

# THE BURDEN OF PRENATAL EXPOSURE TO ALCOHOL: MEASUREMENT OF COST

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## ABSTRACT

### Background

In Canada the incidence of Fetal Alcohol Spectrum Disorder (FASD) is estimated at 1 to 6 in 1000 live births. FASD is the leading cause of developmental and cognitive disabilities among Canadian children. There is a paucity of research examining the economic costs of FASD.

### Objective

To estimate direct and indirect costs associated with FASD at the patient level.

### Methods

**Design:** Cross-sectional study design was used. **Sample and Setting:** One-hundred and forty-eight (148) parents of children with FASD, aged 1 to 21 years, living in urban and rural communities throughout Canada. **Procedure:** Participants completed the Health Services Utilization Inventory (HSUI). Key cost components were elicited: direct costs: medical, education, social services, out-of-pocket costs; and indirect costs: productivity losses. **Data Analysis:** Total average costs per child were calculated by summing the costs for each child in each cost component, and dividing by the sample size. Costs were extrapolated to one year. A stepwise multiple regression analysis was used to identify significant determinants of costs and to calculate the adjusted annual costs associated with FASD.

### Results

Total adjusted annual costs associated with FASD per child were \$14,342 (95% CI, \$12,986; \$15,698). Severity of the child's condition, age of the child, and geographical setting were significant determinants of costs ( $p < 0.001$ ). Cost of FASD annually to Canada of those 1 to 21 years old, was \$344,208,000 (95% CI \$311,664,000; \$376,752,000).

### Conclusions

Study results demonstrated the cost burden of FASD was profound. Implications for practice, policy, and research are discussed.

**Key Words:** alcohol, pregnancy, cost, economic burden, fetal alcohol syndrome, fetal alcohol effects, fetal alcohol spectrum disorder

In Canada the incidence of Fetal Alcohol Spectrum Disorder (FASD) has been estimated to be 1 to 6 in 1000 live births.<sup>1,2,3</sup> Caused by prenatal exposure to alcohol, the disorder is the leading cause of developmental and cognitive disabilities among Canadian children and its effects are life lasting.<sup>1-3</sup> Fetal Alcohol Spectrum Disorder acknowledges that Fetal Alcohol Syndrome is a continuum, with differing

degrees of expression of dysfunction and malformation. The full Fetal Alcohol Syndrome (FAS) is characterized by a triad of signs:

- 1) prenatal and or postnatal growth retardation;
- 2) characteristic facial anomalies including short palpebral fissures, flat philtrum, and thin vermilion border of the upper lip; and
- 3) central nervous system dysfunction demonstrated by intellectual impairment and/or

structural abnormalities, microcephaly, developmental delay, and a complex behaviours problems.

Children with FAS often display characteristics such as extreme hyperactivity, aggressiveness, poor judgment, speech and language difficulties. Other clinical manifestations of FAS may include cardiac anomalies, urogenital defects, skeletal abnormalities, visual and hearing problems.<sup>1,2,3,4,5,6,7,8,9,10</sup> The term "Fetal Alcohol Effects" (FAE) and more recent diagnostic terms such as "Partial FAS" (PFAS), "Alcohol-Related Neurodevelopmental Disorder" (ARND) and "Neurobehavioral Disorder - Alcohol Exposed" are used to describe cases of lesser severity in terms of cognitive function and organ anomalies, but often with very serious evidence of neurotoxicity problems.<sup>1,2,3,10</sup>

While programs to prevent and treat Fetal Alcohol Syndrome have been identified throughout Canada, these initiatives have lacked consistency and coordination. Comprehensive programs for the prevention and treatment of FASD must be developed. This research examined the necessity of allocating funds to FASD prevention and treatment programs by measuring the economic costs of FASD to society and to its victims. The research objectives were to determine the average annual direct and indirect costs per case from the societal perspective and to identify the significant determinants of costs. The current research addresses a gap in knowledge as analyses of costs existing to date have calculated only partial costs of the syndrome, reflecting gross underestimates of the true cost of FASD, and none has been conducted in a Canadian setting.

### **Review of the Relevant Literature**

The economic impact of Fetal Alcohol Syndrome was measured in four American-based studies.<sup>11,12,13</sup> Abel & Sokol measured the economic cost of FAS from the perspective of the health care system and estimated that the burden of FAS in the United States was US \$321 million in 1984, based on an average incidence of 1.9 FAS cases per 1000 live births.<sup>11</sup> The incidence rate was an average, drawn from several prospective and retrospective studies.

