CME/MOC

A Multicountry Updated Assessment of the Economic Impact of Fetal Alcohol Spectrum Disorder: Costs for Children and Adults

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Aim: To conduct a systematic review and quantitative analysis of the world literature on the economic impact of fetal alcohol spectrum disorder (FASD).

Methods: A comprehensive literature review was conducted using multiple electronic databases and reference materials.

Results: Thirty-two studies from 4 countries met the inclusion criteria (United States [n = 20], Canada [n = 9], Sweden [n = 2], and New Zealand [n = 1]). The studies reported the economic impact of FASD on health care, special education, residential care, criminal justice system, productivity losses due to morbidity and premature mortality, productivity losses of caregivers of children with FASD, and intangible costs. The economic estimates vary considerably due to the different methodologies used by different studies. The mean annual cost for children with FASD was estimated to be $22,810 and for adults $24,308. Residential costs for children with FASD were 4-fold greater than for adults with FASD. The costs of lost productivity for adults were 6.3-fold greater than for children.

Conclusions: The data on the economic burden of FASD are scarce, and the existing estimates likely underestimate the full economic impact of this disorder on the affected individuals, their caregivers, and society. However, the current research is sufficient to demonstrate that FASD is a serious public health problem associated with tremendous economic burden.

Key Words: costs, fetal alcohol spectrum disorder, morbidity, productivity losses, systematic review

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Individuals who are prenatally exposed to alcohol exhibit a wide variety of symptoms originally described as fetal alcohol syndrome (Jones et al., 1973), later referred to as fetal alcohol spectrum disorders (FASDs). The umbrella term FASD includes fetal alcohol syndrome (FAS), partial fetal alcohol syndrome (pFAS), and alcohol-related neurodevelopmental disorder (ARND), and depending on the diagnostic guideline, alcohol-related birth defects (ARBDs) (Chudley et al., 2005; Hoyme et al., 2016).

The most recent framework utilizes a single designation of FASD as a diagnostic term with the specification of the presence or absence of the characteristics facial features (which combines the categorical designations of FAS, pFAS, and ARND) (Cook et al., 2016). In addition, the category of neurodevelopmental disorder (the term ''neurodevelopmental'' is often replaced with ''neurobehavioral'' when referring to adults) associated with prenatal alcohol exposure (ND-PAE) has been proposed (American Psychiatric Association, 2013). This diagnostic category is similar to ARND (Johnson et al., 2017).

A recent study identified 428 comorbid conditions that have been reported in persons with FASD (Popova et al., 2016c). Specific health problems experienced by individuals with FASD include congenital malformations, chromosomal abnormalities, prenatal and postnatal growth delays, intellectual disability, behavioral disorders, speech and language difficulties, visual and audiological impairments, cardiac deformities, and urogenital problems. Alcohol is a teratogen that can damage any organ or system of the developing fetus, and may have lifelong health consequences (Popova et al., 2016c).

Due to the complexity of neurological impairments in FASD, people are predisposed to the development of other common comorbid adverse outcomes later in their life such as difficulties at school, mental health problems, lower levels of productivity, unemployment, drug and alcohol dependence, homelessness, and increased contact with the criminal justice system (Streissguth et al., 1996). People with FASD often require lifelong and multidimensional services to address their ever-changing and complex needs. Unsurprisingly, such interventions generate substantial costs to society.

Understanding the extent of the costs of FASD to society is critical for program development, resource

prioritization, and funding prevention programs. The financial trade-off of cost of prevention programs for short-term and long-term savings should be a practical motivator of public policy surrounding funding of FASD prevention efforts. Cost estimates aid in the prioritization of funding for substance use disorder treatment programs and for targeted interventions for women (Single et al., 2003). Therefore, improving the quality of cost studies and resulting cost estimates will enhance the capacity of policy makers and funders to make informed decisions regarding the advantages and disadvantages of programs directed at diminishing the effects of alcohol use during pregnancy and FASD.

The objectives of this study were to conduct a comprehensive systematic review of the existing literature on the economic cost of FASD for children and adults; to assess the quality of existing cost studies; and to suggest areas of future research.

