

## Cost of speech-language interventions for children and youth with foetal alcohol spectrum disorder in Canada

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### Abstract

This study, which is part of a large economic project on the overall burden and cost associated with Foetal Alcohol Spectrum Disorder (FASD) in Canada, estimated the cost of 1:1 speech-language interventions among children and youth with FASD for Canada in 2011. The number of children and youth with FASD and speech-language disorder(s) (SLD), the distribution of the level of severity, and the number of hours needed to treat were estimated using data from the available literature. 1:1 speech-language interventions were computed using the average cost per hour for speech-language pathologists. It was estimated that ~ 37,928 children and youth with FASD had SLD in Canada in 2011. Using the most conservative approach, the annual cost of 1:1 speech-language interventions among children and youth with FASD is substantial, ranging from \$72.5 million to \$144.1 million Canadian dollars. Speech-language pathologists should be aware of the disproportionate number of children and youth with FASD who have SLD and the need for early identification to improve access to early intervention. Early identification and access to high quality services may have a role in decreasing the risk of developing the secondary disabilities and in reducing the economic burden of FASD on society.

**Keywords:** Foetal alcohol spectrum disorder, foetal alcohol syndrome, speech disorders, language disorders, intervention, economic cost.

### Introduction

According to the Participation and Activity Limitation Surveys, 2001 and 2006, speech disability was the third most prevalent disability (after learning and chronic disabilities) among the 5–14 years age group in the general population of Canada (Statistics Canada, 2007). In 2006, ~ 45% of children aged 5–14 with one or more disabilities reported a speech disability, meaning that ~ 78,240 Canadian school-age children experienced difficulty speaking and/or being understood (Statistics Canada, 2007).

Among these children (the broad term “children” will be used to refer to children and youth) with disabilities there is a special population with a very high proportion of speech-language disorders (SLD)—children who were pre-natally exposed to alcohol (CDC, 1995; Church, Eldis, Blakley, & Bawle, 1997; Egeland, Perhain-Hestet, Gessnet, Ingle, Berne, & Micldalgh, 1998; Elliott, Payne, Morris, Haan, &

Bower, 2008; Kvigne, Leonardson, Neff-Smith, Brock, Borzelleca, & Welty, 2004; Spohr, Willms, & Steinhausen, 1994; Steinhausen, Nestler, & Spohr 1982). Alcohol is a teratogen that causes a spectrum of neurological, cognitive, and behavioural deficits in infants exposed to alcohol *in utero* (Stratton, Howe, & Battaglia, 1996). Foetal Alcohol Spectrum Disorder (FASD) is an umbrella term used to characterize the full range of damage caused by pre-natal alcohol exposure, varying from mild-to-severe and encompassing a broad array of physical defects as well as cognitive, behavioural, emotional, and adaptive functioning deficits (Chudley, Conry, Cook, Looock, Rosales, & LeBlanc, 2005). FASD includes the following four diagnoses: Foetal Alcohol Syndrome (FAS), Partial FAS (pFAS), Alcohol-related Neurodevelopmental Disorder (ARND), and Alcohol-related Birth Defects (ARBD; Chudley et al., 2005)—all of which may include congenital

anomalies, such as malformations and dysplasia of the cardiac, skeletal, renal, ocular, auditory, and other systems. Individuals with FASD are at an increased risk for neglect, abuse, foster care, and disrupted family life (Kvigne et al., 2004), which further increase their risk of developmental adversity.

People with FASD often suffer from SLD due to brain damage caused by the teratogenic effects of pre-natal alcohol exposure (Church et al., 1997). Pre-natal alcohol exposure can disrupt both an individual's development and their use of language (Mattson & Riley, 1998; Streissguth, Barr, Kogan, & Bookstein, 1996). Individuals with FASD may also be vulnerable to developing SLD as a result of their greater propensity for atypical or adverse social interactions (Coggins, Timler, & Olswang, 2007). Among such SLD are poor receptive and expressive language skills, phonological deficits, fluency and articulation difficulties, and associated neurocognitive abnormalities that can adversely impact speech-language development (Church et al., 1997). In addition, individuals with FASD often have other mental disorders, a well-established dimension of FASD that is also likely to affect language acquisition that may subsequently lead to difficulties with speech and articulation (Church et al., 1997).

Several studies have reported a high occurrence of SLD among individuals with FASD (Church et al., 1997; Egeland et al., 1998; Elliott et al., 2008; Kvigne et al., 2004; Pensiero, Manna, Michieletto, & Perissutti, 2007). Specifically, unspecified SLD have been reported to occur in 40% (CDC, 1995) to 90% (Church et al., 1997) of individuals with FAS, and 37% (Spohr et al., 1994) to 60% (Elliott et al., 2008) of individuals with FASD. Furthermore, Mills, McLennan, and Caza (2006) found, based on self-reports obtained from caregivers, that ~ 79% of children with FASD under the age of 7 years have visited a speech-language pathologist, and 57% of them received speech and language therapy in Alberta, Canada.

