ADDICTION
SSA SOCIETY FOR THE STUDY OF ADDICTION
REVIEW
doi:10.1111/add.14841

Fetal alcohol spectrum disorder: a systematic review of the cost of and savings from prevention in the United States and Canada

Jacob R. Greenmyer[1], Svetlana Popova[2], Marilyn G. Klug[3] & Larry Burd[1]

Department of Pediatrics, University of North Dakota School of Medicine and Health Sciences, Grand Forks, ND, USA,[1] Institute for Mental Health Policy Research, Centre for Addiction and Mental Health, Dalla Lana School of Public Health, University of Toronto, Toronto, Ontario, Canada[2] and Department of Population Health, University of North Dakota School of Medicine and Health Sciences, Grand forks, ND, USA[3]

ABSTRACT

Background and aims Fetal alcohol spectrum disorder (FASD) is a preventable condition that imposes a significant financial burden on societies. Funding of FASD prevention is a small portion of the total expenditures associated with FASD. This paper aimed to review the literature on the costs of and savings from prevention of FASD and present a model for the United States and Canada of projected savings based on expansion of existing evidence-based prevention models. **Methods** A systematic review of published literature on the cost of FASD prevention was conducted and experts in the field were interviewed. Studies that reported the cost of primary prevention of FASD were eligible for further consideration. **Results** Applying evidenced-based prevention programs to women at highest risk to have a future child with FASD greatly reduces the cost of prevention. In the United States, one case of FASD can be prevented for as little as USD $20 200 – 47 615. Cost of prevention is considerably less expensive than cost of care for a case of FASD. **Conclusion** Expansion of risk-based prevention strategies for fetal alcohol spectrum disorder in the United States and Canada would be an economically efficient and worthwhile investment for society.

Keywords Annual cost per case, cost, fetal alcohol spectrum disorders, fetal alcohol syndrome, mothers, prevention.

Correspondence to: Larry Burd PhD, North Dakota Fetal Alcohol Syndrome Center, University of North Dakota School of Medicine and Health Sciences, 1301 North Columbia Road Stop 9037, Grand Forks, ND 58202-9037 USA. E-mail: larry.burd@med.und.edu
Submitted 29 March 2019; initial review completed 10 June 2019; final version accepted 1 October 2019

INTRODUCTION

Fetal alcohol spectrum disorder (FASD) is a significant financial and emotional burden to society. Economic costs of FASD—a preventable disease—involve multiple sectors of society including direct costs (e.g. health care, education, social services, criminal justice system) and indirect costs (e.g. productivity losses) [1,2]. The medical costs associated with FASD are higher than costs for other conditions, such as autism spectrum disorder [1,3]. The costs accrued by FASD are the result of both primary fetal exposure to alcohol and adverse developmental outcomes. Primary disabilities in individuals with FASD include congenital malformations, growth impairments, intellectual disabilities and neurobehavioral disorders [4]. People with FASD are at increased risk for the development of secondary disabilities, such as difficulties at school, mental health disorders and involvement in the criminal justice system [5–7]. People with FASD typically require life-long assistance in many dimensions of their lives, which gives rise to the multi-faceted economic impact of this condition.

Annual estimates of nation-wide costs for all individuals with FASD in the United States have been mainly limited to people with fetal alcohol syndrome (FAS). For the remainder of this paper we will refer to the broad category of FASD, rather than the narrow category of people with FAS. FASD broadly impacts the economy and leads to increased spending on health care, residential care, special education and corrections. FASD also impacts the economy through the indirect cost of productivity loss. Despite the absence of cost data from multiple important cost categories, the annual estimates for cost of care for all individuals with FASD ranges from USD $926 million to $3.2 billion [8–15]. These dollar figures are reported in the year of the study and are much higher when inflation adjusted: USD $1.29 billion to $10.1 billion. Annual estimates for

nation-wide costs of FASD for Canada do not include productivity losses from care-givers, but still suggest a massive burden to society, estimated at CAD $1.8–9.7 billion [16–19]. These dollar figures are also higher when inflation-adjusted: CAD $1.9–10.5 billion. Despite the massive economic burden of this preventable disorder, a modest amount of money is spent on research and prevention. In the United States, only 0.19% of the total cost of FASD is used to fund research—a 526-fold difference [13,14]. Similarly, in Canada 0.4% of the total cost of FASD was associated with prevention and research from the largest Canadian agencies—a 250-fold difference [20].

