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Prevalence of fetal alcohol spectrum disorder among special subpopulations: a systematic review and meta-analysis

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ABSTRACT

Aim To collate prevalence estimates of fetal alcohol spectrum disorder (FASD) among special subpopulations (defined by service use). **Design** Systematic literature review and meta-analysis of original, quantitative studies published between 1 November 1973 and 1 December 2018. The PRISMAGATHER were adhered to. The review protocol [includes FASD prevalence in (a) general and (b) special populations] is available on PROSPERO (registration number: CRD42016033837). Prevalence estimates were collated for all included studies with country-, disorder- [FASD and fetal alcohol syndrome (FAS)] and population-specific random-effects meta-analyses conducted. **Setting and Participants** A number of service-defined subpopulations globally (see Findings). **Measurements** The main outcome was the prevalence of FASD among special subpopulations. The critical appraisal of each study was conducted using the Joanna Briggs Institute tool. **Findings** We identified 69 studies, comprising 6177 individuals diagnosed with FASD from 17 countries: Australia (n = 5), Brazil (n = 2), Canada (n = 15), Chile (n = 4), eastern Europe (Moldova, Romania and Ukraine; n = 1), Germany (n = 1), Israel (n = 1), Lithuania (n = 1), the Netherlands (n = 1), Poland (n = 1), Russia (n = 9), South Korea (n = 1), Spain (n = 1), Sweden (n = 1) and United States (n = 25). FAS and FASD prevalence rates were collated for the following five subpopulations: children in care, correctional, special education, specialized clinical and Aboriginal populations. The estimated prevalence of FASD in these special subpopulations was 10–40 times higher compared with the 7.7 per 1000 (95% confidence interval = 4.9–11.7) global FASD prevalence in the general population. **Conclusions** Global subpopulations of children in care, correctional, special education, specialized clinical and Aboriginal populations have a significantly higher prevalence of fetal alcohol spectrum disorder compared with the general population, which poses a substantial global health problem.

Keywords Fetal alcohol spectrum disorder, fetal alcohol syndrome, prenatal alcohol exposure, prevalence, special subpopulations, systematic literature review and meta-analysis.

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INTRODUCTION

World-wide, nearly one in 10 (9.8%) women in the general population consume alcohol during pregnancy [1]. Prenatal alcohol exposure places these pregnancies at risk for many adverse outcomes, including fetal alcohol spectrum disorder (FASD), which is a life-long disability that requires assistance from a wide range of service providers including health, community and remedial education, among many others [2]. FASD has a very broad phenotype [3] and is further complicated by high rates of comorbidity—over 400 disease conditions have been reported to co-occur in people with FASD [4], with the most prevalent conditions occurring within the congenital malformations, deformities and chromosomal abnormalities (43%) and mental and behavioural disorders (19%) chapters of the International Statistical Classification of Diseases and Related Health Problems, 10th revision (ICD-10) [5]. Some comorbid

conditions (e.g. language, auditory, visual, developmental, cognitive, mental and behavioural problems) are highly prevalent, ranging from 50 to 91% [4]. Further, it was recently estimated that approximately one in every 13 prenatally alcohol exposed infants will have FASD, which results in approximately 630 000 infants being born with FASD in the world each year [6]. Given that FASD is a life-long disability, it is estimated that more than 11 million individuals between 0 and 18 years of age, and 25 million individuals between 0–40 years of age, have FASD in the general population world-wide [1].

Several studies have provided estimates of the cost of care for FASD among several populations or service providers [7–11]. These cost estimates demonstrate that FASD poses a life-time cost of approximately 1 million dollars [11] and, as such, the prevalence of FASD is a key factor in understanding the service demands and burden of FASD across different populations and various systems of care.

The prevalence of FASD in the general population as well as patterns of prenatal alcohol exposure during pregnancy (e.g. binge drinking, drinking throughout pregnancy or, most commonly reported, drinking during the first trimester of pregnancy) also appear to vary widely between countries and regions [1,6,12]. Understandably, the prevalence of FASD varies not only between countries, but also between different subpopulations and service systems [6]. However, no study consolidating all available data on the prevalence of FASD among all special subpopulations (e.g. children in care, psychiatric care populations, etc.) currently exists. Consolidating all existing evidence on the prevalence of FASD among special subpopulations will aid in the identification of knowledge gaps and areas of study for which evidence is limited or absent, with the intention of ultimately improving prevalence estimates. Improving estimates of FASD within special subpopulations and service-defined populations would provide improved data to plan services and budgets to serve people affected by prenatal alcohol exposure.

This is the first study, to our knowledge, to collate prevalence estimates of FASD among special subpopulations (defined by service utilization), utilizing all published studies in the world literature. In addition, country-, disorder- (FASD and Fetal Alcohol Syndrome (FAS; the dysmorphic subtype form of FASD)) and population-specific random-effects meta-analyses were conducted for countries with available data. The meta-analysed FASD prevalence estimates were compared with the global FAS/FASD prevalence [1,6].

METHODS

The systematic literature search and meta-analyses were conducted and reported according to the standards set out in the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA), provided in the PRISMA Checklist in the Supporting information, Appendix S1 [13]. We have also adhered to the Guidelines for Accurate and Transparent Health Estimates Reporting guidelines [14].