SLD in children with FASD have been linked to learning problems and social difficulties. Due to hearing, speech, and language acquisition problems, individuals with FASD may struggle with social and interpersonal communication, behavioural problems, following social norms, social reasoning, and information processing (Carmichael Olson, 1994; Church et al., 1997; Coggins, Olswang, Carmichael Olsen, & Timler, 2003). For individuals with FASD, these impairments may persist into adolescence and adulthood. Without the crucial support required, people affected by FASD are at a high risk for developing secondary disabilities, such as school failure and drop-out, addictions, mental health problems, sexually deviant behaviour, dependent living, unemployment, homelessness, involvement with the law, and incarceration (Streissguth, Bookstein, Barr, Sampson, O'Mally, & Young, 2004). When combined with the child's primary deficits, these secondary

disabilities increase the complexity of care and result in significant social and economic costs to society (Abel and Sokol, 1987; Harwood 2000; Legge, Roberts, & Butler, 2001; Lupton, Burd, & Harwood, 2004; PHAC 2003, 2005; Popova, Stade, Bekmuradov, Lange, & Rehm, 2011; Popova, Stade, Bekmuradov, Lange, & Rehm, 2012; Stade, Ali, Bennett, Campbell, Johnston, Lens, et al., 2009).

The purpose of this study was 2-fold: first, to estimate the number of children with FASD and SLD by age group (2–4, 5–9, 10–14, 15–19 years), sex, and level of severity (mildly and moderately-to-severely impaired); and, second, to estimate the cost of 1:1 speech-language interventions associated with these disorders in Canada in 2011.

SLD fall under the communication disorders umbrella. A speech disorder refers to an impairment of the respiratory system, phonation system, and/or articulation system of speech production (Justice, 2010). This group of disorders includes articulation and phonological disorders, fluency disorders, voice disorders, and motor speech disorders. A language disorder refers to deficit(s) in the linguistic system that ultimately impacts semantics, syntax, morphology phonology, and/or pragmatics (Justice, 2010).

In the current paper, the term SLD will be used throughout to describe all of the above disorders, and it is assumed that the rates of each are comparable in regard to their treatment and the frequency at which they occur in individuals with FASD.

The current study is part of a large economic project, with multiple components, that is working towards estimating the overall burden and cost associated with FASD in Canada, including the cost of healthcare, law enforcement, children in care, productivity losses of individuals with FASD and their caregivers, along with many others (Popova, Lange, Burd, & Rehm, 2012; Popova, Lange, Mihic, Bekmuradov, & Rehm, 2011; Popova, Stade, Lange, & Rehm, 2012; Popova, Stade, Lange, Mihic, & Rehm, 2012; Popova, Lange, Burd, Chudley, Clarren, & Rehm, 2013). Until now, an estimation of the number of children with FASD and SLD and the associated cost of 1:1 speech-language interventions, at the national level, has not been undertaken in Canada, or in any other country.

## Method

### *Estimation of the number of children with FASD by age group and sex in Canada in 2011*

In order to estimate the number of children, aged 2–19, with FASD in Canada in 2011, the only available and most commonly cited prevalence of FASD (nine per 1000; Roberts & Nanson, 2000) was applied to the general population of Canada by age group and sex in 2011 (Statistics Canada, 2012). Please note that children below 2 years of age were excluded due to their natural level of speech-language

development during these years. Further, this study did not estimate the cost of 1:1 speech-language interventions among individuals over the age of 19, as most treatments for children with SLD in Canada are linked to the educational system.

*Estimation of the number of children with FASD and SLD by age group and sex in Canada 2011*

There were three steps involved in estimating the number of children with FASD and SLD by age group and sex in Canada in 2011.

(1) *Systematic literature search.* A systematic literature search of available epidemiological and medical literature was performed in order to identify studies that have reported the prevalence of SLD among individuals with FASD. The search was conducted in multiple electronic bibliographic databases, including: Ovid MEDLINE, PubMed, EMBASE, Web of Science (including Science Citation Index, Social Sciences Citation Index, Arts and Humanities Citation Index), PsycINFO, ERIC, Scopus, and Social Work Abstracts. The search was conducted using multiple combinations of the following key words: foetal alcohol spectrum disorder, foetal alcohol syndrome, partial foetal alcohol syndrome, foetal alcohol effects, alcohol-related neurodevelopmental disorder, alcohol-related birth defects, pre-natal alcohol exposure, speech, language, disorder\*, disabilit\*, impairments\*, delay\*, deficit\*, prevalence, frequenc\*, and occurrence.