The estimation of life-time costs of FASD is crucial to prioritizing available funding when evaluating prevention programs and developing public policy [9]. The most useful value to consider when dealing with public policy is present discounted value (PDV) [9,21]. PDV refers to the money that needs to be deposited today at a given interest rate so that principal and interest will be dispersed over the person's life-time [22]. A PDV calculation is useful from a public policy perspective, because it converts the estimated life-time cost of an individual with FASD into a easily comparable present dollar amount that would fund life-time costs of care [9,21,22].

The Parent–Child Assistance Program (P-CAP) is an evidence-based prevention program for alcohol- and drug-exposed births that utilizes case management, home visitation and harm reduction mentorship [23]. This program was launched by the University of Washington, and has been replicated at 40 sites in Canada (http://depts.washtingon.edu/pcapuw/). The P-CAP enrolls substance-using (high-risk) women who are pregnant or up to 6 months postpartum. FASD programs have been successful in reducing maternal drinking during pregnancy and the prevalence thereof and have generated substantial social and economic benefits for society [24,25].

Calculating the cost of prevention of FASD is also critical in informing clinical practice and public policy regarding intervention for mothers at risk for giving birth to an individual with FASD. Economic costs of prevention can be compared with the economic savings of prevention to determine the extent that prevention is justified on economic grounds alone. However, there have been few attempts to consider the cost of prevention of FASD alongside the savings from prevention of FASD. Thus, while FASD is a preventable condition that imposes a large economic burden to society, there is a surprising scarcity of literature that analyzes the costs and benefits of prevention efforts for this disorder. These calculations can inform various stakeholders, including policymakers, health-care providers and the public regarding the systemic economic effects of FASD and the financial efficacy of differing intervention strategies for at-risk women.

This study sought to understand if funding of existing interventions in the United States and Canada for primary prevention among women at high risk of having a child with FASD could reduce spending when compared to the cost of care for an individual with FASD. The objectives of this study were to: (i) review existing data on the economic cost of prevention of FASD, (ii) compare the economic cost of prevention with the economic savings of prevention and (iii) estimate the projected savings of prevention efforts accompanied with expansion of existing prevention models.

METHODS

Literature review

Two separate search strategies were utilized in this study: to review (1) existing studies on costs of primary prevention of FASD in the United States and Canada and (2) existing studies on national costs of FASD in the United States and Canada.

The first search strategy for this study, described here, was used to determine the cost of prevention efforts in existing programs. An expert reference librarian assisted in the development of a search strategy. A search of literature published before June 2017, inclusive to all languages, was performed to identify studies reporting the cost of prevention for fetal alcohol spectrum disorder. Keywords were used to search the following electronic bibliographic databases: PubMed, Cochrane Database of Systematic Reviews and Google Scholar. Literature searches were conducted using combinations of the following keywords:

1 Disease conditions: 'fetal alcohol', FASD, FAS, pFAS, fetal alcohol effects (FAE), ARND, ARBD, prenatal alcohol exposure; and
2 Cost: prevention: 'cost of prevention', cost, economic, social cost, economic cost.

A manual review of bibliographies in select studies was performed. Experts in the field were consulted in 2017 to identify studies and data that were not found by our search strategy.

The second search strategy for this study was a recently published systematic literature search that included the national costs of FASD in Canada and the United States [1]. This study was used to determine the country-wide cost of FASD, the approximate percentage of total cost that is dedicated to research and prevention, and the present discounted value in the countries that studied the cost of prevention of FASD.

Eligibility criteria

Studies were considered if they were published in a peer-reviewed journal or scholarly report before June 2017. Studies of all languages were considered. Studies were included if they reported costs associated with a specific

intervention for primary prevention of FASD. Studies were excluded if they did not estimate costs of one case of FASD prevention; reported the cost of screening or secondary prevention but not primary prevention; or contained data which were repeated from an original study.

Qualitative assessment of studies

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2009 checklist was referenced to ensure a standardized approach and minimize error and bias in our literature search and data analysis [26]. Studies that met our inclusion criteria were assessed using the 24-item Consolidated Health Economic Evaluation Reporting Standards (CHEERS checklist) for evaluation of reporting economic evaluations of health interventions [27] (Table 1). The 19-item Consensus on Health Economic Criteria (CHEC) list was then used for the appraisal of methodological quality of economic evaluations [31] (Table 2).

Table 1 The 24-item Consolidated Health Economic Evaluation Reporting Standards (CHEERS) checklist for evaluation of reporting in economic studies.

