POLICY

What Do We Know about the Economic Impact of Fetal Alcohol Spectrum Disorder? A Systematic Literature Review

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Abstract — Aims: The objective of this study was to conduct a systematic review of the literature related to the measurement of the economic impact of Fetal Alcohol Spectrum Disorder (FASD) in different countries and to categorize the available literature. Methods: A systematic literature search of the studies concerning the economic impact of FASD was conducted using multiple electronic bibliographic databases. Results: The literature on the economic burden of FASD is scarce. There are a limited number of studies found in Canada and the USA, and data from the rest of the world are absent. Existing estimates of the economic impact of FASD demonstrate significant cost implications on the individual, the family and society. However, these estimates vary considerably due to the different methodologies used by different studies. Conclusion: Limitations and gaps in the existing methodologies of calculating the economic costs of FASD are discussed. It is evident that there is an urgent need to develop a comprehensive and sound methodology for calculating the economic impact of FASD to the society.

INTRODUCTION

Fetal Alcohol Spectrum Disorder (FASD) is a serious public health, social and economic issue that affects people throughout the world. FASD is a non-diagnostic umbrella term used to describe the range of disabilities that may affect people whose mothers consumed alcohol during pregnancy. This term covers several alcohol-related medical diagnoses, which include: fetal alcohol syndrome (FAS), partial fetal alcohol syndrome (pFAS), alcohol-related neurodevelopmental disorder (ARND) and alcohol-related birth defects (ARBD). The disabilities involve a wide continuum of challenges from mild to very serious disabilities, which affect an individual throughout the course of their life.

People who are affected by this disability most often experience an array of health problems such as birth defects, growth problems, cognitive delay, and speech and language difficulties. Furthermore, those who are affected by FASD are also more susceptible to cardiac anomalies, urogenital defects, skeletal abnormalities, and visual and hearing problems.

Due to the possibility of a wide range of disabilities, people who are affected by FASD may have special needs that require life-long help. Without the crucial support, people affected by FASD are at a high risk of developing secondary disabilities such as: mental health problems, trouble with the law, dropping out of school, becoming unemployed, homeless and/or developing alcohol and drug problems. This in turn produces tremendous costs to the society.

Estimation of FASD cost, especially lifetime cost, is central to describing the extent of the problem and to evaluate the benefits to society of prevention programs and thus, useful from a public policy perspective (Harwood and Napolitano, 1985; Bloss, 1994; Public Health Agency of Canada, 2008). According to the revised International Guidelines for Estimating the Costs of Substance Abuse (Single *et al.*, 2001), cost estimates help to prioritize substance abuse issues, provide useful information for targeting programming and identify information gaps. The development of improved cost estimates also offers the potential to develop more complete costbenefit analyses of policies and programs aimed at reducing the harm associated with the use of psychoactive substances.

It is necessary to begin the discussion of economic cost studies with those from other professions by elaborating the assumptions and terms that the economists use. The International Guidelines (Single et al., 2001) outlines the study of the economic costs of problems associated with the use of psychoactive substances as follows: (i) a type of cost-of-illness study (ii) in which the impact of substance abuse on the material welfare of a society is estimated by examining (iii) the social costs of resources expended for treatment, prevention, research and law enforcement, plus (iv) losses of production due to increased morbidity and mortality, plus (v) some measure for the quality of life years lost, relative to a counterfactual scenario in which there is no substance abuse. For further explanation of each part of the above statement please, see a Supplementary data (Glossary of common terms used in economic cost studies adapted from Single et al., 2001).

Few studies to date have overviewed the literature on the cost associated with FASD in the USA (see for example, Lupton, 2003; Lupton *et al.*, 2004). The objective of this study was to conduct a systematic review of the literature related to the measurement of the economic impact and cost drivers associated with FASD in different countries and to categorize the available literature.

METHODS

Systematic literature search

A systematic literature search of studies concerning the economic impact of FAS/FASD was conducted using 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, CINAHL and OVID (combines several databases), Social Work Abstracts, Epscohost, the Cochrane Database of Systematic Reviews, Canadian Centre on Substance Abuse Library Collection Database, Centre for Addiction and Mental Health Library Database, Criminal Justice Abstracts and Google Scholar.