(2) *Meta-analysis.* The available data on the prevalence of SLD among individuals with FASD was analysed by performing a meta-analysis in order to estimate the pooled prevalence of SLD among individuals with FASD. A double arcsine transformation was applied to the prevalence estimates obtained from the epidemiological studies concerning SLD among those with FASD so that the data followed a normal distribution (Freeman & Tukey, 1950). The double arcsine-transformed prevalence estimates were weighted by the inverse variance of the double arcsine-transformed prevalence. The pooled prevalence of SLD among individuals with FASD was calculated using the Mantel-Haenszel method, assuming a random-effects model (Mantel & Haenszel, 1959). Heterogeneity between studies was assessed using the  $I^2$  statistic (Higgins & Thompson, 2002). Results of the meta-analysis were displayed using a Forest plot.

Publication bias was tested by visually inspecting a funnel plot for skewed distribution, and was adjusted for, if present, using the trim and fill method (Duval & Tweedie, 2000).

(3) *Estimation.* The pooled prevalence of SLD among individuals with FASD was applied to the number of children with FASD by age group and sex in

Canada, in order to estimate the number of children with FASD and SLD by age group and sex in Canada in 2011.

*Estimation of the number of children with FASD and SLD by level of severity in Canada 2011*

The number of children with FASD and SLD by level of severity was estimated based on data from the only available study (Coggins et al., 2007). The authors of this study examined communication deficits among 393 school-aged children with FASD. These children completed numerous standardized tests of language performance, which assessed their fundamental language skills, language comprehension, language development, overall language competence, and word knowledge. Further, the authors indexed the data on a language severity scale containing three levels—normal, mildly impaired, and moderately-to-severely impaired. The data were gathered from each participant using norm-referenced, standardized language tests. The mildly impaired severity level applied to children who obtained scores that ranged between  $-1.25$  SD and  $-2.00$  SD from test means, while the moderately-to-severely impaired level applied to children with test scores that fell more than  $-2.00$  SD below test means. Sixty-nine per cent of the children with FASD displayed significant language deficits, with 31% scoring in the mildly impaired range and 38% classified as moderately-to-severely impaired. This distribution of severity was applied to the total number of children with FASD and SLD in order to estimate the number of children with FASD and SLD by level of severity in Canada in 2011.

*Estimation of the cost associated with 1:1 speech-language interventions among children with FASD and SLD by age group, sex, and level of severity in Canada 2011*

There were two steps involved in estimating the cost associated with 1:1 speech-language interventions among children with FASD and SLD by age group, sex, and level of severity in Canada in 2011:

(1) *Estimation of the average number of hours required for 1:1 speech-language interventions.* In order to calculate the average number of hours required for 1:1 speech-language interventions, a systematic review and meta-analysis of randomized control trials on speech-language therapy was used (Law, Garrett, & Nye, 2003). This study revealed an overall positive effect of speech-language interventions for children with expressive phonological and expressive vocabulary difficulties. Based on the 27 studies included in the review by Law et al. (2003), the average number of hours for interventions ranged from 20–30 hours (Table I). Therefore, 20

hours was used as the number of hours needed to treat those who were mildly impaired, and 30 hours was used as the number of hours needed to treat those who were moderately-to-severely impaired.

(2) *Calculation of the cost associated with 1:1 speech-language interventions.* The cost per hour for a speech-language pathologist was obtained from the Canadian Association of Speech-Language Pathologists and Audiologists (CASLPA; personal communication with Joanne Charlebois, Executive Director), which was estimated to range from \$75 (lower estimate) to \$149 (upper estimate) in 2011. These figures were obtained from a fee survey conducted by the CASLPA of their members, and represent the average provincial responses to the survey (this survey is available to members of the association only).

In order to estimate the cost associated with 1:1 speech-language interventions, the lower (\$75) and upper (\$149) estimates of the cost per hour were applied to the number of hours needed to treat (20 hours for mildly impaired and 30 hours for moderately-to-severely impaired), and multiplied by the

respective number of children with FASD and SLD estimated for each province/territory in Canada, by age group and sex.

To estimate the costs for Canada in 2011 associated with 1:1 speech-language interventions among children with FASD and SLD, the following assumptions were made. First, it was assumed that the incidences of FASD and SLD and of the mortality rate (especially in people of younger ages with FASD and SLD) remain stable over time. It was also assumed that the population and the percentage of the population that is screened for FASD (based on the existing clinical capacity of all FASD multidisciplinary clinics in Canada; Clarren, Lutke, & Sherbuck, 2011) and SLD are also stable over time. Furthermore, it was assumed that the screening techniques used to diagnose FASD and SLD have not changed over time in terms of their sensitivity and specificity. However, since there have been increases in the population (Statistics Canada, 2013), improvements in the use of screening techniques for FASD (Chudley et al., 2005), and stable rates of consuming alcohol while pregnant (Statistics Canada, 2011), the use

Table I. Length of therapy provided to participants in 27 randomized control trials for 1:1 speech-language interventions.

