99 ($p = 0.6795$)
2nd vs highest	0.92 ($p = 0.0197$)	1.10 ($p = 0.0053$)	1.08 ($p = 0.0269$)	0.92 ($p = 0.0108$)
3rd vs highest	0.89 ($p = 0.0013$)	0.97 ($p = 0.2950$)	1.04 ($p = 0.3085$)	0.91 ($p = 0.0027$)
4th vs highest	0.96 ($p = 0.2428$)	1.02 ($p = 0.5150$)	0.98 ($p = 0.5137$)	0.94 ($p = 0.0555$)
Receipt of an MSP subsidy	1.08 ($p = 0.0049$)	0.88 ($p < 0.0001$)	1.20 ($p < 0.0001$)	1.15 ($p < 0.0001$)
Health authority				
Interior vs Vancouver Coastal	0.98 ($p = 0.5452$)	0.91 ($p = 0.0066$)	0.98 ($p = 0.4683$)	0.84 ($p < 0.0001$)
Fraser vs Vancouver Coastal	0.91 ($p = 0.0026$)	0.92 ($p = 0.0035$)	0.93 ($p = 0.0086$)	0.91 ($p = 0.0008$)
Island vs Vancouver Coastal	1.01 ($p = 0.8996$)	0.94 ($p = 0.0677$)	1.11 ($p = 0.0044$)	0.94 ($p = 0.0492$)
Northern vs Vancouver Coastal	0.79 ($p < 0.0001$)	0.65 ($p < 0.0001$)	0.78 ($p < 0.0001$)	0.71 ($p < 0.0001$)

Table 9

Predictors of the Number of Hospital/Physician Visits for a Mental Health Problem Diagnosis for Siblings of DD Children Born 2000–2005, (exp)β (probability).

Variable	Younger Sisters	Older Sisters	Younger Brothers	Older Brothers
Cohort (siblings of children with a DD vs siblings of children without a DD)	1.41 ($p = 0.0002$)	1.33 ($p < 0.0001$)	N.S.	1.40 ($p < 0.0001$)
Age at birth of or nearness of age to the child with the DD	1.04 ($p = 0.0005$)	1.04 ($p < 0.0001$)	1.06 ($p < 0.0001$)	1.03 ($p = 0.0001$)
Sex of the child with the DD (female vs male)	N.S.	N.S.	0.92 ($p = 0.0150$)	N.S.
Number of children in the family	N.S.	N.S.	N.S.	N.S.
Income quintile				
Lowest vs highest	N.S.	0.96 ($p = 0.3501$)	N.S.	N.S.
2nd vs highest	N.S.	0.87 ($p = 0.0014$)	N.S.	N.S.
3rd vs highest	N.S.	0.94 ($p = 0.1931$)	N.S.	N.S.
4th vs highest	N.S.	0.89 ($p = 0.0104$)	N.S.	N.S.
Receipt of an MSP subsidy	N.S.	1.10 ($p = 0.0082$)	1.21 ($p < 0.0001$)	1.11 ($p = 0.0004$)
Health authority				
Interior vs Vancouver Coastal	0.75 ($p < 0.0001$)	0.88 ($p = 0.0057$)	N.S.	N.S.
Fraser vs Vancouver Coastal	0.93 ($p = 0.2067$)	0.84 ($p = 0.9347$)	N.S.	N.S.
Island vs Vancouver Coastal	0.86 ($p = 0.0328$)	1.00 ($p = 0.9347$)	N.S.	N.S.
Northern vs Vancouver Coastal	0.78 ($p = 0.0499$)	0.69 ($p < 0.0001$)	N.S.	N.S.

ratios and confidence intervals and by assessing the results in light of previous literature.

5. Conclusions

This study found evidence of increased odds of depression and other mental health problems at a population level in children who have a sibling with a DD. This study can be used as a foundation for further studies. It can also inform program planning as it offers evidence that siblings are an important group for consideration.

Declaration of conflict of interest

None.

Appendix A

ICD-9 and ICD-10 codes used for identification of developmental disability (disabilities include Autism Spectrum Disorder, Down syndrome, Fetal Alcohol Syndrome and Other developmental disabilities).

Disclaimer

All inferences, opinions and conclusions drawn in this paper are those of the authors, and do not reflect the opinions or policies of the Data Steward.

Ethical statement

Ethics approval was granted by the University of Victoria Human Research Ethics Board (#15-043).

Table A1

ICD-9 Codes for Developmental Disabilities.