Comprehensive systematic literature search

A comprehensive systematic literature search was performed to identify all studies that have reported the prevalence of FASD among a special sub-population. The search was conducted in multiple electronic bibliographic databases, including (in alphabetical order): Cumulative Index to Nursing and Allied Health Literature, EMBASE, Education Resource Information Center, MEDLINE, MEDLINE in process, PsychINFO, Scopus and Web of Science. The search was conducted using multiple combinations of the following key words: (1) epidemiolog*, frequenc*, incidence*, morbidit*, occurren*, prevalence*, probability, rate* OR statistic*; AND (2) alcohol* embryopath*, alcohol* related* neurodevelopmental* disorder*, alcohol* related* birth defect*, arnd, arbd, fetal* alcohol* effect*, fae, fas, fasd, fetal alcohol syndrome*, fetal alcohol spectrum disorder*, foetal* alcohol* effect, foetal* alcohol syndrome*, foetal* alcohol spectrum disorder*, pfas, partial fetal alcohol syndrome, partial foetal alcohol syndrome, prenatal* alcohol expos* OR pre-natal* alcohol expos*; AND (3) cohort stud*, cross* sectional stud*, prospective cohort stud* OR retrospective cohort stud*. The search was performed to identify studies published between 1 November 1973 and 1 December 2018, without language or geographical restrictions. Further, the content pages of the major epidemiological journals, as well as citations in the relevant articles, were manually screened. The full review protocol is available in PROSPERO [includes FASD prevalence in (a) general and (b) special sub-populations; http://www.crd.york.ac.uk/PROSPERO/), registration number CRD42016033837].

Inclusion/exclusion criteria

Articles were retained if they: (a) consisted of original, quantitative research published in a peer-reviewed journal or scholarly report; and (b) involved a measurement of the prevalence of FASD and/or FAS among a service-defined population. Additionally, articles were retained if they: (a) provided a measure of uncertainty (confidence interval or standard error); or (b) provided the number of cases or sample size (information to derive a measure of uncertainty). Articles were excluded if they: (a) lacked FASD prevalence data; or (b) contained prevalence estimates not specific to special subpopulations (i.e. general populations only). For a detailed list of criteria assessed for each included study please refer to the Supporting information, Appendix S2.

Study selection and data extraction

Study selection began by screening titles and abstracts for inclusion. Then, full-text articles of all studies screened as potentially relevant were considered. A data extraction form was developed to record relevant information, such as location of the study (country; province/territory or state), study year(s), sample size, setting, number of cases (by diagnostic category), prevalence (by diagnostic category), diagnostic guideline used, sex distribution of sample, age range of sample and method of ascertainment. Two investigators conducted each study selection step; any disagreements were reconciled by team discussion. All data were extracted by one investigator and then independently cross-checked by a second investigator; all discrepancies were reconciled by team discussion. Non-English-language studies deemed to be potentially relevant were translated either by colleagues fluent in the respective language or using Google Translate (and subsequently cross-checked by a native speaker).

Critical appraisal of included studies

The critical appraisal of each study was performed using the Joanna Briggs Institute tool, specifically designed for use in systematic reviews addressing questions of prevalence [15]. The following seven criteria were used: (i) representativeness of the sample to the target population, (ii) appropriate recruitment of participants, (iii) adequate sample size, (iv) detailed description of participants and setting, (v) sufficient coverage of the identified sample, (vi) use of an objective, standard criteria for ascertaining FASD and (vii) appropriateness of statistical analysis. The explanation of every criterion included in this tool is available in the Supporting information, Appendix S2.

Two investigators independently appraised the quality of each study, and all discrepancies in quality ratings were reconciled by team discussion.

Meta-analysis

Country-, disorder- (FAS and FASD, inclusive of FAS) and population-specific meta-analyses were performed for those countries with two or more studies that used active case ascertainment (ACA; where cases are actively sought and diagnosed) and/or clinic-based methods (prospectively conducted in prenatal clinics or hospitals) and specified the diagnostic criteria used to ascertain cases of FAS/FASD in the respective population. Although studies that utilized passive surveillance (PS) methods (the use of existing record collections, e.g. birth certificates, registries, medical charts, adoption records) were included in the current review, they were not used in the meta-analyses, as they are known to produce underestimates of the prevalence [16]. It is well known that the majority of the countries do not have the capacity and/or resources to use the ACA approach to identify FASD cases because FASD diagnosis requires a multi-disciplinary team and specialized clinical skills. Due to these circumstances, PS is the only option for the majority of the countries.

For all analyses, logit-transformed results were pooled using a Bayesian meta-analysis and non-informative (flat) prior distributions. The combined estimates were based on the mean of the posterior distributions and the 2.5th and 97.5th percentiles. The between-study variances were quantified using the τ^2 and I^2 statistics [17]. All models assumed fixed effects, as between-study heterogeneity is difficult to assess when there are only a small number of studies [17]. Publication bias was tested by visually inspecting a funnel plot for skewed distribution, using a ranked correlation test proposed by Begg & Mazumdar [18] and by employing a weighted regression test proposed by Egger and colleagues [19] (see the Supporting information, Appendix S3). Publication bias was assessed, as studies which measure FAS and FASD may have been established in specific segments of subpopulations where the prevalence of FAS and/or FASD is high (compared to other segments of the same subpopulation). Analyses were performed using the statistical software R, version 3.3.2 [20], and Stata statistical software, version 14.2 [21].

RESULTS

A total of 11 871 studies were identified in the search. Sixty-nine studies, comprising 6177 individuals diagnosed with FASD in total, were retained for data extraction. These studies represented the following 17 countries: Australia (n = 5), Brazil (n = 2), Canada (n = 15), Chile (n = 4), eastern Europe (Moldova, Romania and Ukraine; n = 1), Germany (n = 1), Israel (n = 1), Lithuania (n = 1), the Netherlands (n = 1), Poland (n = 1), Russia (n = 9), South Korea (n = 1), Spain (n = 1), Sweden (n = 1) and the United States (n = 25). A schematic diagram depicting the search strategy employed is presented in Fig. 1.