Reference	Length of intervention (as reported in the original article)	Length of intervention (hours)
Almost & Rosenbaum (1998)	80 minutes a week over 4 months	21.3
Barratt, Littlejohns, & Thompson (1992)	40 minutes weekly over 6 months	16
Cole & Dale (1986)	600 minutes a week given for 8 months	320
Dixon, Joffe, & Bench (2001)	30 minutes a week for 10 weeks	5
Fey, Cleave, Long, & Hughes (1993)	180 minutes a week for 4.5 months	54
Fudala, England, & Ganoung (1972)	25 minutes a week for 4.5 months	7.5
Gibbard (1994) (study 1)	40 minutes a week over 6 months	16
Gibbard (1994) (study 2)	30 minutes a week for 6 months	12
Girolametto, Pearce, & Weitzman (1996a)	150 minutes a week for 10 weeks	25
Girolametto, Pearce, & Weitzman (1996b)	150 minutes a week for 11 weeks	27.5
Glogowska, Roulstone, Enderby, & Peters (2000)	10 minutes a week for 8.4 months	5.6
Lancaster (1991)	17 minutes of therapy a week over 6 months	6.8
Law, Kot, & Barnett (1999)	450 minutes a week for 6 weeks	45
Mulac & Tomlinson (1977)	67 minutes of therapy a week for 4 weeks	4.47
Munro (1999)	60 minutes a week for 6 weeks	6
Reid, Donaldson, Howell, Dean, & Greive (1996)	30 minutes given a week lasting up to 10 weeks	5
Robertson (1997)	20 minutes of therapy a week provided over 3 weeks	1
Robertson & Weismer (1999)	150 minutes a week for 12 weeks	30
Ruscello, Cartwright, Haines, & Shuster (1993)	120 minutes a week for 8 weeks	16
Rvachew (1994)	45 minutes a week over 6 weeks	4.5
Rvachew & Nowak (2001)	30 minutes a week for 12 weeks	6
Shelton, Johnson, Ruscella, & Arndt (1978)	57 days (listening for 5 minutes a day and reading and talking for 15 minutes a day)	19
Sommers (1962)	200 minutes a week for 4 weeks	13.3
Sommers, Furlong, Rhodes, Fichter, Bowser, Copetas, et al. (1964)	200 minutes a week for 4 weeks	13.3
Sommers, Schaeffer, Leiss, Gerber, Bray, Fundrella, et al. (1966)	40 minutes of therapy a week for 8.5 months	22.7
Tufts & Holliday (1959)	60 minutes a week for 7 months	28
Wilcox, Kouri, & Caswell (1991)	90 minutes a week for 3 months in individual condition and 360 minutes a week in group condition for three months	90

Calculated average: 20 hours (outliers removed: Robertson, 1997; Cole & Dale, 1986) to 30.4 hours (all studies). Adapted from Law et al. (2003).

of the previously-noted assumptions allowed for a conservative estimate of the cost associated with 1:1 speech-language interventions among children with FASD and SLD for Canada in 2011 to be made.

All cost figures are presented in 2011 Canadian dollars.

### Sensitivity analysis

The main analysis was performed using the estimated pooled prevalence of SLD among individuals with FASD. Subsequently, a sensitivity analysis was performed using the 95% confidence interval (CI) as the lower and upper estimates.

## Results

### Estimated number of children with FASD by age group and sex in Canada in 2011

In Canada in 2011, it was estimated that, in total, there were 63,637 children (32,650 boys and 30,986 girls), aged 2–19, with FASD.

### Estimated number of children with FASD and SLD by age group and sex in Canada in 2011

(1) *Systematic literature search.* The systematic literature search revealed seven studies that reported the prevalence of SLD among individuals with FASD (CDC, 1995; Church et al., 1997; Egeland et al., 1998; Elliott et al., 2008; Kvigne et al., 2004; Spohr et al., 1994; Steinhausen et al., 1982; Table II).

(2) *Meta-analysis.* The random effects analysis of the seven studies that reported the prevalence of SLD among children with FASD indicated an overall pooled prevalence of 59.6% (95% CI = 43.4–74.8%). The  $I^2$  statistic demonstrated that heterogeneity between studies was present [ $I^2 = 92.0%$ ,  $p = .000$ ]. Figure 1 depicts the Forest plot for the meta-analysis of the double arcsine-transformed prevalence of SLD among children with FASD with the studies presented in the order they appear in Table I.