Components of costs included treatment of care of low birth weight babies with FAS; costs for surgical correction of FAS-related birth

defects; care for those with moderate or severe cognitive disabilities; and the cost of semi-independent supervised support for mildly cognitively disabled patients with FAS who were 21 years of age and under.

In 1991, Abel & Sokol again estimated the cost of FAS from the perspective of the health care system and produced a much lower annual cost estimate of US \$74.6 million, based on an incidence rate of 0.33 FAS cases per 1000 live births.<sup>12</sup> This conservative estimate was derived entirely from prospective studies, which yield lower estimates of FAS incidence than do retrospective studies, in part because, unlike the retrospective studies, there are no prospective data for Native Americans and other racial/ethnic groups that may face risks of FAS. In addition, the cost of semi-independent supervised support for mildly cognitively disabled patients ages 21 and under was excluded from the later study, on the grounds that such care was generally required only after age 21. Neither of the Abel and Sokol studies included costs beyond age 21.

Harwood & Napolitano used a societal perspective and generated cost estimates of US \$ 1.95, 3.2, and 9.69 billion dollars using alternative FAS incidence rates of 1.0; 1.67; and 5.0 per 1000 live births in the United States.<sup>13</sup> Incidence rates were based on a review of prospective studies. Costs included estimates of the value of productivity lost as a result of cognitive disabilities, as well as the cost of treatment and residential care for patients of all ages with FAS.

Finally, Rice, Kelman et al. estimated the cost of FAS from the perspective of the health care system and placed the annual cost of treating the birth defects associated with FAS in the United States at US \$1.6 billion, based on an incidence of 1.9 FAS cases per 1000 live births.<sup>14</sup> The incidence was based on a review of several prospective and retrospective studies. Components of costs included the cost of care for FAS-related birth defects and cognitive disability, as well as the cost of residential care for patients over 21 years. The cost of residential care accounted for 80 percent of the total cost estimate.

The four studies reviewed demonstrate that the physical, behavioural, and cognitive sequelae of FAS and FAE are costly to treat and to rehabilitate, and may limit an individual's ability to contribute to society's productivity. The

economic burden of FAS calculated in these studies provides impetus for implementing prevention and treatment strategies. However, there are several limitations to past research examining the costs of FAS.

In past research estimates of costs are strictly limited to FAS and do not reflect costs of FAE or other alcohol related effects, which are more common. Thus, the results of these studies may significantly underestimate the economic burden associated with prenatal exposure to alcohol. In addition, all of the past studies are USA-based and thus may yield lower estimates of FAS incidence than found in Canada. For example, FAS is more prevalent among Native People, both in Canada and in the US. In some Native communities, the estimated prevalence of FAS is as high as 25 to 190 per 1000 children.<sup>15</sup> However, Native Canadians represent a higher percentage of the total population than Native Americans.<sup>16,17</sup> The overall incidence of FAS in Canada has been estimated to be 1 cases per 100 live births, which is significantly higher than incidence rates cited in American studies.<sup>2</sup> In addition, precisely what costs should be measured and included in an economic evaluation depends on the perspective selected. In 3 of the 4 studies reviewed, the perspective chosen resulted in under-estimation of the total costs of FAS. Specifically, Abel & Sokol and Rice, Kelman et al. failed to include direct costs to the family/patient or productivity losses associated with prenatal exposure to alcohol.<sup>11,12,13,14</sup> Discrepancies in cost estimates obtained in past studies were due, in part, to the time horizon used in the evaluation. For example, Abel & Sokol did not include costs beyond 21 years of age.<sup>11,12</sup> Rice, Kelman et al., included the costs of treatment and care for individuals under the age of 22 years as well as residential care for all ages.<sup>14</sup> Harwood & Napolitano, with the largest cost estimates, included treatment, care and lost productivity for all ages in his research.<sup>13</sup> No research has examined the cost of crime related to Alcohol Related Birth Defects. The severe behavioral problems associated with prenatal exposure to alcohol put adults with FAS at higher risk for criminal behavior. Streissguth et al. found that 6.2% of adolescents and adults with FAS exhibit serious levels of maladaptive behaviors.<sup>18,19</sup>

## METHODS

### Research Design

Prospective cross-sectional research design was used.

### Setting

This study was conducted in urban and rural settings throughout Canada.