Following the identification of 69 studies, they were categorized into the following five special subpopulations: children in care (e.g. adoptees, foster children; n = 36), correctional (n = 8), special education (n = 3), specialized clinical (n = 5) and Aboriginal (n = 17).

The quality appraisals of the included studies indicated that 100% (n = 69) of studies were conducted on samples that were representative of the target population; 97.1% (n = 67) of studies appropriately recruited participants; 65.2% (n = 45) of studies had an adequate sample size; 84.1% (n = 58) of studies provided a detailed description of participants and setting; 95.7% (n = 66) of studies had sufficient coverage of the identified sample; 60.9% (n = 42) of studies used objective, standard criteria for ascertaining FASD; and 100% (n = 69) of studies used an

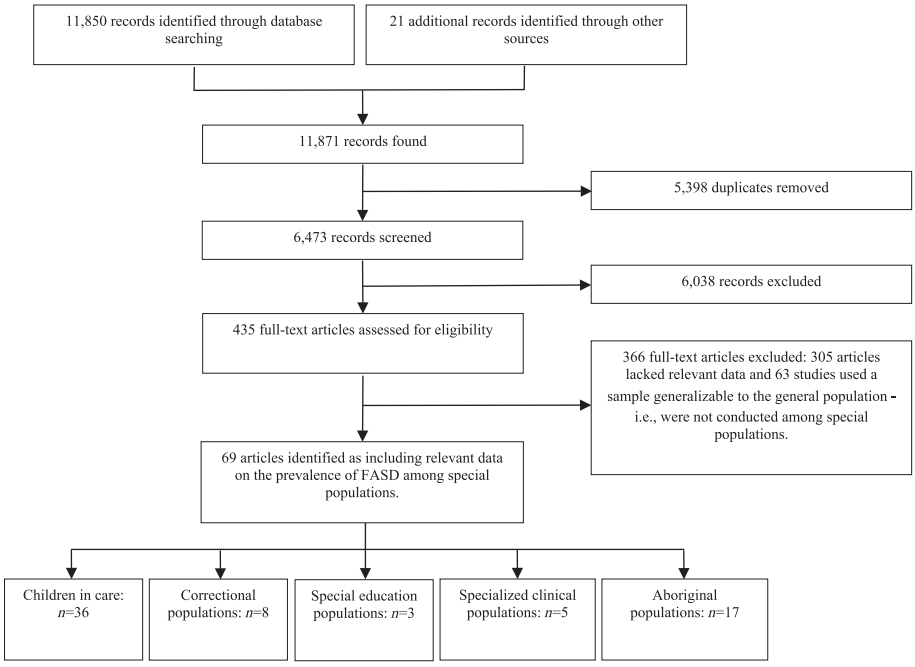

Figure 1 Schematic diagram depicting the search strategy employed

appropriate statistical analysis. Overall, 29.0% (n = 20) of studies met all seven criteria. The quality appraisals of the included studies are presented in the Supporting information, Appendix S2.

Prevalence of FASD among children in care

The prevalence of FASD among children in care was available for the following countries: Brazil (n = 1), Canada (n = 4), Chile (n = 2), Germany (n = 1), Israel (n = 1), Lithuania (n = 1), the Netherlands (n = 1), Poland (n = 1), Russia (n = 9), Spain (n = 1), Sweden (n = 1) and the United States (n = 12); one study [22] reported the prevalence of FAS among children in care from eastern Europe (Moldova, Romania and Ukraine; n = 1). Twenty studies used ACA, two studies used clinic-based methods, 10 studies used PS and four studies used mixed methods. Twenty (of 36) studies reported the diagnostic guideline/case definition used, with the majority (35.0%) using the four-digit diagnostic code [23] (see Table 1).

The prevalence of FAS was reported to be the lowest among pre-adoption children in orphanages and foster care in eastern Europe at 0.0 per 1000 (obtained via ACA) [22] and the highest among orphanages for children with developmental abnormalities in Russia at 680.0 per 1000 (obtained via ACA) [48], with median 79.1. The prevalence of FASD was reported to be the lowest among permanent wards in Canada at 32.6 per 1000 (obtained via PS) [26] and the highest among children in child welfare and homes for those with mental deficiencies in Chile at 611.7 per 1000 (obtained via ACA) [32], with median 177.3 per 1000.

A meta-analysis on the prevalence of FAS/FASD among children in care was conducted for the following three countries: Chile, Russia and the United States. Based on two studies [30,32], the pooled prevalence of FAS and FASD among children in care in Chile was estimated to be 51.9 per 1000 (95% CI = 40.3–64.9 per 1000) and 312.4 per 1000 (95% CI = 283.6–339.1 per 1000), respectively. In Russia, the pooled prevalence of FAS among children in care was estimated to be 95.5 per 1000 (95% CI = 85.3–105.4 per 1000) [39,41,44,46,47]. The pooled prevalence of FAS and FASD among children in care in the United States was estimated to be 142.3 per 1000 (95% CI = 117.3–167.8 per 1000) [51,53,54] and 251.5 per 1000 (95% CI = 220.0–281.7 per 1000) [54,57,61], respectively (Table 2 and Figs 2 and 3).