In addition, the following economic databases were searched: the Alcohol Database ETOH (http://etoh.niaaa.nih. gov/Archive.htm) and the NHS EED (http://www.crd.york. ac.uk/crdweb/).

Moreover, other web sites were searched for relevant literature: Alberta Alcohol and Drug Abuse Commission; Canadian Institutes of Health Research; Canadian Paediatric Society; Canadian Public Health Association; Centre of Excellence for Early Childhood Development; Centres for Excellence in Women's Health; Health Canada; Journal of Fetal Alcohol Research; National Center on Birth Defects and Developmental Disabilities; Public Health Agency of Canada (PHAC); SAMHSA FASD Center for Excellence; Society of Obstetricians and Gynaecologists of Canada; Status of Women Canada; The Women's Addiction Foundation; HRSDC Office of Disability Issues; INAC; FNIHB; Centres for Excellence for Children with Special Needs and Centers for Disease Control and Prevention, USA.

In addition, manual reviews of the content pages of the major epidemiological journals were conducted, as well as citations in the relevant articles. Experts in the relevant field were also consulted in order to obtain more comprehensive data. The search was not limited geographically or to only English language publications. The available published and unpublished literature was searched from January 1960 to July 2010, inclusive.

The search was conducted using multiple combinations of the following key words, in a systematic manner:

- (1) disease conditions: FASD, FAS, pFAS, fetal alcohol effects (FAE), ARND, ARBD, as well as prenatal alcohol exposure, pregnancy and alcohol use/abuse;
- (2) outcomes: disability, disability adjusted life years, quality adjusted life years, morbidity, premature mortality, potential years of life lost and productivity losses;
- (3) cost: social cost, economic cost, direct and indirect costs and intangible cost;
- (4) systems/categories of cost: health care (hospitalization, hospital days, ambulatory care, emergency room visits, family physician visits and prescription drugs), mental health, addiction services, child welfare, early childcare, education (special needs, assessment, suspensions, staff time and salaries),

social services (home support services, residential care and respite care).

Data extraction

Information from the identified studies was independently extracted by two investigators (D.B. and S.P.). Training of coders to achieve sufficient (>0.80) interrater reliability (IRR) was conducted. In order to calculate IRR, Fleiss' kappa statistics using attribute agreement analytic method was used. All analyses related to IRR were computed using Minitab[®] statistical software (2007). A third investigator (S.L.) checked the table entries for accuracy, against the original article.

Using a standardized spreadsheet (MS-Excel), each study was coded for the following variables: reference, year(s) of study, country where the study was done, and direct, indirect and other cost drivers.

RESULTS

Initially, the literature search identified 233 abstracts. One hundred and thirty-eight abstracts were excluded because the studies were not concerning the economic impact of FAS/ FASD. After reviewing the remaining 95 articles, 72 were excluded due to the absence of data on cost drivers associated with FASD. Upon further screening, only 13 well-documented cost studies with comprehensive methodologies were selected for data extraction: 3 studies from Canada and 10 studies from the USA. There were no studies estimating the cost of FASD found for any other countries other than Canada and the USA.

The results of the systematic search strategy are shown in Fig. 1.

Interrater reliability

There was a very high IRR ($\kappa = 0.81$, P < 0.0001) among the two reviewers across all variables coded. Discrepancies were reconciled by a third investigator (J.R.), independent of the first process.

Derivation of costs for comparison

All studies used the local currency for estimating costs and used the same currency year as the costing year. In order to facilitate comparison, the estimated costs in the Canadian studies were converted to May 2010 currency values, using the inflation calculator of the Bank of Canada (http://www.bankofcanada.ca/en/rates/inflation_calc. html). The estimated costs in the USA studies were converted to June 2010 currency values using the currency inflation rates calculated from the consumer price indexes supplied by US Department of Labor (ftp://ftp.bls.gov/ pub/special.requests/cpi/cpiai.txt).

Canadian studies (all costs in Canadian dollars)

The economic impact of FASD was measured in three Canadian-based studies (Stade *et al.*, 2006, 2009; Thanh and Jonsson, 2009). Table 1 presents the cost estimates of FASD from the Canadian studies.