### Sample

A cross-sectional research design was used. Potential participants were elicited from FASworld Canada, a national parent support agency representing over 11 parent support groups. This agency provides service to a heterogeneous population of over 700 children with FASD and their parents throughout Canada. All participants in this study were diagnosed with either FAS or FAE. The newer diagnostic terms such as ARND, PFAS and others were not assigned to the children in this study. Thus, the study will specifically refer to the terms “FAS” or “FAE”, acknowledging that these 2 terms are diagnostic categories within the continuum of the broader category “Fetal Alcohol Spectrum Disorder”. The children with FAS and FAE vary in the following characteristics: age, gender, educational levels and abilities, ethnicity, and age of entry into their current home. Their parents also vary in terms of age, gender, marital status, education and relationship to the child (adoptive, biological, foster).<sup>20</sup>

The study sample included parents (biological, adoptive, or foster) of one or more children diagnosed with FAS or FAE, aged 1 to 21 years, who were currently, living with the child who has FASD, or responsible for the care and welfare of that child. The study was approved by the joint university/ institutional Research Ethics Board and all participants provided written informed consent.

### Data Collection: Health Services Utilization Inventory (HSUI)

The Health Services Utilization Inventory (HSUI) was modified to collect data and measure the quantity of different types of health services used by parents of children with FAS/FAE.<sup>21,22,23</sup> It consisted of questions about the respondents' direct costs including costs of medical care - hospital admissions, health professional services, medication; costs of educational services - home

schooling, special schooling; costs of social services – respite care, foster care; and direct costs to parents – parking and transportation costs and costs of externalizing behaviors.

The tool also included questions to assess indirect costs such as days missed from work caring for the child with FAS. Questions were restricted to a reliable duration of recall, such as 6 months for remembering a hospitalization, 3 months for a visit to a physician and 1 month for the use of a prescription medication.<sup>22</sup>

The tool contained questions consistent with the original tool but written within categories that were relevant to service utilization in children with FAS: growth problems, birth defects, cognitive delay, and behavioural problems. The modified tool was reviewed by experts in FASD, including parents and professionals throughout Canada for content validity.

Unit prices for services and care identified by the parents were collected separately, and total expenditures for services used were calculated. Unit costs associated with costs of treatment and care identified by the parents in the “Health Inventory” were collected from various sources including Pediatric Hospitals; Ministry of Education; Ontario Ministry of Community and Social Services; Ontario Schedule of Physician Benefits; Ontario Drug Formulary; and others.

Productivity losses were calculated by estimating loss of caregivers’ wages. Finally, costs that are incurred because of the child’s externalizing behaviours, which include acts of aggression such as damage to people/property or stealing, were included in the total estimates of costs whenever possible. Costs were analyzed from the perspective of society, the provincial ministries of health and the patient. All costs incurred by parents and caregivers were assigned to the child as the unit of analysis. An incidence approach which begins at birth was used. All data were entered using Microsoft ACCESS, and analyzed using Statistical Package for the Social Sciences (SPSS).

### Data Analysis

The average direct costs associated with FAS at the individual level, from birth to age 21 years, were calculated by multiplying the volume of resource use by the unit price for each service. Costs for recall intervals of various lengths were

annualized by linear extrapolation. All costs were expressed in 2003 Canadian dollars.

Employed subjects were asked to report their annual salary within a \$10,000 range, and the median was used in the calculation. Wages per day were estimated by dividing the annual salary by the number of potentially productive days per year (240). This is estimated by subtracting vacation time (10 work days) and legal holidays (10 work days) from the number of work days per year (260), resulting in 240 potentially productive days per year.<sup>24</sup> For unpaid laborers or those not reporting a wage, a wage was imputed from sex-specific and age-specific means of the study sample.<sup>24,25</sup>

Productivity costs were measured using the human capital approach. The Health Services Utilization Inventory elicited information about the number of days lost from work, over the preceding month, caring for the child with FAS or FAE. The number of days lost were multiplied by 12 to calculate time losses over a 12 month period. This number was then multiplied by the parent’s average daily wage yielding annual productivity losses per child with FAS or FAE.

Expenses related to the child’s externalizing behaviors, including acts of violence against persons, animals, and or property; and stealing were elicited directly in the inventory. The contributions of key cost components were examined. Costs were categorized as:

- 1) medical;
- 2) education;
- 3) social services;
- 4) patient/family direct;
- 5) productivity losses; and
- 6) externalizing behaviours.

Total unadjusted costs were calculated at the patient level by summing the costs for each child in each cost component. Societal costs were presented from the perspective of society, the Ministry of Health and the patient.

A stepwise multiple regression analysis was used to calculate the average cost per case, controlling for explanatory variables. Variables which could potentially influence cost, included severity of illness defined by degree of cognitive delay and behavioural problems, age of the child at diagnosis, relationship to the child (biological

vs. adoptive), age of the parent, marital status, occupation of the parent, annual wage of parent, ethnic group of child and parent, and geographical setting. P values of <0.05 were considered significant.