The funnel plot (see Figure 2) of all studies on the prevalence of SLD among children with FASD (with the double arcsine-transformed prevalence plotted against the standard error of the double arcsine-transformed prevalence estimates) formed a random scatter plot; since random effects were present, the funnel plot could be used to determine if publication bias was present.

### Estimation of the number of children with FASD and SLD by age group, sex, and level of severity in Canada in 2011

By applying the pooled prevalence of 59.6% (calculated in the meta-analysis described above), it was estimated that there were 37,928 children with FASD and SLD in Canada in 2011. There were 17,067 children with FASD who had mild SLD (8757 boys and 8311 girls), and 20,860 children with FASD who had moderate-to-severe SLD (10,703 boys and 10,157 girls; Table III).

Table II. Studies reporting on the prevalence of SLD among children with FASD.

Condition/reference	ICD-10 Code	Country	Sample size/diagnosis	Age (mean)	FASD (% affected)
Specific developmental disorders of speech and language	F80				
CDC (1995)		USA	60 FAS	8	40%
Church et al. (1997)		USA	22 FAS	11.5	90%
Egeland et al. (1998)		USA	145 FAS	.6 (median)	41%
Elliott et al. (2008)		Australia	92 FASD (25 FAS + 65 pFAS + 2 suspected FAS)	3.3 (median)	60%
Kvigne et al. (2004)		USA	78 FASD (43 FAS + 35 pFAS)	FAS (10.0); pFAS (9.8)	58%
Spohr et al. (1994)		Germany	44 FASD (8 FAE + 36 FAS)	15.3	37%
Steinhausen et al. (1982)		Germany	68 FAS	4.3	88%
Specific speech articulation disorder	F80.0				
Church et al. (1997)		USA	22 FAS	11.5	10%
Expressive language disorder	F80.1				
Church et al. (1997)		USA	22 FAS	11.5	76%
Receptive language disorder	F80.2				
Church et al. (1997)		USA	22 FAS	11.5	82%
Stuttering [stammering]	F98.5				
Church et al. (1997)		USA	22 FAS	11.5	5%
Cluttering	F98.6				
Church et al. (1997)		USA	22 FAS	11.5	5%

FAE, Foetal Alcohol Effects; FAS, Foetal Alcohol Syndrome; FASD, Foetal Alcohol Spectrum Disorder; ICD-10, International Classification of Diseases, Version 10; pFAS, Partial Foetal Alcohol Syndrome.

Note: Meta-analysis was based on F80 code ("Specific developmental disorders of speech and language") only.

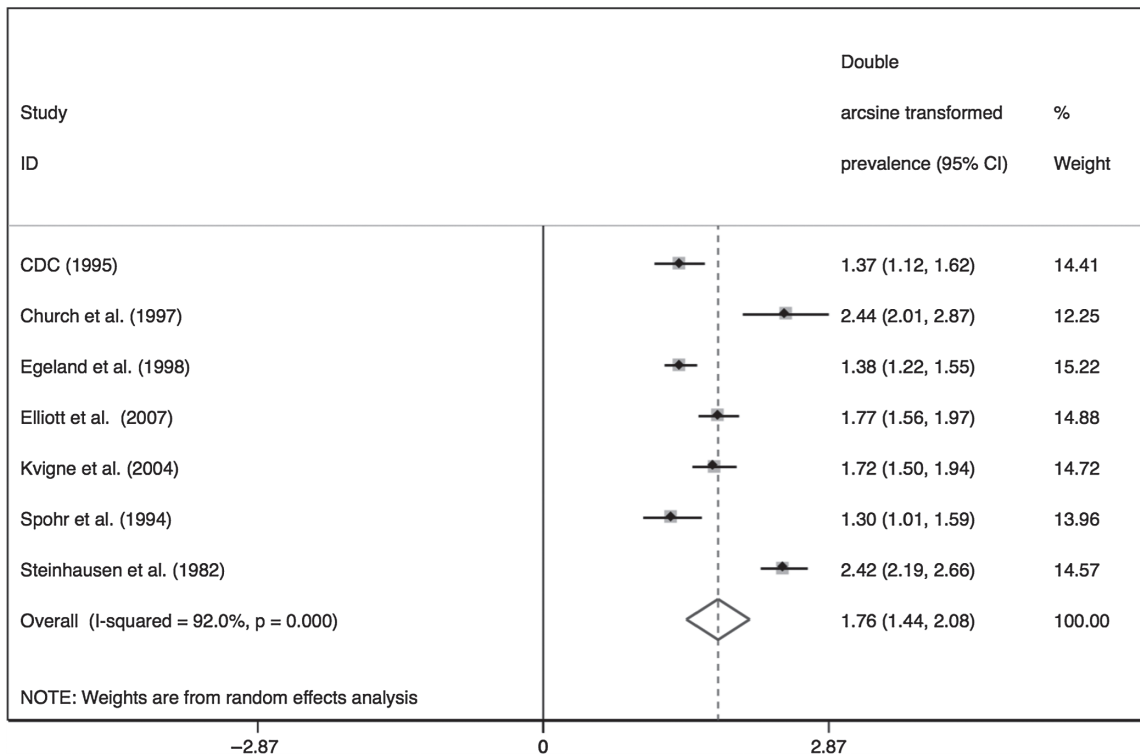


Figure 1. Forest plot of the seven studies that reported the prevalence of SLD among children with FASD. Note: The size of the box around the point estimate is representative of the weight of the estimate used in calculating the aggregated point estimate. CI, Confidence Interval.