Prevalence of FASD among correctional populations

The prevalence of FASD among correctional populations was available for three countries: Australia (n = 1), Canada (n = 6) and the United States (n = 1). Two studies used ACA, one study used clinic-based methods, four studies used PS and one study used mixed methods. Five (of eight) studies reported the diagnostic guideline/case definition used; with the majority (28.6%) using the 2005 Canadian diagnostic guidelines [65] (see Table 3).

In Australia, the prevalence of FASD among a correctional population (73.7% were Aboriginal) was reported to be 363.6 per 1000 (obtained via ACA) [66]. In

Table 1 Study characteristics and prevalence of FAS and FASD among children in care (n = 36) reported in the identified studies, by country.

Reference	*Country (State/ Province/ Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
Children in care												
Strömland *et al.* 2015 [24]	Brazil (Recife)	NA	Orphanage	94	3	31.9	16	170.2	Clarification of the IOM criteria (Hoyme *et al.* 2005 [25])	57.4	3m–14	ACA
Burge, 2007 [26]	Canada (Ontario)	2003	Permanent wards	429	NA	NA	14	32.6	NA	NA	0–18	PS
Fuchs *et al.* 2005 [27]	Canada (Manitoba)	2004–05	Child welfare agencies	5664	NA	NA	640	113.0	NA	NA	0–20	PS
Fuchs & Burnside, 2014 [28]	Canada (Alberta, Manitoba, Ontario)	2010–14	Child welfare agencies	15 623 (Alberta: 6767; Manitoba: 8323; Ontario: 533)	NA	NA	1776 (diagnosed and suspected; Alberta: 699; Manitoba: 1021; Ontario: 56)	113.7	NA	51.3 (Alberta: 52.3; Manitoba: 50.1; Ontario: 57.8)	0–21	PS
Robert *et al.* 2009 [29]	Canada (Quebec)	2004–06	Adoptees from eastern Europe	29	1	34.5	7	241.4	4-digit diagnostic code (Astley & Clarren, 1999 [23])	59.0	4–8	ACA
Mena *et al.* 1987 [30]	Chile (VIII region)	1984	Foster care	931	43	46.2	184	197.6	Guidelines established by the Fetal Alcohol Study Group of the RSA (Rosett, 1980 [31])	57.0	NA	ACA

(*Continues*)

Table 1 (Continued)

Reference	*Country (State/ Province/ Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
Mena *et al.* 1993 [32]	Chile (Metropolitan region)	1989–90	Child welfare and homes for those with metal deficiencies	291	18	61.9	178	611.7	Guidelines established by the Fetal Alcohol Study Group of the RSA (Rosett, 1980 [31])	31.0	1–20+	ACA
Diamond *et al.* 2003 [22]	Eastern Europe (Romania: 73%, Ukraine: 22%, Moldova: 5%)	1999–2001	Pre-adoption: orphanage (84.1%) and foster care (15.9%)	82	0	0.0	NA	NA	NA	51.0	2m–4	ACA
Feldmann, 2012 [33]	Germany	NA	Foster care	267	62	232.2	NA	NA	Fetal Alcohol Syndrome Questionnaire (developed by Feldmann)	NA	NA	PS
Tenenbaum *et al.* 2011 [34]	Israel	NA	Pre-adoption and foster care	100	2	20.0	4	40.0	IOM criteria (Stratton *et al.* 1996 [35])	42.0	0–2	ACA
Kuzmenkoviene *et al.* 2012 [36]	Lithuania	NA	Orphanages	337	74	219.6	134	397.6	Clarification of the IOM criteria (Hoyme *et al.* 2005 [25])	NA	3–5	ACA
Knuiman *et al.* 2012 [37]	Netherlands	1999–2006	Adoptees for Poland	121	26	214.9	37	305.8	NA	52.1	5–17	PS (questionnaire administered to adoptive parents)

(***Continues***)

Table 1 (Continued)

Reference	*Country (State/ Province/ Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
Gyrczuk *et al.* 2014 [38]	Poland (Otwock)	2008–12	Pre-adoption intervention centre	490	108	220.4	NA	NA	NA	46.3	0–1	Clinic-based
Aronson, 1997 [39]	Russia	1994–97	Pre-adoption: orphanages	131	2	15.3	NA	NA	Guidelines established by the Fetal Alcohol Study Group of the RSA (Rosett, 1980 [31])	NA	NA	Mixed methods (ACA and PS)
Konovalova *et al.* 2009 [40]	Russia	NA	41 institutions (boarding schools with special needs programmes for those with mental deficiencies, regular and special needs orphanages, and schools of the social welfare system)	3675	320	87.1	557	151.6	NA	60.0	4–21	ACA
Miller *et al.* 2006 [41]	Russia	NA	Orphanages	234	17	72.7	NA	NA	4-digit diagnostic code (Astley & Clarren, 1999 [23]) and screening tool (Burd *et al.* 1999 [42])	52.0	1.5m–6	ACA
Miller *et al.* 2007 [43]	Russia	2004–05	Orphanages	193	19	98.5	NA	NA		54.4	2m–6	PS
Riley *et al.* 2003 [44]	Russia	1999	Boarding schools and orphanages for children with mental deficiencies	2352	186	79.1	NA	NA	Case definition provided	NA	NA	ACA

(*Continues*)

Table 1 (Continued)