**TABLE 1** Cost Study: Characteristics of the Children

<b>Characteristics</b>		<b>Number</b>	<b>(%)</b>
Sex:	Male	84	56.8%
	Female	64	43.2%
Diagnosis:	FAS	80	54.1%
	FAE	68	45.9%
Cultural Group:	Native	67	45.3%
	Euro-Canadian	81	54.7%
Age:	1-7 years	22	14.9%
	8-12 years	48	32.4%
	13-17 years	40	27.0%
	18-21 years	38	25.7%
	Mean Age = 12.9 years		
Relationship to Parent:	Biological	17	11.5%
	Adoptive	82	55.4%
	Foster	49	33.1%

**TABLE 2** Cost Study: Characteristics of the Parents (n=148)

<b>Gender</b>	
Female	105 (71 %)
Male	43 (29 %)
<b>Age (in years)</b>	
< 20 to 30	6 ( 4 %)
31 to 40	34 (23 %)
41 to 50	46 (31 %)
51 to 60	38 (26 %)
> 60	24 (16 %)
<b>Relationship to the child with FAS/FAE</b>	
Biological	17 (12 %)
Adoptive	82 (55 %)
Foster	49 (33 %)
<b>Marital Status</b>	
Married/Common-in-law	72 (49 %)
Single	31 (21 %)
Divorced/Separated	45 (30 %)
<b>Occupation</b>	
Employed Full-Time	64 (43 %)
Part-Time	34 (23 %)
Unemployed	3 ( 2 %)
Full-Time Homemaker	20 (14 %)
Pension	9 ( 6 %)
Self-Employed	18 (12 %)
<b>Average annual earned salary</b>	
\$10,000 to \$30,000	23 (15 %)
\$30,000 to \$40,000	36 (24 %)
\$40,000 to \$50,000	38 (26 %)
\$50,000 to \$60,000	34 (23 %)
more than \$60,000	16 (11 %)
Preferred not to answer	1 ( 1 %)
<b>Education</b>	
Grade 7 to completion of Grade 12	37 (25 %)
Grade 13 (if applicable and/or Some University or College)	49 (33 %)
Completed University or College	60 (41 %)
Post Graduate Work	2 ( 1 %)
<b>Ethnic Group</b>	
Native	67 (45 %)
Euro-Canadian	81 (55 %)

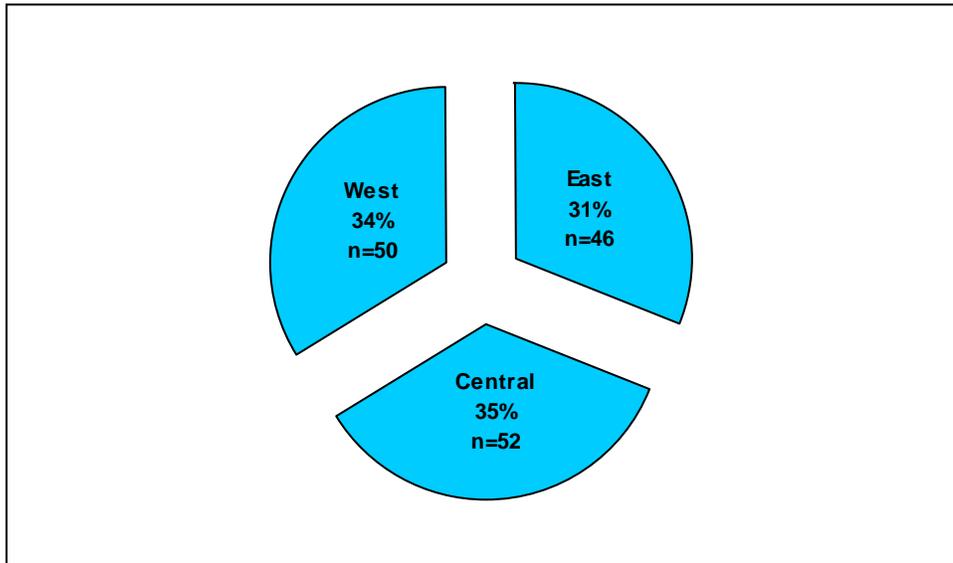
## RESULTS

### Sample Characteristics

One-hundred and forty-eight parents participated in the study. Table 1 presents the children's characteristics; Table 2 presents the

parents' characteristics. Figure 1 demonstrates the number and percentage of participants from the 3 geographical settings: Canada West, Central and East.