Computer assisted + manual search Key words: fetal alcohol spectrum disorder (FASD), fetal alcohol syndrome (FAS), partial fetal alcohol syndrome (pFAS), fetal alcohol effects (FAE), alcoholrelated neurodevelopmental disorder (ARND), alcohol-related birth defects (ARBD), prenatal alcohol exposure, pregnancy, alcohol use/abuse, disability, disability adjusted life years (DALY), quality adjusted life years (QALY), morbidity, premature mortality, potential years of life lost (PYLL), productivity losses, social cost, economic cost, direct and indirect costs, intangible cost, health care (hospitalization, hospital days, ambulatory care, emergency room visits, family physician visits, prescription drugs), mental health, addiction services, child welfare, early childcare, education (special needs, assessment, suspensions, staff time and salaries), social services (home support services, residential care, respite care). Excluded N = 138 N = 95 screened for cost drivers associated with FAS/FASD Excluded N = 72 N = 23 identified for further review Excluded N = 10 N = 13 with comprehensive methodologies selected for data extraction: 3 Canadian studies & 10 studies from the USA

Fig. 1. Schematic diagram of the systematic search strategy.

Stade et al. (2006) measured the economic impact of FASD for 2003 using a modified version of the Health Services Utilization Inventory (Browne et al., 2001) on a sample of 148 parents (biological, adoptive or foster) who were either living with or responsible for the care and welfare of an FASD-affected child. Participants were between the ages of 1 and 21, and were diagnosed with either FAS or FAE. At the patient level, the estimated total adjusted annual cost associated with FASD per child was \$14,342 [95% confidence interval (CI): \$12,986-\$15,698]. At the population level, using a conservative prevalence rate of 3 per 1000 (the literature reports up to 9 per 1000 in Canada; Public Health Agency of Canada, 2003), the cost of FASD annually was \$344.2 million (95% CI: \$311,664,000-\$376,752,000). This prospective cross-sectional research study estimated costs from the perspectives of the society, the Ministry of Health and the patient/family. The cost components included were: direct costs, such as medical care (hospital admissions, health professional services and medication), educational services (home schooling, special schooling and residential programs), social services (respite care, foster care and legal aid), indirect costs (productivity losses; measured using the human capital approach) and other costs (out-of-pocket).

Stade *et al.* (2009) attempted to overcome the limitations of their past study (Stade *et al.*, 2006) by including the cost for infants from the day of birth to 1 year of age, the cost for adults beyond the age of 21 (up to the age of 53) and the cost of children residing in institutions (for a total of 250 participants with diagnosed FAS, pFAS or ARND), as well as by adding a few more cost components (e.g. residential programs, job education, institutionalization and government pensions). The adjusted annual cost of FASD at the patient level, for 2007, was estimated as \$21,642 (95% CI: \$19,842–\$24,041), while at the population level, the estimated adjusted annual cost was \$5.3 billion (95% CI: \$4.12 billion–\$6.4 billion), for persons 0–53 years of age.

Thanh and Jonsson (2009) estimated two societalperspective costs in Alberta, Canada: the annual long-term

				Direct cost,	, cost; percei	ntage of tota	ul cost	T	Other cost				
deference	Year of study; province (if applicable)	РЛ рег 1000	Age	Health care	Education	Social services	Total direct costs	Induced cost (productivity losses), cost; percentage of total cost	(out or pocket), cost; percentage of total cost	Annual cost for all persons with FASD (95% CI)	Annual cost per individual (95% CI)	Adjusted ^a annual cost for all persons with FASD (95% CI)	Adjusted ^a annual cost per individual (95% CI)
stade <i>et al.</i> (2006)	2003	3 (P)	1–21	\$3976; 30.3%	\$4275; 32.6%	\$2866; 21.9%	\$11117; 84.8%	\$1055; 8.1%	\$936; 7.1%	\$344.2 M (\$311.7 M-\$376.8 M)	\$14,342 (\$12,986- \$15,608)	\$390.2 M (\$353.4 M-\$427.2 M)	\$16,259 (\$14,722- \$17,796)
stade <i>et al.</i> (2009)	2007	3 (P)	0-53	\$6630; 35%	\$5260; 28%	\$4075; 18.8%	\$15,965; 82%	\$1431; 6.6%	\$2814; 13%	\$5.3 B (\$4.12 B- \$6.4 B)	\$21,642 (\$19,842– \$24.041)	\$5.5 B (\$4.3 B- \$6.7 B)	\$22,473 (\$20,604– \$24 965)
Thanh and Jonsson (2009) (Stade et al., 2006, cost data used)	2002–2005 (2008 CND); Alberta	3 and 9 (I)	0-71.6	Included; 30.3%	Included; 32.6%	Included; 21.9%	84.8%	Included; 8.1%	Included; 7.1%	\$130 M-\$400 M (long-term cost); \$48 M-\$143 M (short-term cost)	\$1.1 M (lifetime cost per individual)	\$148.4 M-\$428.4 M (long-term cost); \$48.5 M-\$144.5 M (short-term cost)	\$1.12 Note cost per individual)