ICD-9 Number	Disorder
299	Pervasive development disorders (e.g. autism)
317	Mild mental retardation
318	Moderate severe and profound mental retardation
319	Unspecified mental retardation
7580–7583	Chromosomal anomalies for which a developmental disability is typically present (e.g. Down syndrome, cri-du-chat syndrome)
7585	Other conditions due to autosomal anomalies
7588	Other conditions due to chromosome anomalies
7589	Conditions due to anomaly of unspecified chromosome
7595	Tuberous sclerosis
75981	Other and unspecified congenital anomalies: Prader–Willi
75983	Other and unspecified congenital anomalies: Fragile X
75989	Other and unspecified congenital anomalies: other (e.g. Menkes disease, Laurence–Moon–Biedl, Rubinstein–Taybi syndrome, etc.)
76071	Foetal alcohol syndrome
76077	Foetal hydantoin syndrome

Table A2
ICD-10 Codes for Developmental Disabilities.

ICD-10 Number	Disorder
F700	Mild mental retardation with the statement of no, or minimal, impairment of behavior
F701	Mild mental retardation, significant impairment of behavior requiring attention or treatment
F708	Mild mental retardation, other impairments of behavior
F709	Mild mental retardation without mention of impairment of behavior
F710	Moderate mental retardation with the statement of no, or minimal, impairment of behavior
F711	Moderate mental retardation, significant impairment of behavior requiring attention or treatment
F718	Moderate mental retardation, other impairments of behavior
F719	Moderate mental retardation without mention of impairment of behavior
F720	Severe mental retardation with the statement of no, or minimal, impairment of behavior
F721	Severe mental retardation, significant impairment of behavior requiring attention or treatment
F728	Severe mental retardation, other impairments of behavior
F729	Severe mental retardation without mention of impairment of behavior
F730	Profound mental retardation with the statement of no, or minimal, impairment of behavior
F731	Profound mental retardation, significant impairment of behavior requiring attention or treatment
F738	Profound mental retardation, other impairments of behavior
F739	Profound mental retardation without mention of impairment of behavior
F780	Other mental retardation with the statement of no, or minimal, impairment of behavior
F781	Other mental retardation, significant impairment of behavior requiring attention or treatment
F788	Other mental retardation, other impairments of behavior
F789	Other mental retardation without mention of impairment of behavior
F790	Unspecified mental retardation with the statement of no, or minimal, impairment of behavior
F791	Unspecified mental retardation, significant impairment of behavior requiring attention or treatment
F798	Unspecified mental retardation, other impairments of behavior
F799	Unspecified mental retardation without mention of impairment of behavior
F840	Childhood autism
F841	Atypical autism
F843	Other childhood disintegrative disorder
F844	Overactive disorder associated with mental retardation and stereotyped movements
F845	Asperger's syndrome
F848	Other pervasive developmental disorders
F849	Pervasive developmental disorder, unspecified
Q851	Tuberous sclerosis
Q860	Foetal alcohol syndrome
Q861	Foetal hydantoin syndrome
Q871	Aarskog, Prader–Willi, de Lange, Seckel, etc.
Q8723	Rubinstein–Taybi
Q8731	Sotos
Q878	Other
Q90	Down syndrome
Q91 – Q939	Chromosomal abnormalities not elsewhere classified
Q971	Female with more than three chromosomes
Q992	Fragile X Syndrome
Q998	Other specified chromosomal abnormalities

Appendix B

Table B1
ICD Numbers for Depression.

ICD Numbers	Disorder
ICD-9	
309	Adjustment reaction
311	Depressive disorder, not elsewhere classified
50B	Anxiety/depression
ICD-10	
F32	Depressive episode
F33	Recurrent depressive episode
F34	Persistent mood disorder
F38	Other mood disorders

Table B2
ICD Numbers for Mental Health Problems.

ICD Numbers	Disorder
ICD-9	
291–293	Psychotic conditions due to use of psychoactive substances
295–298	Other psychoses
300–308	Neurotic disorders, personality disorders and other nonpsychotic mental disorders
312	Disturbance of conduct not elsewhere classified
313	Disturbance of emotions specific to childhood and adolescence
314	Hyperkinetic syndrome of childhood
ICD-10	
F10 – F19	Mental and behavioral disorders due to use of psychoactive substances
F20 – F25, F28, F29	Schizophrenia, schizotypal and delusional disorders
F30	Manic episode
F31	Bipolar affective disorder
F40 – F45, F48	Neurotic, stress-related and somatoform disorders
F50 – F55, F59	Behavioral syndromes associated with physiological disturbances and physical factors
F90 – F95, F98	Behavioral and emotional disorders with onset usually occurring in childhood and adolescence

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