**FIG. 1** Cost: Participants by Geographical Region\*



\*West: Saskatchewan, Alberta, British Columbia and the Yukon/North West Territories  
Central: Ontario and Manitoba  
East: Newfoundland, New Brunswick, Nova Scotia and Quebec

### Direct and Indirect Costs

The contributions of the direct and indirect cost components from the perspectives of society, the Ministry of Health, and the patient are displayed in Table 3. From the societal perspective, the average unadjusted annual cost was \$13,109 per child with FAS/FAE.

### Components of Direct Costs

Components from the societal perspective included medical, education, social services and out-of-pocket costs. As illustrated in Table 3, education (32.6 %) and medical services (30.3 %) represented the greatest percentage of costs. It is striking that families pay 19 % of the total costs.

**TABLE 3** Average Annual Cost of FAS/FAE per Case

Component	Societal		Ministry of Health/Social Service		Patient	
	Cost (\$)	% of total	Cost (\$)	% of total	Cost (\$)	% of total
<b>Direct Costs: Medical</b>	<b>\$15,975.80 (Neonatal)</b>		<b>\$15,975.80 (Neonatal)</b>			
Hospitalization	<b>\$160.00</b>		<b>\$ 160.00</b>		N/A	
Emergency Room/Clinic Visits	<b>\$ 53.00</b>		<b>\$ 53.00</b>		N/A	
<b>Total</b>	<b>\$213.00</b>		<b>\$213.00</b>			
<u>Visits to Health Professionals</u>						
Family Doctor	\$167.72		\$167.72		N/A	
Orthopedic Surgery	\$ 76.80		\$ 76.80			
Urologist	\$ 55.95		\$ 55.95			
Allergist	\$ 5.71		\$ 5.71			
Pediatrician	\$ 97.93		\$ 97.93			
Psychiatrist	\$858.65		\$858.65			
Occupational Therapist	\$115.88		\$115.88			
Physiotherapist	\$115.80		\$115.80			
Speech Therapist	\$ 7.00		\$ 7.00			
Psychologist	\$925.00		\$925.00			
Medical Devices	\$550.00		\$470.00		\$ 42.50	
Medication Dispensing Fees	\$ 41.94		\$ 25.16		\$ 4.20	
Prescription Medications	\$449.79		\$245.80		\$166.80	
Non- Prescription Medication	\$249.60		N/A		\$249.60	
Diagnostic Tests	\$ 45.72		\$ 45.72		N/A	
<b>TOTAL</b>	<b>\$3,975.88</b>	<b>30.3%</b>	<b>\$3,425.51</b>	<b>32.4%</b>	<b>\$463.10</b>	<b>18.9 %</b>
<b>Direct Costs: Education</b>						
Home Schooling	\$ 27.30		\$ 27.30		N/A	
Special Schooling	\$1,560.00		\$1,560.00		N/A	
Residential Program	\$2,688.00		\$2,688.00		N/A	
<b>TOTAL</b>	<b>\$4,275.30</b>	<b>32.6%</b>	<b>\$4,275.30</b>	<b>40.5%</b>		
<b>Direct Costs: Social Services</b>						
Respite Care	\$ 144.00		\$ 144.00		N/A	
Foster Care	\$2,122.03		\$2,122.03		N/A	
Legal Aid	\$ 600.00		\$ 600.00		N/A	
<b>TOTAL</b>	<b>\$2,866.03</b>	<b>21.9%</b>	<b>\$2,866.03</b>	<b>27.1%</b>		
<b>Out-of-Pocket</b>						
Transportation per visit	\$ 40.86		N/A		\$ 40.86	
Parking	\$ 55.25		N/A		\$ 55.25	
Externalizing Behaviours	\$840.00				\$840.00	
<b>TOTAL</b>	<b>\$936.11</b>	<b>7.1%</b>	N/A		<b>\$936.11</b>	<b>38.1 %</b>
<b>TOTAL DIRECT COSTS</b>	<b>\$12,053.32</b>	<b>91.9%</b>				
<b>Indirect Costs: Productivity Losses</b>	<b>\$1055.25</b>	<b>8.1%</b>				
<b>TOTAL COSTS</b>	<b>\$13,108.57</b>		<b>\$10,566.84</b>		<b>\$2,454.46</b>	

**Determinants of Cost**

Table 4 illustrates that severity of the child’s condition, age of the child and geographical setting significantly impacted on costs associated with FASD. Table 5 presents the adjusted annual costs for severity of disability. As expected, cost increased with severity of illness, Figure 2 demonstrates the various resources consumed as the child with FASD goes through life. The youngest age is characterized by utilization of health care, while the older children reflect educational needs and externalizing behaviours. The average annual costs appear to peak in early

adolescence. Table 6 presents the adjusted annual costs by geographical region. Costs are higher the west and decrease as one move to the east coast. The regressions analysis was used to arrive at an adjusted average total annual cost for individuals diagnosed with FAS or FAE, ages 1 to 21 years, in Canada. Specifically, when adjusted for severity of disability, age, and geographical region, the summary adjusted value of average annual total costs was \$14,342.00 (95% CI, \$12,986; \$15,698).