Reporting quality				
		Astley et al. *2000 [28]*	*Burd* et al. *2003 [29]*	*Thanh* et al. *2015 [30]*
CHEERS checklist item [27]	1			X
	2			X
	3	X	X	X
	4	X	X	X
	5	X	X	X
	6			X
	7	X	X	X
	8	X	X	X
	9			X
	10	X	X	X
	11		X	X
	12	X	X	X
	13		X	X
	14			X
	15		X	X
	16			X
	17			X
	18			X
	19	X	X	X
	20			X
	21		X	X
	22	X	X	X
	23	X	X	X
	24	X	X	X
	Score	11	15	24

Table 2 The 19-item Consensus on Health Economic Criteria (CHEC) list for evaluation of economic study methodology.

Methodological quality				
		Astley et al. *2000 [28]*	*Burd* et al. *2003 [29]*	*Thanh* et al. *2015 [30]*
CHEC-list item [31]	1	X	X	X
	2	X	X	X
	3	X	X	X
	4		X	X
	5	X	X	X
	6	X	X	X
	7	X	X	X
	8	X	X	X
	9	X	X	X
	10		X	X
	11	X	X	X
	12		X	X
	13			X
	14			X
	15			X
	16	X	X	X
	17		X	X
	18	X	X	X
	19	X	X	X
	Score	12	16	19

Data extraction

A standardized spreadsheet (MS-Excel) was used to code each study in both of the search strategies. For cost of prevention studies, the following variables were coded: reference, year(s) of study, year of reported currency value, country that was studied, patient population that was studied and cost to prevent one case of FASD. For studies estimating of cost of care, the following variables were coded: reference, year(s) of study and annual cost for all people with FASD. Data from investigators were confirmed using published peer-reviewed literature or published departmental documents.

Conversion calculations

Dollar figures were extracted from studies in their original values in the year of study. To facilitate accurate and current calculations, the costs of prevention, total annual costs of all people with FASD and life-time costs of individuals with FASD were converted to June 2017 values using inflation calculators. Updated currency values were then used for calculations to determine the economic benefit of prevention. United States costs were converted to June 2017 values using the on-line inflation calculator provided by the US Bureau of Labor Statistics (https://www.bls.gov/data/inflation_calculator.htm). Canadian costs were

converted to 2017 values using the on-line inflation calculator provided by the Bank of Canada (http://www.bankofcanada.ca/rates/related/inflation-calculator/?page_moved=1). Both inflation calculators use currency inflation rates from the Consumer Price Index to derive adjusted values.

Derivation of projected savings

Data were extracted from studies containing information on: (1) cost of prevention per case for FASD in the United States and Canada; (2) annual cost for all individuals with FASD in both United States and Canada; and (3) PDV of lifetime costs for an individual with FASD in the United States and Canada. These values were updated to 2017 currency values using conversion calculators. First, a theoretical funding level of 1% total annual cost of care for all individuals with FASD was determined for both the United States and Canada. National costs of FASD in Canada were based on two studies, and in the United States were based on three studies. These costs were then multiplied by 0.01 [8,9,20,30,32]. These studies provided the high and low ranges in the estimate of the country-wide cost for FASD. To determine how many cases of FASD could be prevented based on 1% funding, 1% of the estimated total annual cost of care was divided by the cost to prevent one case of FASD [28–30]. The number of cases prevented in this theoretical model was then multiplied by the PDV of the cost to society from one case of FASD. This value was used as the cost savings during the life-time of a prevented case. Net savings were then calculated by taking the cost savings during the life-time of individuals with FASD minus the cost of prevention. Dollar figures in tables represent USD in Tables 4 and 5 and CAD in Table 6. Table 3 contains costs in both USD and CAD. Meta-analysis was not performed due to disparities in the quality of studies, studies being conducted in multiple countries with different resource utilization and the paucity of studies available.

RESULTS

Search results

The literature search, focused on cost of prevention of FASD, produced 845 abstracts. Due to significant overlap, there were 522 unique abstracts that were considered further. We found that 424 of these studies did not contain cost information related to FASD. Of the 128 studies that contained cost information related to FASD, only three studies reported data on costs associated with interventions for primary prevention of FASD [28–30] (Fig. 1). While our original literature search was inclusive to all countries, studies that were identified through our inclusive literature review were limited to the United States and Canada. Thus, only data from the United States and Canada were further considered. A recent systematic review provided estimates of national cost of care for individuals with FASD and PDV for the United States and Canada [1]. The lowest and highest estimates from studies reviewed in this recent report were used to estimate projected savings. We identified three studies that estimated national cost of care in the United States that met inclusion criteria. Two studies that estimated national cost of care in Canada were included. Cost of care and PDV estimates from these countries were then used to calculate the savings from prevention.