Reference	*Country (State/ Province/ Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
The St. Petersburg-USA Orphanage Research Team, 2005 [45]	Russia	1997–2002	Orphanages	1167	112	96.0	NA	NA		NA	0–6	PS
Warren *et al.* 2001 [46]	Russia	NA	Boarding schools and orphanages	184	26	141.3	NA	NA	IOM criteria (Stratton *et al.* 1996 [35])	67.0	8–17	ACA
Bubnov, 2010 [47]	Russia, Yekaterinburg	2005–09	Orphanages	445	67	150.6	177	397.8	Clarification of the IOM criteria (Hoyme *et al.* 2005 [25])	NA	2m–4	ACA
Legonkova, 2011 [48]	Russia, St Petersburg	2004–10	Orphanages for children with psychoneurological problems and orphanages for children with developmental abnormalities	NA	NA	46.0–93.0 in orphanages for children with psycho-neurological problems; and 464.0–680.0 in orphanages for children with developmental abnormalities	NA	NA	4-digit diagnostic code (Astley & Clarren, 1999 [23])	NA	0–7	ACA
Olivan-Gonzalvo, 2011 [49]	Spain	2000–10	Adoptees from eastern Europe (Russia: 92%)	1062	117	110.2	NA	NA	4-digit diagnostic code (Astley & Clarren, 1999 [23])	60.0	NA	ACA
Landgren *et al.* 2010 [50]	Sweden	NA	Adoptees from eastern Europe (Estonia, Latvia, Poland Romania, Russia)	71	21	295.8	37	521.1	IOM criteria (Stratton *et al.* 1996 [35])	56.0	5–10	ACA

(***Continues***)

Table 1 (Continued)

Reference	*Country (State/ Province/ Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
Albers *et al.* 1997 [51]	United States	1991–95	Adoptees from Europe	56	1	17.9	NA	NA	Smith's Recognizable Pattens of Human Malformation (Lyons, 1997 [52])	46.0	2.5m–9	ACA
Astley *et al.* 2002 [53]	United States (Washington)	1999–2001	Foster care	600	6	10.0	NA	NA	4-digit diagnostic code (Astley & Clarren, 1999 [23])	52.0	NA	ACA
Chasnoff *et al.* 2015 [54]	United States (Illinois)	NA	Foster and adopted youth referred to a children's mental health centre	547	93	170.0	156	285.2	4-digit diagnostic code (Astley & Clarren, 1999 [23])	63.8	4–18	Clinic-based
Farina *et al.* 2004 [55]	United States	NA	Adoptees from Russia	29	0	0.0	10	344.8	NA	48.0	1–7	ACA
Johnson *et al.* 1996 [56]	United States	NA	Adoptees from Eastern Europe (Belarus: 2%, Poland: 1%, Romania: 4%, Russia: 76%, Other: 17%)	252	6	23.8	NA	NA	NA	NA	0–10	PS
Loman *et al.* 2009 [57]	United States	NA	Adoptees [post-institutionalized and foster care for Eastern Europe (21%), South America (21.5%), Asia (57%) and Africa (0.5%)]	200	NA	NA	8	40.0	CDC diagnostic guidelines (Bertrand *et al.* 2004 [58])	46.5	8–11	Mixed methods (ACA & PS)

(***Continues***)

Table 1 (Continued)

Reference	*Country (State/ Province/ Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
McGuinness *et al.* 2000 [59]	United States	1997	Adoptees from Eastern Europe	105	NA	NA	7	66.7	NA	48.0	6–9	PS
Miller & Hendrie, 2000 [60]	United States	1991–98	Adoptees from China	452	0	0.0	NA	NA	NA	2.0	2m–1	ACA
Miller *et al.* 2005 [61]	United States	1988–2004	Adoptees from Guatemala (orphanages, foster- and mixed-care settings)	103	NA	NA	19	184.5	4-digit diagnostic code (Astley & Clarren, 1999 [23])	53.0		Mixed methods (ACA & PS)
Miller *et al.* 2009 [62]	United States	2004–07	Adoptees from eastern Europe	138	NA	NA	10	72.5	NA	51.0	7m–5	ACA
Miller *et al.* 2009 [63]	United States	NA	Adoptees from eastern Europe (Bulgaria: 2%, Lithuania: 6%, Latvia: 2%, Moldova: 6%, Romania: 26%, Russia: 52%, Ukraine: 6%)	50	NA	NA	2	40.0	NA	52.0	8–11	Mixed methods (ACA & PS)
Ringeisen *et al.* 2008 [64]	United States	1999–2000	Child welfare agencies	5496	29	5.3	NA	NA	NA	50.0	0–14	PS

ACA = active case ascertainment; DSM = Diagnostic and Statistical Manual of Mental Disorders; FAS= fetal alcohol syndrome; FASD = fetal alcohol spectrum disorder; IOM = Institute of Medicine; m = months; NA = not available; ND–PAE: neurodevelopmental disorder associated with prenatal alcohol exposure; PS = passive surveillance; RSA = Research Society on Alcoholism.

Table 2 Pooled prevalence of FAS and FASD among special subpopulations.

Country	FAS/FASD	No. of studies	Prevalence per 1000 (%)	95% confidence interval per 1000	
				Lower	Upper
Children in care					
Chile	FAS	2	51.9 (5.2)	40.3	64.9
	FASD	2	312.4 (31.2)	283.6	339.1
Russia	FAS	5	95.5 (6.6)	85.3	105.4
United States	FAS	3	142.3 (14.2)	117.3	167.8
	FASD	3	251.5 (25.2)	220.0	281.7
Correctional populations					
Canada	FASD (adult)	2	146.7 (14.7)	98.2	204.9
Special education populations					
Chile	FAS	2	29.1 (2.9)	19.2	42.0
	FASD	2	84.2 (8.4)	66.6	103.1
Aboriginal populations					
Australia	FAS	2	2.3 (0.2)	1.4	3.5
	FASD	2	14.8 (1.5)	11.4	18.6
Canada	FAS	3	60.8 (6.1)	42.1	83.4
	FASD	3	43.6 (4.4)	37.9	49.3
United States	FAS	3	2.8 (0.3)	2.2	3.5
	FASD	2	4.4 (0.4)	3.5	5.3

Only studies that used active case ascertainment and/or clinic-based methods and specified the diagnostic criteria used to ascertain cases of fetal alcohol syndrome/fetal alcohol spectrum disorder (FAS/FASD) in the respective population were included in the meta-analyses. Studies that utilized passive surveillance methods were excluded from the meta-analyses.