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Table 1. Annual cost and lifetime costs (in Canadian dollars) per individual associated with FASD in Canadian studies

cost and the annual short-term cost of FASD. The annual long-term cost refers to the projected amount of money incurred by the cohort of children born with FASD each year (lifetime cost), which was calculated using the FAS incidence rates of 3 per 1000 live births (lower rate) and 9 per 1000 live births (upper rate) along with the total number of live births per year in 2002-2005. The annual short-term cost refers to the amount of money incurred by people who are presently living with FASD, calculated using the FASD cost calculator developed by the FAS centre at the University of Dakota (available at http://www.online-clinic. com). The cost calculator included the following cost components: healthcare costs, special education, juvenile justice services, adult corrections and service delivery systems. The costs and disorder rates used in the calculator were in 1996/ 97 United States Dollar (USD), and were adjusted to 2008 Canadian Dollar (CND) value using a 5% discount rate and a USD/CND exchange rate of 1.03. The lifetime cost of caring for each individual child born with FASD was estimated as ~\$1.1 million (annual cost per child was used from Stade *et al.*, 2006): $$15,812 \times average$ age at death (71.6). The total annual cost of FASD (short-term cost) for Alberta was estimated as \$48 million (based on the lower incidence rate) to \$143 million (based on the upper incidence rate), and the daily cost as \$105,000 (lower incidence rate) to \$316,000 (upper incidence rate). It is also worth noting that the long-term economic cost for the disorders associated with FASD rose from \$130 to \$400 million from 2002 to 2005, respectively.

USA studies (all costs in US dollars)

The economic impact of FAS was measured in 10 American-based studies (Harwood et al., 1984, 1998; Harwood and Napolitano, 1985; Abel and Sokol, 1987, 1991a,b; Weeks, 1989; Rice et al., 1990, 1991; Rice, 1993; Harwood, 2000, 2003). Please note that Harwood et al. (1984) and Harwood and Napolitano (1985) as well as Rice et al. (1990, 1991) were published in repetition. Only two studies from the USA estimated the total lifetime cost for a person with FAS (Harwood and Napolitano, 1985; Weeks, 1989). Table 2 presents the cost estimates of FAS from the USA studies.

Using a societal perspective and data from Russell (1980), Harwood et al. (1984) and Harwood and Napolitano (1985) generated 1980 cost estimates of between \$1.937 billion and \$9.687 billion with a median estimate of \$3.236 billion, using alternative FAS incidence rates (based on a review of prospective studies) of 1.0, 5.0 and 1.67 per 1000 live births, respectively. The total lifetime cost was estimated at \$596,000 per FAS birth. Costs included estimates of medical treatment, home and residential care, special educational services and productivity losses for patients with FAS of all ages (0-65). Based on this lifetime estimate, Lupton et al. (2004) demonstrated how these costs for one case of FAS are spread out over the person's lifetime. After adjustment for inflation the \$596,000 in 1980 became \$2 million in 2002. This estimate is composed of \$1.6 million (80%) for health care, special education and residential care for individuals with mental retardation, and \$0.4 million (20%) for productivity losses (Lupton et al., 2004). Furthermore, the cumulative cost of one case of FAS to age 65 would have a

billion; I, incidence; M, million; P, prevalence.