**TABLE 4** Determinants of Costs from Societal Perspective

Variable	F	p
Severity of Disability	45.36	<0.001
Age of Child	25.85	<0.001
Geographical Setting	13.78	<0.01
Relationship to Child	5.84	NS*
Age of Parent	3.82	NS*
Occupation of Parent	3.22	NS*
Education of Parent	1.78	NS*
Cultural Group of Child	1.52	NS*
Marital Status	1.06	NS*

\* NS=Not significant

**TABLE 5** Adjusted Annual Costs per Child According to Severity of Disability

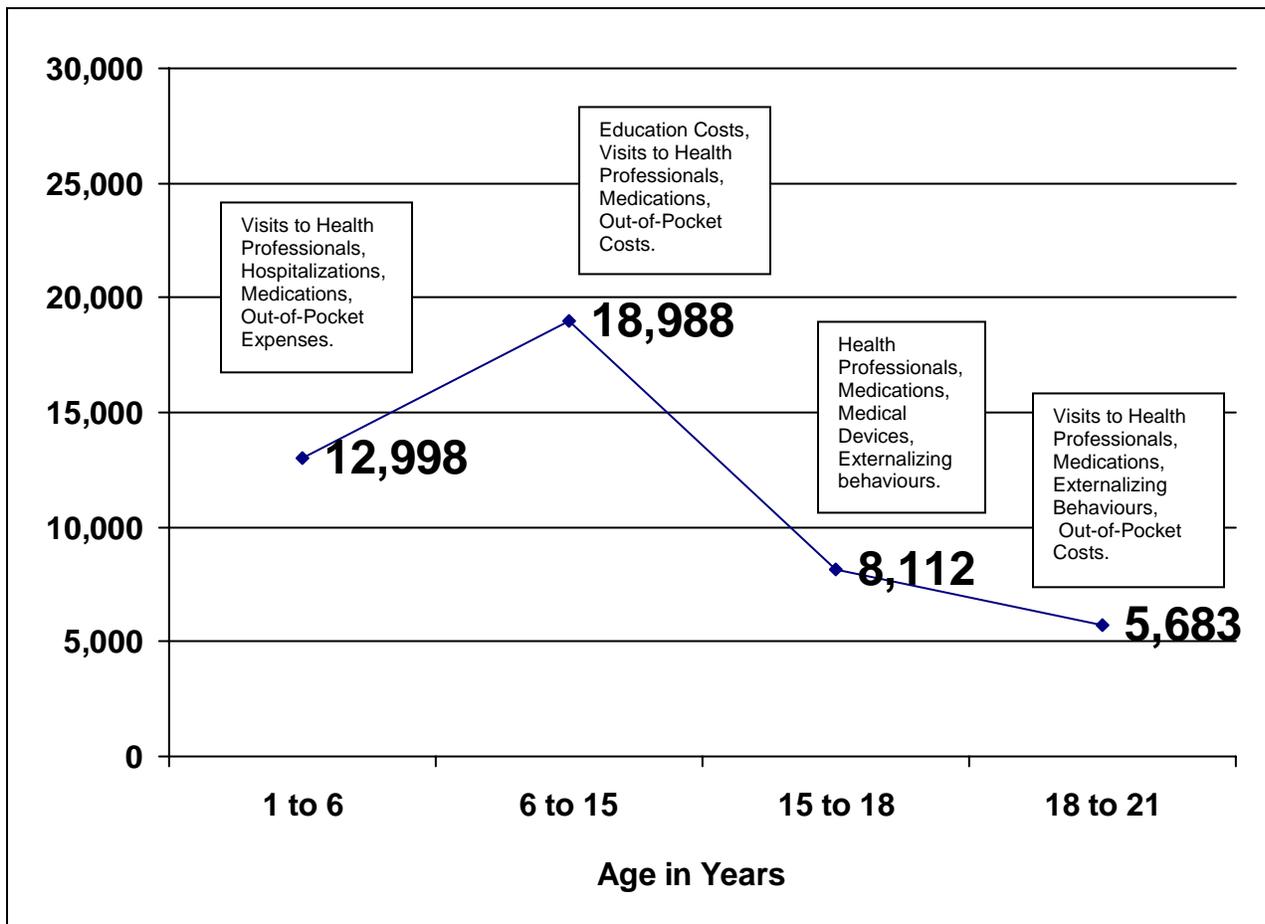
Disease Severity	n	Mean	95% CI
Mild	77	\$9,756	\$ 5,828, \$13,684
Moderate	39	\$14,323	\$13,020, \$15,755
Severe	22	\$19,494	\$10,137, \$28,851