Canada, the reported prevalence of FAS and FASD ranged from 0.0 per 1000 (obtained via ACA) [71] to 10.5 per 1000 (obtained via clinic-based methods) [69] and 17.5 per 1000 (obtained via ACA) [66] to 233.5 per 1000 (obtained via clinic-based methods) [69], with median 108.7. In the United States, the reported prevalence of FAS was 0.0003 per 1000 (obtained via PS) [75]. The medians for FAS and FASD prevalence estimates in this special subpopulation (all countries) were 0.05 per 1000 and 112.8 per 1000, respectively. The pooled prevalence of FASD among adults in the correctional system in Canada was estimated to be 146.7 per 1000 (95% CI = 98.2–204.9 per 1000) [70,71] (see Table 2 and Figs 2 and 3).

Prevalence of FASD among special education populations

The prevalence of FASD among special education populations was available for Chile (n = 2) and South Korea (n = 1). The reported prevalence of FAS and FASD among special education populations, obtained via ACA using the guidelines established by the Fetal Alcohol Study Group of the RSA [31], ranged from 21.1 per 1000 [77] to 42.3 per 1000 [78] with median 33.7 for FAS, and 75.8 per 1000 [77] to 88.1 per 1000 [76] with median 82.0 for FASD. The reported prevalence of FAS among a special education population in South Korea was 42.3 per 1000 (obtained via ACA using a study-specific case definition) [78] (see Table 3).

The pooled prevalence of FAS and FASD among special education populations in Chile was estimated to be 29.1 per 1000 (95% CI = 19.2–42.0 per 1000) [76,77] and 84.2 per 1000 (95% CI = 66.6–103.1 per 1000) [76,77], respectively (see Table 2 and Figs 2 and 3).

Prevalence of FASD among specialized clinical populations

The prevalence of FASD among specialized clinical populations was available for two countries: Brazil (n = 1) and the United States (n = 4). Three studies used clinic-based methods and two studies used PS. The reported prevalence of FAS among babies referred to genetic clinics in Brazil was 1.0 per 1000 (obtained via PS; diagnostic guideline/case definition used not specified) [79]. The prevalence of FASD was reported for three specialized clinical populations in the United States: psychiatric care population (n = 2), patients evaluated at genetic clinics (n = 1) and a developmentally disabled clinical population (n = 1). One study [80] used the DSM-5 criteria of ND-PAE [81] and one study [83] used the four-digit diagnostic code [23]; the remaining two studies did not report the diagnostic guideline/case definition used. The lowest prevalence of FAS was reported among patients evaluated at genetic clinics at 6.4 per 1000 (obtained via clinic-based methods) [82] and the highest prevalence was reported among a psychiatric care population at 82.0 per 1000 (obtained via PS) [83], with median 10.4. The lowest prevalence of FASD was reported among a developmentally disabled