*Estimation of the cost association with 1:1 speech-language interventions among children with FASD and SLD by age group, sex, and level of severity in Canada in 2011*

It was estimated that the total cost associated with 1:1 speech-language interventions for children with FASD with mild SLD ranged from \$25.6–50.9 million. For children with FASD with moderate-to-severe SLD, the total costs associated with 1:1 speech-language interventions ranged from \$46.9–94.8 million. The total cost of 1:1 speech-language interventions for children with FASD ranged from \$72.5–144.1 million (Table III). Please

note that the estimated number of children with FASD and SLD, and the costs associated with 1:1 speech-language interventions by age group, sex, and level of severity for each province/territory in Canada in 2011, are available from the authors.

*Sensitivity analysis*

Assuming that 43.4% (as the lower estimate) of children with FASD have SLD, it was estimated that there were 27,618 children with FASD and SLD (12,428 with mild SLD and 15,190 with moderate-to-severe SLD) in Canada in 2011. In turn, it was estimated that the cost associated with 1:1 speech-language interventions for these children ranged from \$52.8–104.9 million. Assuming that 74.8% (as the upper estimate) of children with FASD have SLD, it was estimated that there were 47,600 children with FASD and SLD (21,420 with mild SLD and 26,180 with moderate-to-severe SLD) in Canada in 2011. The resulting estimated cost associated with 1:1 speech-language interventions for these children ranged from \$91–180.9 million. The results of the sensitivity analysis are presented in Table III.

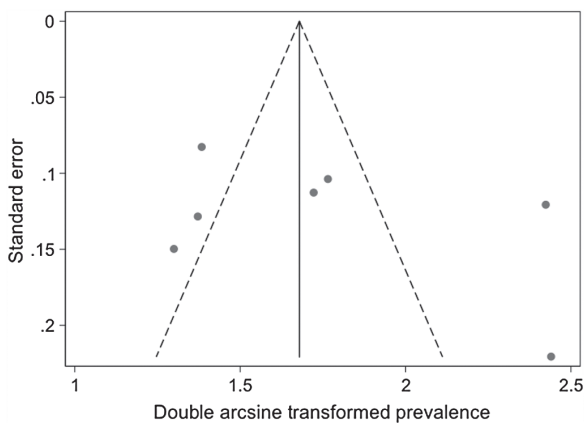


Figure 2. Funnel plot of the seven studies that measured the prevalence of SLD among children with FASD used in the meta-analysis with pseudo 95% Confidence Interval.

**Discussion**

This is the first study that has attempted to estimate the number of children with FASD and SLD and the cost associated with speech-language interventions (not only in Canada, but also in any other country of the world). The results of this study demonstrate that

Table III. Total number of children with FASD and SLD, and the cost associated with 1:1 speech-language interventions by age group and sex in Canada in 2011.