Critical appraisal of literature

The qualitative assessment of the included studies found the optimal study according to the CHEERS checklist and the CHEC list was by Thanh *et al.* [30] (Tables 1 and 2). This was the only study that performed sensitivity analysis,

Table 3 Systematic review results: estimated cost of prevention of one case of FASD in the United States and Canada.

United States (all costs in USD$)			
Source	*Risk factors or patient population*	*Cost to prevent 1 case (reported in year of study)*	*2017 Equivalent*
Burd *et al.* 2003 [29]	Weekly alcohol use, no binges	$100 000 000	$134 810 000
	Frequent drinkers, non-smokers	$3 450 000	$4 651 000
	Heavy drinkers, low-income, smokers	$235 000	$316 800
	Women who have had a previous child with FAS	$15 000	$20 200
Astley *et al.* 2000 [28]	Women who have had a previous child with FAS	$33 000	$47 615
Canada (all costs in CAD$)			
Thanh *et al.* 2015 [30]	Heavy alcohol consumers	$97 000 (range = $72 000–153000)	$102 914 (range = $76 390–162329)

FAS = fetal alcohol syndrome; FASD = fetal alcohol spectrum disorder.

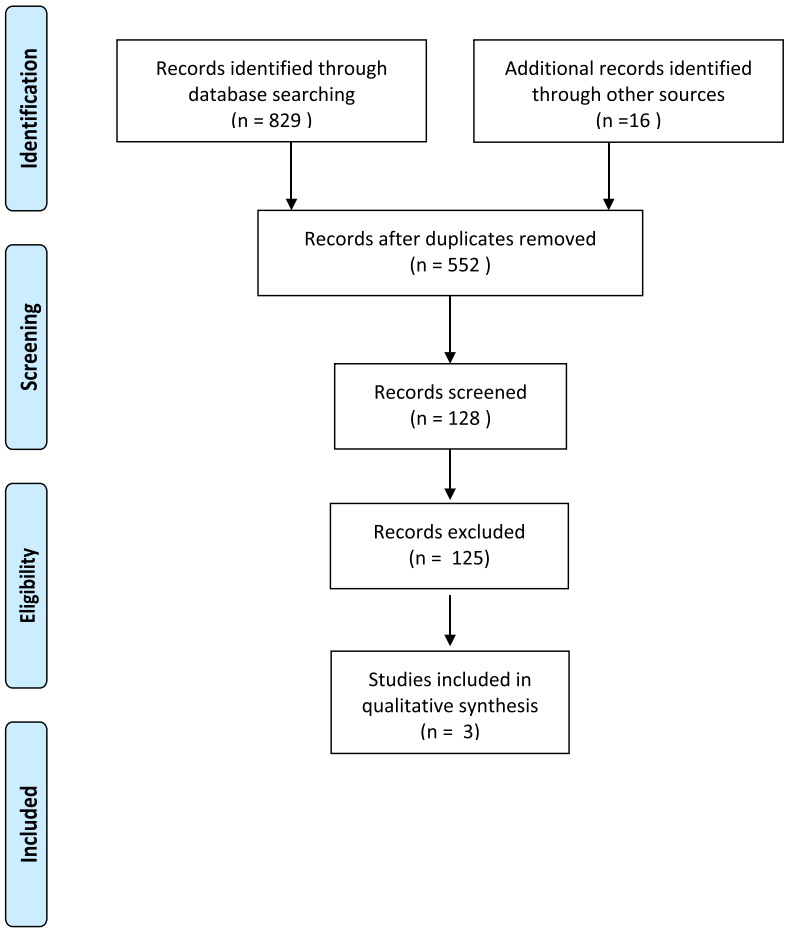

Figure 1 Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) 2009 flow diagram [Colour figure can be viewed at wileyonlinelibrary.com]

incremental analysis and reported methods for discounting future costs. Only one study reported cost estimates for different groups of women [29].