inflation (May 2010)

for

^aAdjusted

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				Dir	ect cost, cost; percentage of	Indirect cost (productivity	Other direct	Annual		Adjusted ^a annual cost	Adjusted ^a		
Reference	Year of study; state	P/I per 1000	Age	Health care	Residential care and home care	Special education	Total direct costs	losses), cost; percentage of total cost	cost, cost; percentage of total cost	cost for all persons with FAS	Lifetime cost per individual	for all persons with FAS	lifetime cost per individual
Harwood <i>et al.</i> (1984); Harwood and Napolitano (1985)	1980	1.0, 5.0 and 1.67 (cost estimates based on 1.67)	0–65	\$699 M (children \$125 M; adults \$574 M); 22%	HC and RC with day services: \$694 M; 22%	\$990 M; 31%	\$2.4 B; 75%	\$853.3 M; 25%	Not included	\$3.2 B	\$596,000	\$8.5 B	\$1.6 M
Abel and Sokol (1987)	1984	1.9	0–21	Growth retardation: \$118 M; Cleft palate, tetralogy of fallot and sensorineural anomalies: \$18M; 42%	24-h RC due to MR: 109 M; semi-independent supervised support \$75.8 M; 58%	Not included	\$321 M	Not included	Not included	\$321 M	n/a	\$674.1 M	n/a
Weeks (1989) Abel and Sokol (1991a) (revision	1988, Alaska 1984	1.67 0.33	0–65 0–21	Included Treatment cost: \$16.9 M; 22.7%	Included 24-h RC due to MR: \$57.7 M; 77.3%	Included Not included	n/a \$74.6 M	Included Not included	Not included Not included	n/a \$74.6 M	\$1.4 M n/a	n/a \$156.7 M	\$2.5 M n/a
of 1987 study) Abel and Sokol (1991b) (revision of 1987 study)	1987	1.9	0–21	Treatment cost: \$104.5 M; 42%	24-h RC due to MR: \$145.2 M; 58%	Not included	\$249.7 M	Not included	Not included	\$249.7 M	n/a	\$479.4 M	n/a
Rice <i>et al.</i> (1990, 1991)	1985	1.9	0–65	Treatment: \$135 M; 8.4%	RC for age 21+: \$1287 B, 79.9%; Full-time RC <21 years \$110 M, 6.8%; Semi-independent supervised care \$76 M 4.7%	Not included	\$1.6 B	Not included	Research \$3 M; 0.2%	\$1.6 B	n/a	\$3.25 B	n/a
Rice (1993) (update of Rice <i>et al.</i> , 1990, 1991)	1990	1.9	0–65	Included	Included	Not included	n/a	Not included	Included	\$2.1 B	n/a	\$3.6 B	n/a
Harwood <i>et al.</i> (1998) (update of Harwood <i>et al.</i> , 1984 and Harwood and Napolitano, 1985 study)	1992	2	0–65	Included	Included	Included	n/a	Included	Not included	\$1.9 B	n/a	\$3.04 B	n/a
Harwood (2000) (update of Harwood at al = 1008)	1998	2	0–65	Included	Included	Included	\$2.9 B	\$1.25 B	Not included	\$4.2 B	n/a	\$5.5 B	n/a
et al., 1998) Harwood (2003) (updated Harwood, 2000)	2003	2	0–65	Included	Included	Included	\$3.9 B	\$1.5 B	Not included	\$5.4 B	n/a	\$6.5 B	n/a

Table 2	Annual	cost and	lifetime	cost (in U	S do	llars)	per	indivi	idual	associated	with	FAS	in the	USA	studies
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B, billion; HC, home care; I, incidence; M, million; MR, mental retardation; P, prevalence; RC, residential care. ^aAdjusted for inflation (June 2010).

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break down as follows: \$130,000 in the first 5 years, \$360,000 in 10 years, \$587,000 in 15 years and more than \$1 million in 30 years (Lupton *et al.*, 2004).

Abel and Sokol (1987) measured the economic cost of FAS from the perspective of the health care system from birth to 21 years of age, and estimated that the economic burden of FAS in the USA was \$321 million in 1984, using an incidence rate of 1.9 per 1000 live births. The incidence rate was an average drawn from several prospective and retrospective studies. The study estimated the cost of providing specialized services for pre- and post-natal growth retardation requiring neonatal intensive care; surgical repair of FAS-related birth defects and subsequent treatment; care for FAS patients with moderate or severe cognitive disabilities; and the cost of semi-independent supervised support for mildly cognitively disabled patients with FAS.