Sex	Age group; severity level of impairment	Total population (2011) <sup>a</sup>	Estimated number of children with FASD <sup>b</sup>	Number of children with FASD and SLD <sup>c,d</sup>	Total cost of 1:1 speech-language interventions for children with FASD <sup>e,f</sup>	
					Lower boundary (20 hours; \$75 per hour)	Upper boundary (30 hours; \$149 per hour)
Boys	2-4 years	588,548	5,297	3,157	\$6,037,708	\$11,994,913
	Mildly impaired			1,421	\$2,130,956	\$4,233,499
	Moderately-to-severely impaired			1,736	\$3,906,752	\$7,761,414
	5-9 years	939,635	8,457	5,040	\$9,639,387	\$19,150,248
	Mildly impaired			2,268	\$3,402,136	\$6,758,911
	Moderately-to-severely impaired			2,772	\$6,237,250	\$12,391,337
	10-14 years	975,941	8,783	5,235	\$10,011,837	\$19,890,183
	Mildly impaired			2,356	\$3,533,590	\$7,020,065
	Moderately-to-severely impaired			2,879	\$6,478,248	\$12,870,118
	15-19 years	1,123,671	10,113	6,027	\$11,527,348	\$22,900,997
Girls	Mildly impaired			2,712	\$4,068,476	\$8,082,705
	Moderately-to-severely impaired			3,315	\$7,458,872	\$14,818,292
	2-4 years	560,268	5,042	3,005	\$5,747,593	\$11,418,552
	Mildly impaired			1,352	\$2,028,562	\$4,030,077
	Moderately-to-severely impaired			1,653	\$3,719,031	\$7,388,475
	5-9 years	885,964	7,974	4,752	\$9,088,795	\$18,056,405
	Mildly impaired			2,139	\$3,207,810	\$6,372,849
	Moderately-to-severely impaired			2,614	\$5,880,985	\$11,683,556
	10-14 years	924,042	8,316	4,957	\$9,479,423	\$18,832,455
	Mildly impaired			2,230	\$3,345,679	\$6,646,749
Total	Moderately-to-severely impaired			2,726	\$6,133,745	\$12,185,706
	15-19 years	1,072,690	9,654	5,754	\$11,004,351	\$21,861,978
	Mildly impaired			2,589	\$3,883,889	\$7,715,992
	Moderately-to-severely impaired			3,165	\$7,120,463	\$14,145,986
	Sensitivity analysis, assuming 43.4% (lower estimate) of children with FASD have SLD			37,928	\$72,536,442	\$144,105,731
	Mildly impaired			12,428	\$18,642,459	\$37,036,352
	Moderately-to-severely impaired			15,190	\$34,177,842	\$67,899,979
	Total	7,070,759	63,637	27,618	\$52,820,301	\$104,936,331
	Sensitivity analysis, assuming 74.8% (upper estimate) of children with FASD have SLD					
	Mildly impaired			21,420	\$32,130,321	\$63,832,238
Moderately-to-severely impaired			26,180	\$58,905,589	\$117,025,770	
Total	7,070,759	63,637	47,600	\$91,035,910	\$180,858,009	

FASD, Foetal Alcohol Spectrum Disorder; SLD, speech-language disorder(s).

Note: Column numbers may not add up due to rounding error.

<sup>a</sup>Obtained from Statistics Canada (2012).

<sup>b</sup>Calculated based on a prevalence of nine per 1000 (Roberts & Nanson, 2000).

<sup>c</sup>Based on data by Coggins et al. (2007).

<sup>d</sup>Calculated based on a prevalence of 596 per 1000.

<sup>e</sup>Calculated using data on the average number of hours adapted from Law et al. (2003).

<sup>f</sup>Calculated using data on the average cost per hour for a speech-language pathologist obtained from the Canadian Association for Speech-Language Pathologists and Audiologists (CASLPA).

the number of children with FASD and SLD and the annual cost associated with 1:1 speech-language interventions in Canada is substantial. It is important to highlight that the cost of speech-language interventions is only part of the overall direct healthcare costs associated with FASD in Canada.

There are several limitations of the current study which have to be considered. To begin, it should be noted that the prevalence of FASD is unknown in Canada; therefore, for the purpose of this analysis, the only available and most commonly cited rough estimate of nine per 1000 (Roberts & Nanson, 2000) was used to estimate the number of children with FASD, which may not be accurate.

Further, the presented cost of speech-language interventions may be over-estimated due to the following reasons. First, it was assumed that all children with FASD and SLD require and receive 1:1 speech-language interventions. Second, the severity distribution of SLD was taken from the study of Coggins et al. (2007), where 69% of children with FASD had a significant language impairment. However, more than 40% of these children had experienced substantial adverse environmental conditions (e.g., abuse, neglect, negative caregiving, etc.). Such environments are likely to exert a negative influence on language development, particularly social communication, in a way that children with FASD without these adverse environments would not encounter. Third, the available literature on the prevalence of SLD among children with FASD, which was used to calculate the pooled FASD prevalence in this study, is dominated by FAS-specific data (Table II).

Furthermore, it should be considered when interpreting these findings that there is no agreement as to the number of treatment sessions or the length of the sessions that speech-language pathologists should provide for specific disorders, and it is likely that interventions are client-specific rather than disorder-specific. As such, there is the possibility that the number of hours for speech-language interventions may be much longer or shorter for some children than those used in this study. For example, according to Dr Larry Burd (Director), the North Dakota Foetal Alcohol Syndrome Centre offers younger children and intensive therapy for a longer duration. Most children with FASD in this centre receive therapy at least once a week, with many receiving therapy two or more times a week, and therapy often continues for a couple of years. For at least 15% of children with FASD at this centre, therapy will last for 4–6 years. Depending on the length of the intervention, the costs could fluctuate (can be higher or lower).

Moreover, speech-language interventions can take on many forms, for example, clinic-based or parent-administered and 1:1 sessions or group sessions. Like all types of disability, there is no cure for SLD; however, there are effective therapies. Children with SLD often qualify for an Individualized Education Plan, as well as other particular services. These

services typically include 1:1 interventions with a speech-language pathologist. Even though 1:1 intervention may be advantageous for some SLD, there are other treatment models available and utilized.