METHODS

Systematic Literature Search

A comprehensive systematic search of literature published before June, 2017, inclusive of all languages, was performed to identify studies reporting on the economic impact of FASD. The search was not limited geographically. Subject headings and keywords were utilized to search the following electronic bibliographic databases: PubMed, Scopus, Cochrane Database of Systematic Reviews, ClinicalKey, CINAHL, Google Scholar, and PsychINFO. The economic databases EconLit and Business Source Complete were also searched. Finally, a manual review of bibliographies in select studies that fulfilled inclusion criteria was performed.

Literature searches in the above bibliographic databases were conducted using multiple combinations of the following key words:

(1) Disease conditions: fetal alcohol, FASD, FAS, pFAS, fetal alcohol effects (FAE), ARND, ARBD, prenatal alcohol exposure;
(2) Outcomes: disability, disability adjusted life years, quality adjusted life years, morbidity, premature mortality, potential years of life lost, productivity losses;
(3) Cost: cost, economic, social cost, economic cost, direct cost, indirect cost, intangible cost; and
(4) Categories of cost: hospitalization, hospital days, ambulatory care, emergency room visits, family physician visits, prescription drugs, addiction services, child welfare, early childcare, special needs, assessment, suspensions, staff time, salaries, home support services, residential care, respite care, and corrections.

An example of a keyword search used is: (''fetal alcohol'' OR fasd) AND (cost$ OR economic). A graphic depiction of the search strategy is presented in Appendix 1, http://links.lww.com/JAM/A92.

Inclusion Criteria

Studies were included if they consisted of original research published in a peer-reviewed journal or scholarly report; reported the cost associated with a particular cost category for individuals with FASD; and specified the age of studied population.

Exclusion Criteria

Studies were excluded if they did not estimate costs of FASD; did not report the sample size; reported a pooled economic estimate by combining several studies; and were published in iteration.

Data Extraction

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, sample size, age of study population, direct and indirect cost categories, intangible costs, annual cost and lifetime cost per person with FASD, and also annual cost for all persons with FASD.

Two investigators conducted each study selection step independently; any disagreements were reconciled by team discussion.

The literature search produced 522 abstracts. Of those, 318 studies that did not report on the economic impact of FASD were excluded, leaving 204 studies that contained information regarding the economic toll of FASD. One hundred fifty studies were subsequently excluded due to the absence of information regarding monetary cost and/or cost categories associated with the economic impact of FASD. In total, 32 studies that met the inclusion criteria were retained for data extraction and analysis. A flow diagram of the systematic search strategy is presented in Appendix 1 (http://links.lww.com/JAM/A92).

Analysis

Sample size, and also costs per person or total cost were used to estimate the cost per person for each study and the overall cost per person and per child for each cost category. Minimum, median, and maximum sample sizes and costs per person for each cost category were estimated.

Bubble graphs of the average cost per person for each study using sample size as the size of the bubble were made for total costs, health care, residential care, productivity losses, and special education. The overall mean for the studies' cost per person and cost per child, and for each of the cost categories were provided as grand means by taking the sum of total costs for each study divided by the sum of sample sizes. This weighted the means proportionally to their sample sizes. Z tests for proportions and 95% confidence intervals (CIs) were used to estimate the significant proportions of child costs to all costs.

Comparison of Cost

Identified studies used the local currency for estimating costs and used the same currency year as the costing year. To facilitate comparison between individual studies and to standardize past costs to present equivalents, annual and lifetime total costs were converted to May, 2017 currency values using online inflation calculators. The estimated costs in the USA and South Africa were converted to May, 2017 values using currency inflation rates from the Consumer Price Index provided by the US Bureau of Labor Statistics

(https://www.bls.gov/data/inflation_calculator.htm). The estimated costs in Canada were converted to 2017 values using currency inflation rates from the Consumer Price Index provided by the Bank of Canada (http://www.bankofcanada.ca/rates/related/inflation-calculator/?page_moved=1). The costs in New Zealand were converted to 2017 currency values using the inflation calculator provided by the Reserve Bank of New Zealand (http://www.rbnz.govt.nz/monetary-policy/inflation-calculator). Costs for Sweden were converted to 2017 costs using https://www.statbureau.org/en/eurozone/inflation-calculators.

Qualitative Ranking of Studies

The Drummond 10-point *Qualitative ranking of checklists* is a widely used tool for the critical appraisal of economic evaluations (Drummond et al., 2005). The Drummond checklist was designed to assess economic evaluations, which include a critique of cost studies. A modification of both the Drummond checklist and the variation of the Drummond checklist used by Navarro et al. was adapted to make a 9-point quality rating index checklist that was more specific and functional for analyzing FASD-specific cost studies (Navarro et al., 2011). A study with a score of 1 was deemed of low quality and a study with a score of 9 was deemed of high quality (see Appendix 2, http://links.lww.com/JAM/A93). We found no study meeting criteria 2 in the Drummond Checklist, so we excluded this criterion from the table.