In 1991, Abel and Sokol (1991a) revised their 1987 study, with a much lower and more conservative incidence rate of 0.33 per 1000 live births; derived from prospective studies that were comprised of primarily Caucasian samples and did not include any other racial/ethnic groups (e.g. Native Americans). This study produced a much lower annual cost estimate of \$74.6 million. More than 77% of this economic burden was associated with residential care due to mental retardation of FAS individuals. However, Abel and Sokol (1991b) further refined their methodology and the costs included and excluded. The updated 1987 annual estimate of \$250 million was, again, based on an incidence rate of 1.9 per 1000 live births.

Again, from the perspective of the health care system, Rice *et al.* (1990, 1991), using the method of Abel and Sokol (1987), estimated the annual cost of treating birth defects associated with FAS in the USA in 1985 at \$1.6 billion, based on an incidence rate of 1.9 FAS cases per 1000 live births. The cost drivers included the cost of care for FAS-related birth defects and cognitive disability and residential care for patients with mental retardation over 21 years of age (up to age 65). Then, based on increasing population rates and healthcare costs, between 1985 and 1990, Rice (1993) projected a \$2.1 billion annual cost for 1990.

Harwood *et al.* (1998) estimated the 1992 annual cost to be \$1.944 billion based on a prevalence rate of 2.0 per 1000 live births. This study re-estimated: (i) the nature and cost of specific types of treatment, (ii) the proportion of FAS cases requiring services and (iii) the duration of services. This cost estimate included the following drivers: treatment and care services to age 21, home and residential care to age 65 of people with moderate to severe retardation, special education services and productivity losses.

Further, Harwood (2000, 2003) updated the 1992 study by adjusting for the change in national health care expenditures, the consumer price index for medical services, for changes in the USA adult population, and in the hourly compensation index for productivity losses. Based on these adjustments, estimated costs rose to \$4.15 billion by 1998 (direct cost \$2.9 billion; indirect cost \$1.25 billion), and to \$5.4 billion by 2003 (direct cost \$3.9 billion, 6.1% annual increase; indirect cost \$1.50 billion, 4% annual increase).

A study by Weeks (1989) reported that a lifetime cost for each child born with FAS is \$1,374 million in 1988. This study adapted the methodology used by Harwood and Napolitano (1985) and included the following costs: developmental disability services, special education, social service costs, adult vocational services and institutional care for mental retardation to age 65. This estimate is much higher than the study by Harwood and Napolitano (1985) estimate because costs in Alaska are generally higher than national costs.

DISCUSSION

Based on the few existing studies for Canada and the USA, it is clear that FASD is a serious public health problem and is associated with tremendous monetary costs.

Even though many cost components have been taken into account in the reviewed studies, the total cost associated with FASD is still underestimated. This is primarily due to the fact that several cost components have not been included in the studies to date, likely because the data are not readily available. Among those costs drivers commonly not included are: child welfare costs/payments, law enforcement costs, cost of research and prevention and intangible costs (i.e. the costs of pain and suffering), just to name a few. It is a very difficult task to estimate the cost of FASD since the total cost accrues from many sectors of society. Regardless, it is very important to include all cost components (as long as it is possible to estimate them in a reliable manner, but if not at least all cost components should, at the very least, be noted) in order to get a true and valid estimate, or at least as close to the reality as possible.

One important cost driver-the child welfare system-was not accounted for in any of the existing Canadian or USA estimates. However, there is reason to believe that children with FASD are overrepresented in the child welfare system and thus, must be included to get a true cost estimate (Farris-Manning and Zandstra, 2003; Hutson, 2006). Specifically, Fuchs et al. (2010) reported that in Manitoba, Canada 17% of children in care are affected by diagnosed or suspected FASD. In Alberta, it was determined that there are children in the care of 15.032 social services (Farris-Manning and Zandstra, 2003) and almost half of them have FASD (Hutson, 2006). In one of the first studies of its kind, Fuchs et al. (2008) reported the total annual cost of children in care in Manitoba as \$9.5 million for a sample of 400 children with FASD in 2006. These authors found that the daily cost of caring for a child with FASD in the child welfare system was \$65 (or \$23,760 per annum).