The current estimates provide both national and international policy-makers and decision-makers alike with a clear perspective on the magnitude of the burden and cost associated with children with FASD and SLD. The findings of this study are important for a variety of reasons, namely: (1) early intervention may improve the outcome of children with FASD and SLD; (2) early intervention may reduce rates or risk of developing secondary disabilities; (3) speech-language pathologists have an opportunity to play a key role in first appreciating risk factors for FASD; (4) speech-language interventions settings may be one of several optimal locations to screen for FASD; (5) supporting the implementation of prevention strategies to reduce/eliminate the consumption of alcohol during pregnancy could have a cost-saving effect; (6) they will raise awareness of not only the consequences of consuming alcohol during pregnancy, but also of the wide range of impairments seen in individuals with FASD; and (7) they will lead to increased awareness of speech-language pathologists to the disproportionate number of children with FASD that they will likely work with during their careers. Moreover, the cost estimates presented can be used to promote the allocation of resources to programs specifically for children with FASD.

Given the high prevalence of SLD among children with FASD, speech-language pathologists will frequently come into contact with children affected by FASD. Speech-language pathologists working in early intervention, serving children with birth defects, behavioural problems, developmental delays, and children in foster care or juvenile corrections settings may have significantly high rates of children with FASD in their caseload. Unfortunately, FASD is not widely recognized by healthcare practitioners (Clark & Tough, 2003); therefore, FASD is largely underdiagnosed (Paintner, Williams, & Burd, 2012). Early screening may lead to early diagnosis, which can lead to early participation in developmental interventions, and this, in turn, can prevent the development of secondary disabilities and improve the quality-of-life for people with FASD and their families (see for example, Paintner et al., 2012).

Speech-language pathologists are in a unique position, as they may be one of the first contacts with the healthcare system for children with FASD; there are a number of ways in which this opportunity can be maximized. First, it would be useful to review the pre-service training and in-service training to determine if additional information on FASD may be needed, such as FASD recognition and the specific needs of children with FASD. Second, improving the capacity for speech-language pathologists to effectively screen children on their caseload for FASD



and provide referrals to diagnostic clinics where a diagnosis can be established, if appropriate. The FASD Canadian Diagnostic Guidelines clearly states that speech-language pathologists should be part of the core diagnostic team (Chudley et al., 2005). Third, improved screening and increased training on FASD could increase referral rates for other related services (audiologists, occupational therapists, physical therapists, etc.). A comprehensive approach to intervention and early participation of the other disciplines, where needed, could increase the amount of early intervention and, thus, could potentially improve outcomes (Olson, Jirikowic, Kartin, & Astley, 2007).

Lastly, the high rates of SLD among children with FASD suggest that there is an increased need for comprehensive treatment plans that specifically target individuals with FASD. Designing targeted interventions for children with FASD and SLD may have implications for learning and behaviour, interpersonal communication, and social skill development (Wyper & Rasmussen, 2011).

According to clinical observations, the link between SLD and behaviour problems is common. Children with SLD appear to be over-represented in the group of children with temper tantrums, sleep disturbance, increased irritability, aggression, and attention deficit hyperactivity disorder. Speech-language pathologists are often the first professionals to begin the process of managing these behavioural problems in the context of therapy. Anecdotal data suggest that this strategy is often effective. Further research to understand improved management pathways may be very useful for speech-language pathologists working with this population. Further efforts to better understand the link between SLD and behavioural disorders may have important influence on improved early intervention and decreasing the risk of developing secondary disabilities in this population.

Further studies are needed to determine the types of speech-language interventions children with FASD are currently receiving and whether children with FASD are effectively treated with the same interventions developed for children without FASD. Such information is necessary to determine if program accommodations are necessary for children with FASD. In addition, the limitations of the current study draw attention to the need for accurate prevalence estimates of SLD among individuals with FASD, and the need for cost analyses to be conducted using other delivery methods, not only in Canada, but in other countries as well.

Although this study has several limitations, it does provide a working estimate, which is a powerful tool for understanding the enormous economic burden that FASD has on the healthcare system overall—speech-language interventions being only one facet of the total healthcare cost. Additional research can refine these estimates over time.

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### Notice of correction

The version of this article published online ahead of print on 18 Dec 2013 contained two errors on page 7. In Table III, the column total reported for the number of children with FASD and SLD “33,091” should have read “37,928”, and the row heading “Sensitivity analysis, assuming 74.8% (lower estimate) of children with FASD and SLD” should have read “Sensitivity analysis, assuming 74.8% (upper estimate) of children with FASD and SLD”. Lastly, the Acknowledgements were omitted. This omission and the above errors have been corrected for this version.