Cost of prevention

Table 3 summarizes the results of the literature review and contains data pertinent to the cost of preventing one case of FASD. Astley *et al*. studied the P-CAP model in Washington State [28]. They reported that by treating women who have had a previous child with FASD (the highest-risk population) the cost to prevent one case of FASD was $47 615 (converted to 2017 USD$). Burd *et al*. conducted a cost analysis in the United States that estimated the cost to prevent FASD based on maternal risk factors [29]. This cost analysis used data from previous studies and modeling data from North Dakota, United States [34]. The cost estimates of preventing one case of FASD varied dramatically based on the risk factors of the woman being treated. The estimated cost per prevented case of FASD was $134 810 000 among women who drink compared to $20 200 for the highest-risk population: a 6674-fold cost reduction. The highest-risk patient populations were women who were heavy drinkers and smokers with low income ($316 800 in 2017 USD$ to prevent one case) and women who have had a previous child with FAS ($20 200 in 2017 USD$). One study examined costs from 366 women who utilized the P-CAP model in Alberta, Canada, from 2008 to 2011 [30]. The decision analytical modeling technique utilized in this study estimated that the cost of preventing one case of FASD by treating women with heavy alcohol consumption in the P-CAP program averaged $99 400 (range = $76 390–162 329) in 2017 CAD$.

Savings from prevention

Annual cost of care estimates for FASD in the United States range from (2017) USD $1.29–10.1 billion [1,8,9]. A theoretical funding for prevention services of 1% the estimated cost of care in the United States would translate to USD $12 900 000–101 000 000. Astley *et al*. reported that the cost to prevent one case of FASD treating the highest-risk women was the equivalent of 2017 USD $47 615 [28]. Thus, using the low and high estimates of cost of care, 1% funding could prevent between 270 and 2121 cases in the P-CAP in Washington State (Table 2). The 2017 adjusted PDV of one case of FASD in the United States is

$1.24 million which means that, for every case prevented, stakeholders in society have saved $1.24 million in spending [22]. Therefore, the net savings of funding a similar prevention program to the P-CAP in Washington State at the level of 1% the national cost of care is predicted to be between $326 220 000 and $2 562 976 000 (Table 4). Burd *et al.* provided a stratified cost analysis that reports estimates for groups of women with different risk factors in North Dakota [29]. It costs approximately $316 800 to prevent one case of FASD among women with low income who are simultaneously heavy drinkers and smokers (2017 USD) [29]. Using the high and low values of a 1% theoretical funding, this correlates to between 40 and 318 prevented cases (Table 5) and to net savings of $37 340 000–298 408 000. By further stratifying maternal risk factors and focusing on women with a previous child with FASD, between 638 and 5000 cases would be prevented (Table 3). This would save the United States between $788 428 000 and $6 179 000 000.

Annual costs of care for FASD in Canada range between $1.9 and 10.5 billion (2017 CAD) [1,20,33]. Funding equivalent to 1% the annual national cost of care would be $19 000 000–105 000 000. Thanh *et al.* reported that the cost to prevent one case of FASD among women with heavy alcohol consumption in the P-CAP program is CAD $102 914 [30]. Using the high and low estimates for 1% funding predicts between 184 and 1020 prevented cases (Table 6). The 2017 adjusted PDV of one case of FASD in Canada is CAD $849 000. Based on the cost of prevention estimates for Alberta, P-CAP funding equivalent to 1% the annual cost of care would provide a net savings of $137 216 000–761 208 679 (Table 4).

Additional savings

Expert consultation highlighted that economic benefits to the P-CAP model are not limited to prevention of FASD. Other sources of significant cost savings in this model were

Table 4 Projected savings of Washington State, US P-CAP with funding equivalent to 1% of annual national costs of FASD (derived from Astley *et al.* 2000) [28] (all costs in 2017 USD$).

	Cases prevented based on funding of prevention programs at 1% the annual cost of FASD in the US (1% of funding/cost to prevent 1 case)		*Range of cost savings over life-time of individuals (number of cases prevented × PDV)*[c]	*Range of net savings (cost savings minus cost of funding)*
Maternal risk factors	*Low estimate*[a]	*High estimate*[b]		
Women who have had a previous child with FAS	270	2121	$339 120 000–2 663 976 000	$326 220 000–2 562 976 000

[a]$1 240 000 PDV based on Lupton *et al.* 2004 [22]. [b]$101 000 000 based on 1% of estimated national cost of care in Harwood *et al.* 1984 [8] and Harwood & Napolitano, 1985 [9]. [c]$12 900 000 based on 1% of estimated national cost of care in Miller, 2006 [32]. FAS = fetal alcohol syndrome; FASD = fetal alcohol spectrum disorder); P-CAP = Parent–Child Assistance Program; PDV = present discounted value.