**TABLE 6** Adjusted Annual Cost per Child According to Geographical Region

Geographical Region	n	Mean	95% CI
East	46	\$9,756	\$ 8,781, \$10,731
Central	52	\$12,922	\$10,338, \$15,506
West	50	\$16,450	\$15,957, \$16,943

**FIG. 2** Adjusted Annual Costs and Most Common Cost Components According to Age Groups

**Annual Costs (\$)**



**DISCUSSION**

**Current and Past Research**

This study has illustrated that the burden of prenatal exposure to alcohol is profound. The adjusted average annual costs per child with FAS and FAE, ages 1 to 21 years, in Canada was \$14,342. The largest single component of costs was education costs and medical costs accounting for 32.6 % and 30.3 % respectively. The current study attempted to overcome the limitations of past research by including estimates of costs of FAE, direct costs to the family/patient and measuring productivity losses of parent(s). Discrepancies in cost estimates obtained in past studies were due, in part, to the time horizon used in the evaluation. Similar to research conducted

by Harwood & Napolitano, the current study estimated cost of treatment, care and lost productivity for all ages.<sup>13</sup> The current research presents detailed and disaggregated information on resource use and unit costs, and measured the costs of externalizing behaviours which were commonly not provided in past research. Finally, past studies provide estimates of the total cost of FAS to the nation. In the current research, estimates of the average cost associated per case of FAS will be more helpful in providing guidance as to what policies and interventions for prevention and treatment of FAS are appropriate. Case-specific costs provide more information, such as what types of costs are incurred at different ages, which can be used for developing programs and services.

### **Cost of FAS/FAE and Other Disabilities**

There is a paucity of research that examines the cost of childhood disability or illness at the individual patients. Thus, there were only two studies measuring cost associated with childhood disability or illness that could be directly compared to the results of the current study.<sup>26,27</sup> Leibson et al., found that children, ages 5 to 19 years, with attention deficit disorder (ADHD) (n=4119) have much higher medical expenses than their healthy peers.<sup>26</sup> Specifically, the annual medical costs of a child with ADHD was US \$4,306 versus US \$1,944 in the healthy child. The annual medical costs of a child with ADHD, is less than the annual medical costs, US \$5,686, of a child with FAS/FAE measured in the current research.

Although the prevalence of FAS/FAE is 5 to 20 times less than that of childhood asthma in Canada, the cost per case of FAS/FAE is considerably higher. For example, Ungar & Coyte assessed the cost of asthma in Ontario children. They found that adjusted annual societal costs per child in 1995 Canadian dollars varied from \$1,122 in children aged 4 -14 years to \$1,386 in children under 4 years of age.<sup>27</sup>

### **Determinants of Cost**

Severity of the disability as defined by cognitive functioning, severity of behaviour problems and organ anomalies impacted on the cost of FAS and FAE. This is in keeping with clinical findings, as children more severely affected will require more surgeries to correct heart defects, skeletal problems, hearing impairment and others. Similarly, children with severe cognitive and behavioral disabilities will require more specialized educational services, and health services such as psychologists, occupational therapists and others.

Age of the child impacted on the cost of FAS and FAE. Costs for children 6 to 15 years were higher than in all other groups. This reflects the costs of specialized education, the use of health specialists such as psychologists, psychiatrists, occupational therapists, and others. Costs began to plateau between the ages of 18 to 21 years. It is not clear if this reflects a decreased need for services, or a lack of available services as individuals leave the childhood/adolescent years. Qualitative data suggest that there is a lack of

services to assist young adults with FAS/FAE to enter the work force and leave the parental home. Living in the East (Newfoundland, New Brunswick, Nova Scotia, Quebec) resulted in significantly lower costs than central or western Canada. Interestingly, the lower costs did not reflect lower morbidity or lower utilization of services. Rather, parents living in the eastern region of Canada consistently noted that there were lack of educational, and health services for their children with FAS or FAE. This may cause the cost per case to be under-estimated. Future research that examines availability of resources per geographical region is needed.