RESULTS

A total of 32 studies (Table 1) were identified and included in the analysis from the following 4 countries: United States (n = 20 studies) (Russell, 1980; Stanage et al., 1983; H.J. Harwood et al., 1984; Harwood and Napolitano, 1985; Abel and Sokol, 1987; Parker et al., 1987; Weeks, 1989; D. Rice et al., 1990; D.P. Rice et al., 1991; D.P. Rice, 1993; H. Harwood et al., 1999; H. Harwood, 2000; Popova et al., 2011b; Klug and Burd, 2003; Miller et al., 2006; Rosen et al., 2008; Popovici et al., 2009; Amendah et al., 2011; Bouchery et al., 2011); Canada (n = 9) (Stade et al., 2006, 2009; Fuchs et al., 2008, 2009; Thanh and Jonsson, 2009, 2010, 2014; Thanh et al., 2015; Popova et al., 2016b; Alaska Mental Health Trust Authority, 2017); Sweden (n = 2) (Johansson et al., 2006; Ericson et al., 2017); and New Zealand (n = 1) (Easton et al., 2016). Year of study ranged from 1983 to 2013. Nine studies (Stanage et al., 1983; Abel and Sokol, 1987, 1991; Parker et al., 1987; Klug and Burd, 2003; Stade et al., 2006; Fuchs et al., 2008; Fuchs et al., 2009; Amendah et al., 2011) reported cost associated with FASD among children aged birth to 21 years.

Please note that Harwood et al. (1984) and Harwood and Napolitano (1985), and also Rice et al. (1990, 1991) utilized some overlapping data (H.J. Harwood et al., 1984; H.J. Harwood and Napolitano, 1985; D. Rice et al., 1990; D.P. Rice et al., 1991). A Canadian cost study published by Popova et al. (2016b) contained data from 9 other studies (Popova et al.,

TABLE 1. A List of Studies Included in the Current Analysis

Study Number	Reference	Year	Country	Only Children
1	Harwood et al., 1984; Harwood and Napolitano, 1985	1980	**United States**	No
2	Abel and Sokol, 1987	1984	United States	Yes
3	Abel and Sokol, 1991a	1984	United States	Yes
4	Abel and Sokol, 1991b	1987	United States	Yes
5	Rice et al., 1990; Rice et al., 1991	1985	United States	No
6	Rice, 1993	1990	United States	No
7	Harwood et al., 1998	1992	United States	No
8	Harwood, 2000a	1998	United States	No
9	Popova et al., 2011b	2003	United States	No
10	Miller et al., 2006	2001	United States	No
11	Amendah et al., 2011	2003–2005	United States	Yes
12	Bouchery et al., 2011	2006	United States	No
13	Russell, 1980	1987	United States	No
14	Stanage et al., 1983b	1983	United States	Yes
15	Parker et al., 1987	1983	United States	Yes
16	Weeks, 1989	1988	United States	No
17	Alaska Mental Health Trust Authority, 2017	2016	United States	No
18	Klug and Burd, 2003	1996–1997	United States	Yes
19	Rosen et al., 2008	2005	United States	No
20	Popovici et al., 2009	2007	United States	No
21	Stade et al., 2006	2003	Canada	Yes
22	Stade et al., 2009	2007	Canada	No
23	Thanh and Jonsson, 2010	2009	Canada	No
24	Thanh et al., 2015	2014	Canada	No
25	Popova et al., 2016b	2013	Canada	No
26	Fuchs et al., 2008	2006	Canada	Yes
27	Fuchs et al., 2009	2006	Canada	Yes
28	Thanh and Jonsson, 2009	2008	Canada	No
29	Thanh and Jonsson, 2014	2003–2012	Canada	No
30	Easton et al., 2016	2013	New Zealand	No
31	Johansson et al., 2006	2002	Sweden	No
32	Ericson et al., 2017	2014	Sweden	No