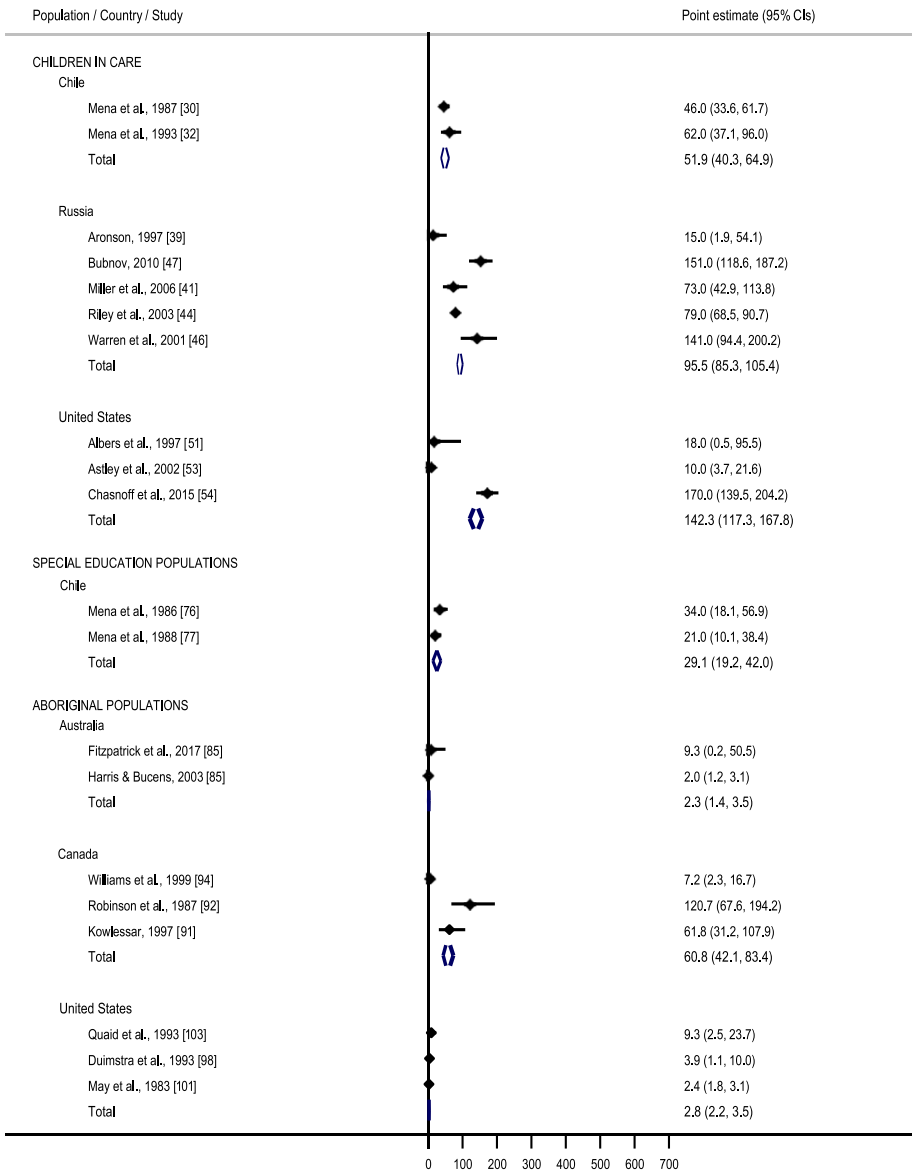

Figure 2 Forest plot of meta-analysed fetal alcohol syndrome (FAS) prevalence studies [Colour figure can be viewed at wileyonlinelibrary.com]

clinical population at 21.0 per 1000 (obtained via clinic-based methods) [84] and the highest among a psychiatric care population at 142.4 per 1000 (obtained via clinic-based methods) [80], with median 81.7 (see Table 3).

Based on inclusion criteria, it was not possible to conduct a meta-analysis on the prevalence of FAS/FASD among specialized clinical populations for any country.

Prevalence of FASD among aboriginal populations

The prevalence of FASD among Aboriginal populations was available for three countries: Australia (n = 4), Canada (n = 5) and the United States (n = 8). Seven studies used ACA, eight studies used PS and two studies used mixed methods. Twelve (of 17) studies reported the diagnostic guideline/case definition used, with the majority (17.6%) using the guidelines established by the Fetal Alcohol Study Group of the Research Society on Alcoholism (RSA) [31] (see Table 3).

In Australia, the reported prevalence of FAS and FASD ranged from 2.0 per 1000 (obtained via PS and clinic-based methods) [86] to 9.3 per 1000 (obtained via ACA) [85], with median 5.7 (FAS), and 4.1 per 1000 (obtained via PS) [88] to 194.4 per 1000 (obtained via ACA) [85], with median 9.7 (FASD), respectively. In Canada, the reported prevalence of FAS and FASD ranged from 7.2 per 1000 (obtained via ACA and PS) [94] to 120.7 per 1000 (obtained via ACA) [92], with median 61.8, and 7.0 per 1000 (obtained via PS) [93] to 189.7 per 1000 (obtained via ACA) [92], with median 66.9, respectively. In the United States, the reported prevalence of FAS and FASD ranged from 0.4 per 1000 (obtained via PS) [102]