The current study was not designed to estimate costs at the population level. Costs were calculated at the individual-patient level. However, a brief discussion on the cost of FAS/FAE to Canada seems warranted. A very conservative estimate of prevalence of FAS/FAE in Canada is approximately 3 in 1000 people.<sup>3</sup> The number of people ages 1 to 21 years in Canada is approximately 8 million (8,000,000).<sup>28</sup> Therefore, a conservative estimate of FAS/FAE in the 1 to 21 year age group in Canada is 24,000 individuals. The adjusted annual cost of FAS/FAE in Canada for ages 1 to 21 years at the individual level is \$14,342 (95% CI, \$12,986; \$15,698). Thus, a very conservative estimate of the cost of FAS/FAE annually to Canada is \$344,208,000 (95% CI \$311,664,000; \$376,752,000 or \$344.2 million (95% CI \$311.7 million; \$376.8 million).

In a review of the economic burden of illness in Canada, the cost of FAS is comparable to the annual cost of all birth defects (\$175.8 million) and all blood diseases (\$213.4 million).<sup>29</sup> Comparison of FAS/FAE to these conditions is limited because Health Canada included cost for all ages and did not include the productivity losses included in the current research. A clearer estimate of the prevalence of FAS/FAE in Canada, and of the cost of the syndrome for all ages, may allow a more confident comparison of FAS/FAE to other illnesses and disabilities.

### **Study Limitations**

While the study did not draw a random sample of children with FAS, the sampling plan included multiple areas of Canada and all children (parents) agreeing to participate were enrolled. Generalisability of the findings to the larger

Canadian population is supported by the heterogeneity of the sample. Individuals residing in institutions such as facilities for disabled children were not included in the sample and it is possible that individuals with severe disability were not well represented in this study. Individuals who were homeless or in the judicial system were not included in the study and may have lead to a somewhat lower cost estimate.

As diagnosis in infancy is rare, no infants were included in the study. We thus, elicited health services utilization information about neonatal care and services from all of the parents who participated. The time lapse between services and data collection limited the accuracy of the data related to neonatal costs. Health care resource data were based on parent reports and were not verified by agencies or services providing care. However, Browne et al. examined the validity of the Health Services Utilization Inventory, by comparing patients' recall of services with clinics' records of services utilized during the 2 months following referral.<sup>30</sup> The observed agreement ranged between 0.72-0.99. The Kappa statistic, which adjusts for chance agreement, ranged between 0.48-0.89, and consistently reflected adequate levels of agreement between the patients' reports and the clinic records.

### **Implications to Practice and Research**

By providing a detailed description of the cost-of-illness and the utilization of health services from various perspectives, this research may assist clinical and policy decision-makers to develop optimal disease management strategies and allocation of scarce health care funds. The increasing focus on screening and diagnosis of FAS demands that greater attention be paid to the delivery of care to this population. The study findings illustrated that 19% of total costs were paid by parents caring for their child with FAS.

Decision-makers should be aware of the substantial long-term economic impact of prenatal exposure to alcohol and should be sensitive to the financial constraints faced by parents, particularly those with low incomes.<sup>31</sup> In addition, given the high adoption rate among this population, the results of this study support the need for subsidized adoption of children with FAS. Finally, the data obtained demonstrated the significant burden of prenatal exposure to alcohol. It is

necessary for nurses and other health professionals to identify and implement health promotion strategies to prevent this completely preventable condition.

Broad economic aggregates, attributable to the sequelae of prenatal exposure, can provide a basis for assessing competing strategies for prevention, treatment and further research. It should be noted that cost data alone can not identify the most efficient allocation of finite health care resources. Rather, it is information on incremental costs and incremental health gains attributable to particular health care activities that can identify the combination of human and material inputs that maximize health benefits.

While there is general agreement among researchers on what constitutes direct, indirect and intangible costs, the methods used to collect cost and utilization information, require further research and development. For example, it was difficult to measure productivity loss of parents of children with FAS. Several parents reported a career change or lack of career opportunity because of the child's disability. Much of the data provided, although honest, was speculative, and more accurate methods for measuring such costs must be developed.

The cost of disability analysis revealed the contribution of indirect costs to the total costs of prenatal exposure to alcohol. The quality of the human capital approach continues to evolve with greater application of sensitivity analysis and exposition of inputs, resulting in more accurate estimates of indirect costs. Further research into this and other methods are required to improve the estimation of indirect costs.

Finally, future studies including prospective data of costs in infancy may capture more completely the economic burden of prenatal exposure to alcohol.

### **CONCLUSION**

This study illustrated that the profound cost burden of prenatal exposure. It anticipated that the results of this research will catalyze future research. Most importantly, it is anticipated by articulating the cost burden of prenatal exposure to alcohol, that this study will help children with FASD.

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