TABLE 2. Economic Cost Per Year Associated With FASD

Type of Cost	No. Studies All Ages (Children Only)	Sample Size			Cost Per Person			Grand Mean Cost/Person All Ages*←	Grand Mean Cost/Person Children†←
		Min	Median	Max	Min	Median	Max		
All costs	30 (8)	18	7138	380,000	$2035	$17,841	$298,975	$23,804	$22,810
United States	21 (7)	18	1949	380,000	$2035	$20,000	$298,975	$68,151	$38,995
Canada	6 (1)	20,481	144,453	307,136	$3691	$7995	$19,787	$12,470	$22,351
Sweden	2 (0)	56		19,000	$4710		$93,735		
New Zealand	1 (0)		44,300			$2035			
Health care	20 (8)	18	1563	380,000	$471	$4310	$38,264	$4954	$2978
Residential	11 (5)	79	1872	267,000	$1209	$14,077	$62,705	$3359	$13,573
Productivity losses	10 (1)	79	16,291	225,000	$300	$5976	$18,696	$6674	$1055
Special education	7 (2)	79	529	114,000	$812	$4941	$15,059	$7179	$4549
Intangible	2 (0)	1097		12,100	$12,713		$24,380	$23,410	
Correction	1 (0)		4148			$72,188		$72,088	

Estimates based on examined studies for all ages and children by country and cost category.
*Calculated based on 1 to 30 available studies.
†Calculated based on 1 to 8 available studies that only included children.

2012, 2013a, 2013b, 2014a, 2014b, 2015, 2016a; Easton et al., 2014, 2015), which were used in the current analysis. Studies with overlapping data were excluded, and only the most complete study was used. One study was from Western Cape, South Africa (Crede et al., 2011), which surveyed caregivers of children with FASD (0–12 years of age) to estimate healthcare services, was excluded from the current quantitative analysis because it reported costs from a country with a developing economy, which is not comparable to the costs of developed countries. We felt that aggregating data from these dissimilar economies would have led to biased cost estimates.

The following cost categories were estimated in the included studies: health care (n = 20 studies), residential care (n = 11), productivity losses due to morbidity and premature mortality and/or productivity losses of caregivers of children with FASD (n = 10), special education (n = 7), corrections systems (n = 1), and intangible cost (n = 2) (Table 2).

Sample sizes in the included studies varied widely, ranging from 18 to 380,000 for people of all ages, and from 18 to 380,000 for children (Table 2). Canadian studies had the largest sample sizes. Such variations in sample sizes were also present for all cost categories. Average total costs per person per year for all ages also varied greatly, with the lowest value being $2035 and the highest value being $298,975 (approximately 150 times greater). The US studies, included in this analysis, had the highest average cost reported ($68,151)—over 4 times higher than any other country. Residential costs had the lowest subcost per person at $3359 and special education the highest cost ($7179). Estimates for intangible and correction costs were not possible to conduct due to data unavailability.

We then considered the individual cost components for the total sample relative to the costs specific for children (Figs. 1–5). The size of the circle is relative to the size of the sample. Estimates of total costs per person (in US dollars) were similar for all persons ($23,804) and children ($22,810). This was only a 4.2% decrease in per person costs ($P = 0.509$) for children (Fig. 1). Greater differences were found for

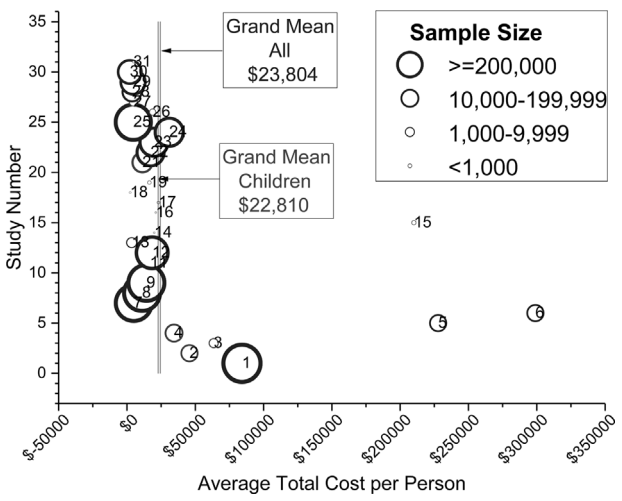

FIGURE 1. Grand mean of total annual cost per person with FASD (estimates from 32 studies; 8 had data for children).