Table 5 Projected stratified savings in the United States from a prevention program with funding equivalent to 1% of annual national cost of FASD (derived from Burd *et al.* 2003) [29] (all costs in 2017 USD$).

	Cases prevented based on funding of prevention programs at 1% the annual cost of FASD in the US (1% of funding/cost to prevent 1 case)		*Range of cost savings over life-time of individuals (number of cases prevented × PDV)*[c]	*Range of net savings (cost savings minus cost of funding)*
Maternal risk factors	*Low estimate*[a]	*High estimate*[b]		
Weekly alcohol use, no binges	0	0	$0	$0
Frequent drinkers, non-smokers	2	12	$2 512 000–26 376 000	$0
Heavy drinkers, low-income, smokers	40	318	$50 240 000–399 408 000	$37 340 000–298 408 000
Women who had a previous child with FAS	638	5000	$801 328 000–6 280 000 000	$788 428 000–6 179 000 000

[a]$1 240 000 PDV based on Lupton *et al.* 2004 [22]. [b]$101 000 000 based on 1% of estimated national cost of care in Harwood *et al.* 1984 [8] and Harwood & Napolitano, 1985 [9]. [c]$12 900 000 based on 1% of estimated national cost of care in Miller, 2006 [32]. FAS = fetal alcohol syndrome; FASD = fetal alcohol spectrum disorder); PDV = present discounted value.

Table 6 Projected savings of Alberta, Canada P-CAP with funding equivalent to 1% of annual national costs of FASD (derived from Thanh *et al.* 2015) [30] (all costs in 2017 CAD$).

Population studied	*Cases prevented based on funding of prevention programs at 1% the annual cost of FASD in the US (1% of funding/cost to prevent 1 case)*		*Range of cost savings over life-time of individuals (number of cases prevented × PDV)*[c]	*Range of net savings (cost savings minus cost of funding)*
	Low estimate[a]	*High estimate*[b]		
Alcohol users who were served by the Alberta FASD-Cross Ministry Committee P-CAP	184	1020	$156 216 000–866 208 679	$137 216 000–761 208 679

[a]$849 000 PDV based on Thanh *et al.* 2011 [33], which modified Harwood & Napolitano, 1985 [9]. [b]$105 000 000 based on 1% of estimated national cost of care in Thanh and Jonsson, 2015 [18]. [c]$19 000 000 based on 1% of estimated national cost of care in Popova *et al.* 2016 [20]. FASD = fetal alcohol spectrum disorder); P-CAP = Parent–Child Assistance Program; PDV = present discounted value.

decreasing time to re-unification with children; reduced dependence on public assistance; increased employment; and increased levels of education [25].

DISCUSSION

Adjusted annual cost estimates for all individuals with FASD in the United States range from $1.29 billion to $10.1 billion and in Canada from $1.9 billion to $10.5 billion [1]. An intervention program in the United States that focused on women who have previously given birth to a child with FASD could cost as little as $20 200 USD per case prevented, and would save society USD $1 235 800 (net) based on the present discounted value of life-time cost [22]. This is a 62-fold cost reduction for women with a high recurrence risk. Prevention efforts that treated women of low socio-economic status who are heavy drinkers and smokers ($316 800/case prevented) would save society USD $939 200 per case prevented. Similar findings have been reported in separate locations inside the United States and in Canada in the P-CAP model of prevention [28,30]. One study noted that the cost of raising a child with FAS would be approximately 30 times the cost of preventing one case of FASD [28]. Another study concluded that funding of cost-effective FASD prevention can provide greater than 700% returns on the investment [30]. These authors also noted that these reported savings estimates are conservative, as they do not include savings associated with other benefits of P-CAP [25,30]. If 1% of estimated 2017 costs associated with care of people with FASD in the United States or Canada was allocated toward prevention in high risk women, the economic benefit during the course of the individuals' life-times would be extraordinary. These data demonstrate the economic acuity of utilizing prevention models that utilize maternal risk stratification.

Future research on cost of prevention would be helpful. Despite the significant cost of care for FASD, estimates based on data from both Canada and United States agencies suggest that prevention and research accounts for less than 0.5% of total FASD spending [13,14,20]. There have been relatively few attempts to calculate the cost of prevention for FASD—despite FASD being a preventable disease with a large economic cost to society. More research is needed to investigate the cost of prevention of FASD. The existing studies that investigate cost of FASD prevention demonstrate that current prevention models provide a large return on investment for society. Additional high-quality studies are needed to validate existing data and direct public health initiatives.