In addition, law enforcement cost was also not considered in the existing Canadian and USA estimates. It is reported that about 50% of young offenders in Canada have FASD (Zakreski, 1998). In one American study of 253 people with FAS or FAE, 60% reported ever being charged, convicted or in trouble with the authorities for any of a list of criminal behaviors, and 42% of adults had been incarcerated for a crime (Streissguth *et al.*, 1996).

There were several other important cost drivers not included in the USA estimates: medical services for physical anomalies, special education, substance abuse, mental health and vocational services, services for mild physical and learning disabilities and lost productivity of caregivers and FASD-affected persons. These insufficiencies have also been noted in several reviews (see Lupton, 2003; Lupton *et al.*, 2004; Hutson, 2006). In addition, the existing USA studies

included the cost of FAS only. However, the prevalence of FASD is suspected to be 10 times higher (Sampson *et al.*, 1997; May and Gossage, 2001; Astley, 2002), and therefore, the existing cost figures are likely underestimated.

In the USA studies, there is a broad range in the reported total cost figures. For instance, the annual cost estimate by Abel and Sokol (1991a) was reported as \$75 million for 1984 (\$157 million adjusted for 2010), while Harwood (2003) reported \$5.4 billion for 2003 (\$6.5 billion adjusted for 2010). These cost disparities reflect the fact that the studies have used different methodologies and assumptions. For example, the studies used different prevalence/incidence rates [e.g. the incidence rate was ranging in the USA studies from 0.33 to 1.9 (Abel and Sokol, 1987, 1991a,b)], used different age categories and different cost components. Another common problem was that the USA studies used terms 'incidence' and 'prevalence' interchangeably (please note that the authors of this paper used the original terminology from the studies). Therefore, it is not clear whether these cost studies were incidence or prevalence-based, which is very important from the economic point of view. The application of FAS prevalence rates to contemporary as well as older birth cohorts can also contribute to the large ranges observed (Lupton et al., 2004).

Another noted inconsistency was that the individual cost drivers accounted for different proportions of the total cost in each of the reviewed studies. Methodological differences can likely account for the different proportions reported. If certain cost components are not included, then other cost components will account for a higher percentage of the overall cost, thus depicting an unrealistic picture.

Furthermore, studies that used a sample to draw conclusions on the total costs incurred (e.g. Stade *et al.*, 2006, 2009) used a method of convenience sampling, which, as a result, limits the generalizability of the Canadian studies.

CONCLUSION

Based on the observed literature, currently, there are no comprehensive assessments of the economic impact of FASD in Canada, the USA or any other country. The majority of the studies that do exist limit their range of the cost components included, are inconsistent with one another or are only generalizable to certain populations. There is an urgent need to provide an accurate cost estimate of FASD that would encompass all aspects of this disorder and the various sectors affected by this disability. A standardized approach would allow for proper comparisons across studies, both nationally and internationally. It would also allow for comparisons between FASD and other public health issues, which is ideally required if the results will be used in economic evaluations and resource allocation decisions.

As evident from the observed methodologies of the existing studies, the next step is to develop a comprehensive and sound methodology for calculating the economic impact of FASD. The PHAC has undertaken the lead in the development of a sound methodological approach for calculating the economic impact of FASD. The current authors will build this methodological model for Canada based on the guidelines developed from the first national Roundtable on The Development of a Canadian Model for Calculating the Economic Impact of FASD, which was held by PHAC on March 21–22, 2007 in Ottawa (Public Health Agency of Canada, 2008). This cost-of-illness economic model will consider the systems that those with FASD come into contact with throughout their lives (i.e. a life-cycle approach); the life/developmental stages of those affected; different levels of severity of disability; the direct and indirect costs to the systems, individuals and families, including the loss of productivity of parents/caregivers, and the lost potential of the affected individuals. An inclusive cost estimate should not only take into consideration the costs accrued due to the illness, but should also account for preventative care, and the money that can be saved by effective social policies and programs. It is hoped that this model will be appropriate not only for use in Canada, but for use in other countries as well.

An accurate and comprehensive economic impact estimate is crucial in order to illustrate to policy developers and decisionmakers the extent of the problem. Thus, the importance of an economic impact model of FASD goes without saying. Once policy-makers can clearly see the impact of burden and cost that FASD has on not only Canada, but throughout the world as well, prevention initiatives are likely to soon follow.

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