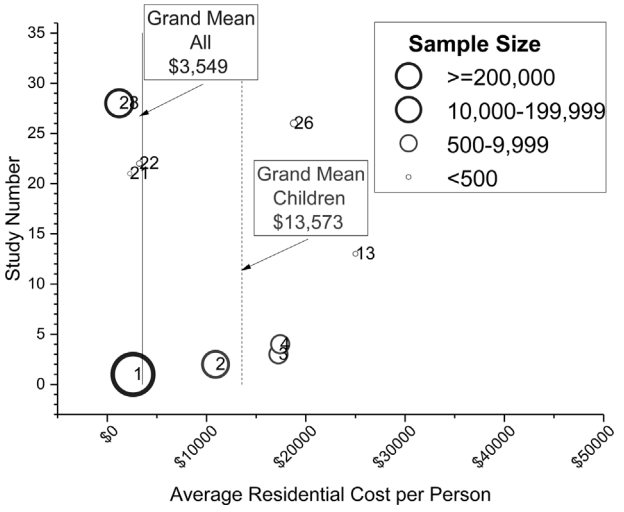

FIGURE 2. Grand mean of average cost of residential care per person (based on 9 studies; 5 had data for children).

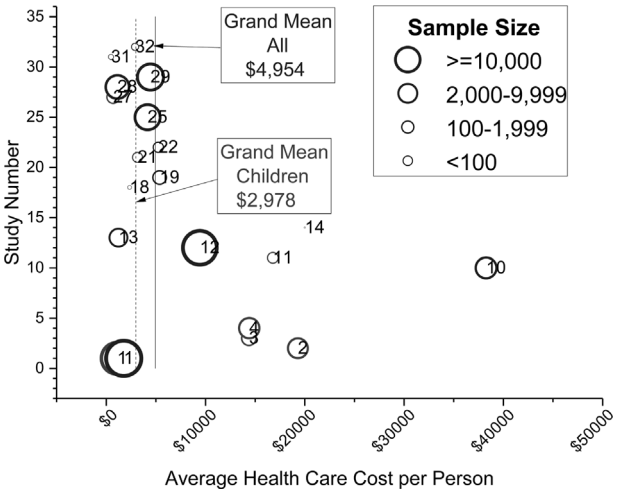

FIGURE 3. Grand mean of health care cost per person with FASD per year (estimates based on 20 studies; 8 had data for children).

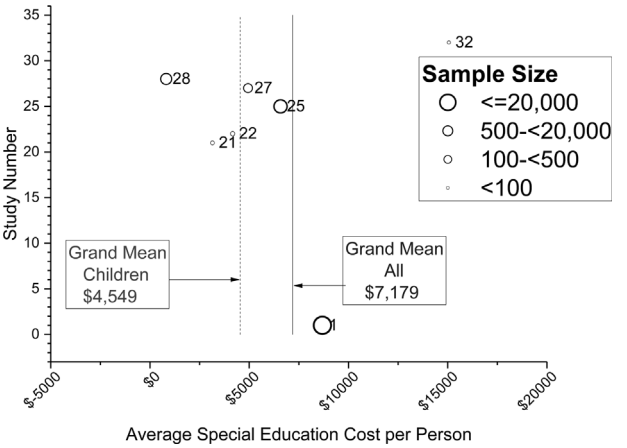

FIGURE 5. Grand mean of special education cost per person with FASD per year for children (estimates based on 2 studies) and all ages (estimates based on 7 studies).

individual cost categories. Costs of residential care were much higher for children ($13,573) than for adults ($3359)—a 282% increase (Fig. 2). In contrast, healthcare costs were lower for children than for adults by 40% (95% CI 10%–70%) (Fig. 3). Productivity losses due to morbidity and premature mortality and/or productivity losses of caregivers of children with FASD were decreased by 84% (95% CI 62%–107%) compared with adults (Fig. 4). Special education costs were also decreased by 37% (95% CI 7%–67%) (Fig. 5). Estimates for intangible and correctional costs could not be estimated due to lack of data.

DISCUSSION

There are only 5 countries in the world (all but 1 are developed countries), which have estimated economic impact of FASD. The studies examined the following cost components: health care, special education, residential care, criminal justice system, productivity losses due to morbidity and premature mortality, productivity losses of caregivers of children with FASD, and intangible losses. These studies demonstrate that FASD is associated with a significant economic burden on the individual, the family, and society. However, the estimated costs vary considerably due to the different methodologies and different cost drivers used by the studies.