To establish a cost-effective model of prevention, a stratified approach based on cost of prevention per case would be optimal. Funding for prevention should focus on women that present the highest risk of giving birth to a child with FASD. Cost-effective prevention strategies should utilize targeted intervention that prioritizes those with the highest risk: (1) women who have had a previous child with FASD; and (2) women of low socio-economic status who are heavy drinkers and smokers and have poor diets [29]. In addition to direct and indirect economic incentives, the intangible costs (suffering) that could be alleviated by thoughtful and cost-effective intervention should encourage policymakers to invest in prevention of FASD based on risk assessment.

LIMITATIONS

Based on the scarcity of existing literature, there is a risk of bias in the studies reviewed in this report. Additional limitations to this study include the inability to exactly predict the cost of care for individuals with FASD and the cost of prevention for FASD. These limitations can be separated into those that would increase the savings of FASD prevention and those that would decrease the savings of prevention. First, our estimated savings are probably underestimated, because most cost studies that examine the life-time and nation-wide costs of FASD do not include data from every cost category [1,20]. By contrast, there are

limitations that may lead to overestimation of savings. The existing studies that examine the cost of prevention have studied programs with existing infrastructure, and if funding were to increase for FASD prevention the cost of prevention may transiently rise, as there would be an increased cost associated with training health-care professionals. Next, when our model predicts the cases prevented based on 1% funding, it assumes that an infinite number of new FASD cases are born to mothers with certain identifiable risk factors.

Declaration of interests

None.

Acknowledgements

No external funding was utilized for this research.