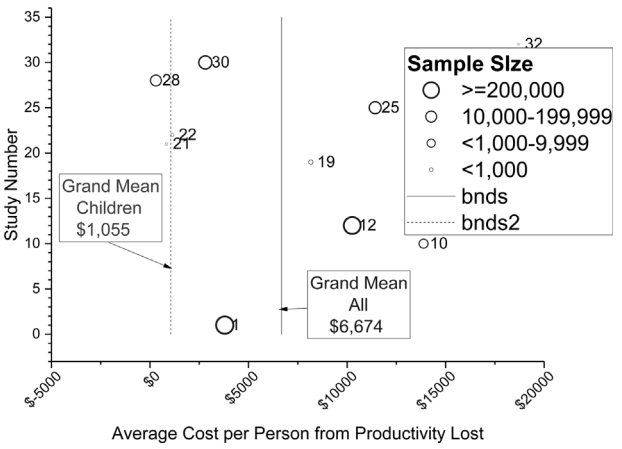

FIGURE 4. Grand mean of productivity losses due to morbidity and premature mortality cost per person with FASD per year for all ages (estimates based on 10 studies; 1 study had data for children).

Canada is the only country to have investigated the costs to the correctional system related to FASD (Popova et al., 2015). Prevalence estimates of FASD in the Canadian correctional system suggest a profound over-representation of individuals with FASD (Fast et al., 1999; Burd et al., 2003; Murphy et al., 2005; Rojas and Gretton, 2007; MacPherson et al., 2011; Popova et al., 2011a). To attest to this, it has been estimated that youth with FASD are 19 times more likely to be incarcerated than youth without FASD (Popova et al., 2011a). Cost studies have estimated that correctional services and the criminal justice system account for an average of 27.2% (range 11.3%–40%) of the economic burden of FASD. Thus, future cost studies on FASD in other countries should consider cost of corrections, and also police and courts as significant cost drivers.

The productivity losses of caregivers of individuals with FASD (informal caring) were recently estimated in a Swedish study (Ericson et al., 2017). The authors estimated that informal caring as €11,633, or 16.5% of the total cost per child with FASD (Ericson et al., 2017). By way of comparison, decreased productivity for adults (age 18–64) was estimated at €19,997 (21.3% of the total cost per adult with FASD). Thus, productivity losses of caregivers of children with FASD were found to be approximately 58% of the productivity losses of FASD-related morbidity. Productivity losses attributable to morbidity were 1 of the highest cost categories in every study where it has been investigated. The study by Ericson et al. (2017) examines productivity losses due to demands on the caregivers for children with FASD as a major cost component of losses in adults with FASD (Ericson et al., 2017).

In the United States, 2 studies estimated the intangible losses associated with FASD including the monetary value of pain, suffering, and reduced quality of life (Miller et al., 2006; Rosen et al., 2008). Theoretical literature supports the inclusion of intangible costs (Manning et al., 1991; Office of Management and Budget, 1992; Miller et al., 2006), and proclaims their importance with respect to regulatory cost-benefit analyses (Office of Management and Budget, 1992). Future studies that aim to demonstrate the significant impact of FASD on individuals and society, overall, should include estimates of intangible losses resulting from FASD.

Based on the scarcity of FASD cost data with respect to the criminal justice system, productivity losses from caregivers, and intangible costs, many of the existing cost estimates should be considered conservative.

Informing Public Policy

It has been estimated that 1% to 5% of live births in the developed world will be affected by FASD (Lange et al., 2017; May et al., 2018). While hundreds of thousands of children are born every year with this largely preventable condition, many countries devote less than 1% of the cost of caring for people with FASD to its prevention (Rice et al., 1991; Popova et al., 2016b).

In Fig. 6, we present cost comparisons for children (A) and adults (B). As a basis of comparison of FASD costs for children, we used autism spectrum disorder (Lavelle et al., 2014) and asthma (Nunes et al., 2017). The annual costs of autism spectrum disorder were $17,081 per year in the USA (Lavelle et al., 2014). A mean cost per patient per year, including all children with asthma (intermittent, mild, moderate, and severe asthma) in Europe is $USD 1900, which seems lower than USA—estimated mean $3100 (Nunes et al., 2017). The annual cost of care for a person with FASD exceeds costs for autism by 26% and asthma by 87% (Fig. 6). FASD now appears to be more prevalent than autism spectrum disorder (Lange et al., 2017; May et al., 2018). For adults, we compared cost of care for FASD with diabetes and epilepsy (Ng et al., 2014; Begley and Durgin, 2015). The cost of FASD exceeded cost of care for diabetes by 13% and for epilepsy by 56% (Fig. 6).

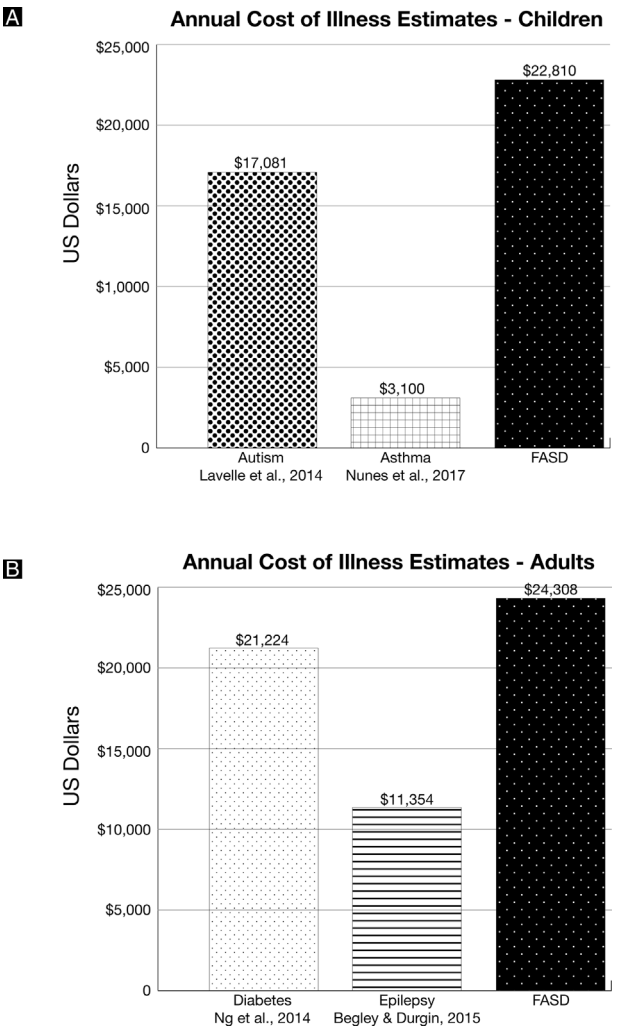

FIGURE 6. (A) Cost of illness comparison of children with fetal alcohol spectrum disorder (FASD with autism spectrum disorders and asthma. (B) Cost of illness comparison of FASD with diabetes and epilepsy in adults.

Limitations of the Examined Cost Studies

The quality of cost studies on FASD has improved since original reports. However, many limitations remain. Firstly, by far the biggest limitation for studies of FASD is the absence of nomenclature to identify and label those with FASD in health claims data. Presently, the International Classification of Disease, Ninth Revision, Clinical Modification (ICD-9-CM) code 760.61 is used; however, no diagnostic code exists for any specific FASD diagnostic category. Secondly, FASD is underdiagnosed, which means that the total costs are much higher at a national level than can currently be estimated. Thirdly, FASD is typically diagnosed around age 5 or 6; therefore, persons with FASD who die before age 5 or 6 would often not be identified as having FASD and would not be included. Fourthly, most cost studies do not include several important cost variables such as criminal justice system, productivity losses and unreimbursed costs of caregivers, cost of premature mortality and intangible losses, and thus, underestimate the overall cost. Fifthly, a current limitation in comparing studies from different countries is the inconsistent inclusion of different cost categories and definitions of these variables. Sixthly, it would be very useful to be able to compare people with FASD to people without FASD across cost categories or to compare people with FASD to people without FASD who have another condition of interest. This is currently difficult due to the large numbers of people with FASD who are undiagnosed. Future cost studies should build on high-quality cost methodologies with a consistent set of cost variables to generate data that can be comparable across other populations.

CONCLUSIONS

Every year, 630,000 new cases of FASD are born around the world (Lange et al., 2017). The annual per-person costs of care are $24,308 for adults and $22,810 per child. Costs for

residential care for children are 4-folds greater than for adults, and the costs of lost productivity are 6.3-folds greater for adults than for children. Huge investments in prevention and treatment of FASD are warranted.

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