References

1. Greenmyer J. R., Klug M. G., Kambeitz C., Popova S., Burd L. A multicountry updated assessment of the economic impact of fetal alcohol spectrum disorder: costs for children and adults. *J Addict Med* 2018; **12**: 466–73.
2. Popova S., Stade B., Bekmuradov D., Lange S., Rehm J. What do we know about the economic impact of fetal alcohol spectrum disorder? A systematic literature review. *Alcohol Alcohol* 2011; **46**: 490–7.
3. Amendah D. D., Grosse S. D., Bertrand J. Medical expenditures of children in the United States with fetal alcohol syndrome. *Neurotoxicol Teratol* 2011; **33**: 322–4.
4. Stratton K. R., Howe C. J., Battaglia F. C., Institute of Medicine, Committee to Study Fetal Alcohol Syndrome *Fetal alcohol syndrome—diagnosis, epidemiology, prevention, and treatment*. Washington, DC: National Academy Press; 1996.
5. Popova S., Lange S., Shield K., Mihic A., Chudley A. E., Mukherjee R. A., *et al*. Comorbidity of fetal alcohol spectrum disorder: a systematic review and meta-analysis. *Lancet* 2016; **387**: 978–87.
6. Burd L. Invited commentary: Fetal alcohol spectrum disorder: complexity from comorbidity. *Lancet* 2016; **387**: 926–7.
7. Streissguth AP, Barr HM, Koga J, Bookstein FL. Understanding the Occurrence of Secondary Disabilities in Clients with FAS and FAE. Seattle: University of Washington Fetal Alcohol and Drug Unit; 1996.
8. Harwood H. J., Napolitano D. M., Kristiansen P. L., Collins J. J. *Economic Costs to Society of Alcohol and Drug Abuse and Mental Illness: 1980*. Research Triangle Institute: Research Triangle Park, NC; 1984.
9. Harwood H. J., Napolitano D. M. Economic implications of the fetal alcohol syndrome. *Alcohol Health Res World* 1985; **10**: 38–75.
10. Abel E. L., Sokol R. J. Incidence of fetal alcohol syndrome and economic impact of FAS-related anomalies. *Drug Alcohol Depend* 1987; **19**: 51–70.
11. Abel E. L., Sokol R. J. A revised conservative estimate of the incidence of FAS and its economic impact. *Alcohol Clin Exp Res* 1991; **15**: 514–24.
12. Abel E. L., Sokol R. J. A revised estimate of the economic impact of fetal alcohol syndrome. In: Glanter J., editor. *Recent Developments in Alcoholism: Children of Alcoholics, 9th edn. New York: Plenum Press; 1991, pp. 117–25.*
13. Rice D, Kelman S, Miller L. The Economic Costs of Alcohol and Drug Abuse and Mental Illness. 1985. DHHS Publication no. (ADM) 90–1694. Rockville, MD: US Department of Health and Human Services; 1990.
14. Rice D. P., Kelman S., Miller L. S. Estimates of economic costs of alcohol and drug abuse and mental illness, 1985 and 1988. *Publ Health Rep* 1991; **106**: 280–92.
15. Bouchery E. E., Harwood H. J., Sacks J. J., Simon C. J., Brewer R. D. Economic costs of excessive alcohol consumption in the U.S., 2006. *Am J Prev Med* 2011; **41**: 516–24.
16. Stade B., Ali A., Bennett D., Campbell D., Johnston M., Lens C., *et al*. The burden of prenatal exposure to alcohol: revised measurement of cost. *Can J Clin Pharmacol* 2009; **16**: 91–102.
17. Thanh N. X., Jonsson E. Drinking alcohol during pregnancy: evidence from Canadian Community Health Survey 2007/2008. *J Popul Ther Clin Pharmacol* 2010; **17**: 302.
18. Thanh N. X., Jonsson E. Costs of fetal alcohol spectrum disorder in the Canadian criminal justice system. *J Popul Ther Clin Pharmacol* 2015; **22**: e125–e131.
19. Popova S., Lange S., Burd L., Rehm J. Cost attributable to fetal alcohol spectrum disorder in the Canadian correctional system. *Int J Law Psychiatry* 2015; **41**: 76–81.
20. Popova S., Lange S., Burd L., Rehm J. The economic burden of fetal alcohol spectrum disorder in Canada in 2013. *Alcohol Alcohol* 2016; **51**: 367–75.
21. Bloss G. The economic cost of FAS. *Alcohol Health Res World* 1994; **18**: 53–4.
22. Lupton C., Burd L., Harwood R. Cost of fetal alcohol spectrum disorders. *Am J Med Genet C Semin Med Genet* 2004; **127C**: 42–50.
23. Ernst C. C., Grant T. M., Streissguth A. P., Sampson P. D. Intervention with high-risk alcohol and drug-abusing mothers: II. Three-year findings from the Seattle model of paraprofessional advocacy. *J Community Psychol* 1999; **27**: 19–38.
24. Astley S. J. Fetal alcohol syndrome prevention in Washington state: evidence of success. *Paediatr Perinat Epidemiol* 2004; **18**: 344–51.
25. Grant T., Casey Family Programs. Parent–Child Assistance Program Outcomes Suggest Sources of Cost Savings for Washington State. Washington, DC: University of Washington; 2013.
26. Moher D., Liberati A., Tetzlaff J., Altman D. G. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *PLOS Med* 2009; **6**: e1000097.
27. Husereau D., Drummond M., Petrou S., Carswell C., Moher D., Greenberg D., *et al*. Consolidated health economic evaluation reporting standards (CHEERS)—explanation and elaboration: a report of the ISPOR health economic evaluation publication guidelines good reporting practices task force. *Value Health* 2013; **16**: 231–50.
28. Astley S. J., Bailey D., Talbot C., Clarren S. K. Fetal alcohol syndrome (FAS) primary prevention through FAS diagnosis: I. Identification of high-risk birth mothers through the diagnosis of their children. *Alcohol Alcohol* 2000; **35**: 499–508.
29. Burd L., Cotsonas-Hassler T. M., Martsolf J., Kerbeshian J. Recognition and management of fetal alcohol syndrome. *Neurotoxicol Teratol* 2003; **25**: 681–8.
30. Thanh N., Jonsson E., Moffat J., Dennett L., Chuck A., Birchard S. An economic evaluation of the parent-child assistance program for preventing fetal alcohol spectrum disorder in Alberta. *Canada. Adm Policy Ment Health* 2015; **42**: 10–8.

31. Evers S., Goossens M., de Vet H., van Tulder M., Ament A. Criteria list for assessment of methodological quality of economic evaluations: consensus on health economic criteria. *Int J Technol Assess Health Care* 2005; **21**: 240–5.
32. Miller T. R., Levy D. T., Spicer R. S., Taylor D. M. Societal costs of underage drinking. *J Stud Alcohol* 2006; **67**: 519–28.
33. Thanh N. X., Jonsson E., Dennett L., Jacobs P. Costs of FASD. In: Riley E. P., Clarren S., Weinberg J., Jonsson E., editors. *Fetal Alcohol Spectrum Disorder: Management and Policy Perspectives of FASD*. Weinheim, Germany: Wiley-Blackwell; 2011, pp. 45–69.
34. Burd L., Wentz T. Fetal alcohol syndrome: the North Dakota experience. *North Dakota J Human Serv* 1997; **1**: 14–9.