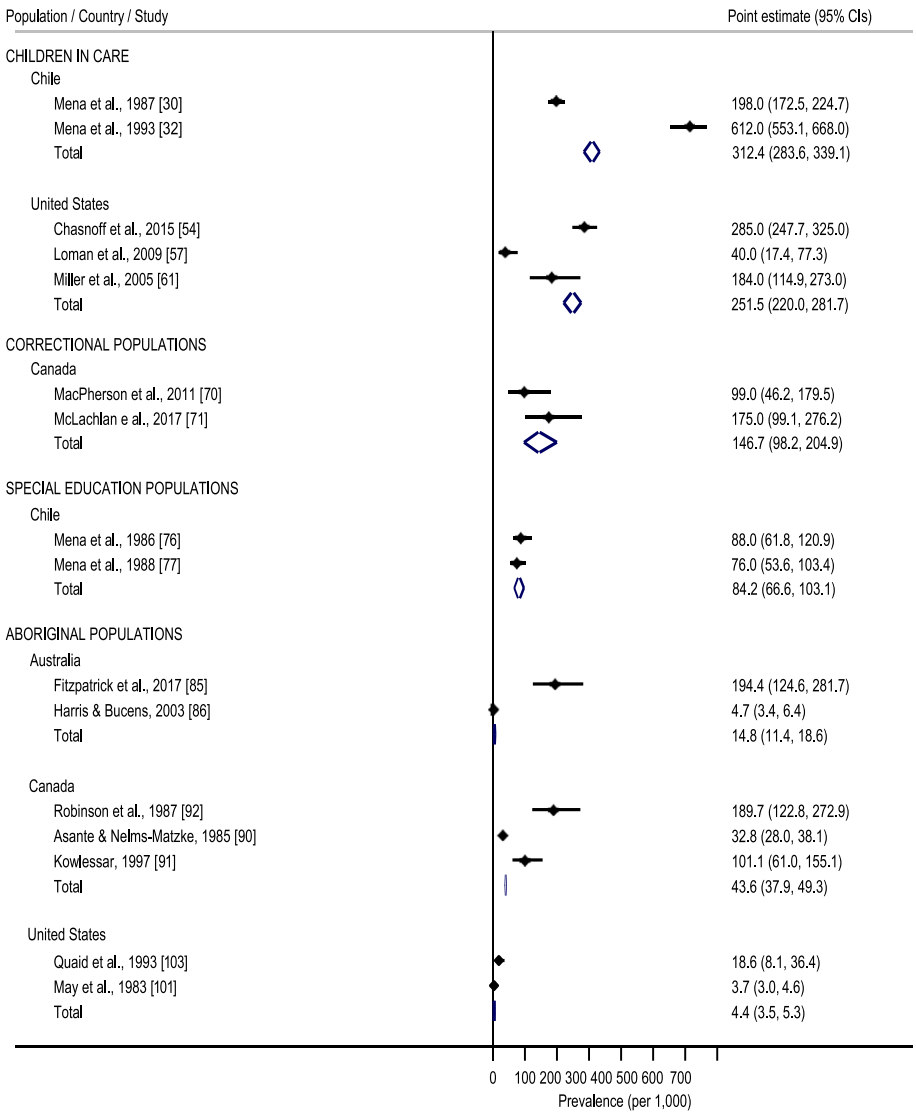

Figure 3 Forest plot of meta-analysed fetal alcohol spectrum disorder (FASD) prevalence studies [Colour figure can be viewed at wileyonlinelibrary.com]

to 9.3 per 1000 (obtained via ACA) [103], with median 2.8 for FAS, and 3.7 per 1000 (obtained via ACA) [103] to 18.7 per 1000 (obtained via ACA) [103], with median 11.2, for FASD.

In Australia, the pooled prevalence of FAS and FASD among Aboriginal populations was estimated to be 2.3 per 1000 (95% CI = 1.4–3.5 per 1000) [85,86] and 14.8 per 1000 (95% CI = 11.4–18.6 per 1000), respectively. In Canada, the pooled prevalence of FAS and FASD among Aboriginal populations was estimated to be 60.8 per 1000 (95% CI = 42.1–83.4 per 1000) [91,92,94] and 43.6 per 1000 (95% CI = 37.9–49.3 per 1000) [90–92], respectively. The pooled prevalence of FAS and FASD among Aboriginal populations in the United States was estimated to be 2.8 per 1000 (95% CI = 2.2–3.5 per 1000) [98,101,103] and 4.4 per 1000 (95% CI = 3.5–5.3 per 1000) [101,103], respectively (see Table 2 and Figs 2 and 3).

The pooled prevalence and results of the tests of heterogeneity and publication bias for the meta-analyses on the prevalence of FAS and FASD among subpopulations by country are presented in the Supporting information, Appendix S3.

Comparison of FASD prevalence in special subpopulations versus global FASD prevalence in general population

The meta-analysed prevalence estimates of FASD among special subpopulations appear to far exceed those found among the general population. For example, compared to the recently estimated global prevalence of FASD in the general population (7.7 per 1000; 95% CI = 4.9–11.7) [6], the prevalence among children in care was 32 times higher in the United States (251.5 per 1000; 95% CI = 220.0–281.7) [54,57,61] and 40 times higher in Chile (312.4 per 1000; 95% CI = 283.6, 339.1) [30,32];

Table 3 Study characteristics and prevalence of FAS and FASD among correctional populations ($n = 8$), special education ($n = 3$), specialized clinical populations ($n = 5$) and Aboriginal populations (17) reported in the identified studies, by country.

Reference	*Country (State/Province/ Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
Correctional populations												
Bower *et al.* 2018 [66]	Australia (western Australia)	2015–16	Youth detention centre; 73.7% Aboriginal	99	–	–	36	363.6	Australian Guide to the Diagnosis of FASD [67]	92.9	10–< 18	ACA
Burd *et al.* 2003 [68]	Canada	2001–02	Federal and provincial prisons	148 797	13	0.1	NA	NA	NA	91.2	NA	PS (survey)
Fast *et al.* 1999 [69]	Canada (British Columbia and Yukon)	1995–96	In-patient assessment unit of youth forensic psychiatric services	287	3	10.5	67	233.5	IOM criteria (Stratton *et al.* 1996 [35])	NA	12–18	Clinic-based
MacPherson *et al.* 2011 [70]	Canada (Manitoba)	2005–06	Male-only medium security penitentiary for adults	91	NA	NA	9	98.9	Canadian diagnostic guidelines (Chudley *et al.* 2005 [65])	100.0	19–30	Mixed methods [ACA and PS (interview)]
McLachlan *et al.* 2017 [71]	Canada (Yukon)	2014–15	Correctional centre, and offender and supervision Services	80	0	0.0	14	175	Canadian diagnostic guidelines (Chudley *et al.* 2005 [65])	NA	18–40	ACA
Murphy *et al.* 2005 [72]	Canada (British Columbia)	2004	Juvenile detention centres	137	NA	NA	16	116.8	NA	89.8	14–19	PS (survey)
Rojas & Gretton, 2007 [73]	Canada (British Columbia)	1985–2004	Youth Sexual Offence Treatment Programme	230	NA	NA	25	108.7	Case definition provided (based on Boland *et al.* 2000 [74])	100.0	12–18	PS
Burd *et al.* 2004 [75]	United States	2001–02	Prison systems and community corrections facilities	3 080 904	1	0.0003	NA	NA	NA	89.7	NA	PS (survey)
Special educational populations												
Mena *et al.* 1986 [76]	Chile (Concepción)	1982	Special schools for mentally handicapped children	386	13	33.7	34	88.1	Guidelines established by the Fetal Alcohol Study Group of the RSA (Rosett, 1980 [31])	NA	NA	ACA
Mena *et al.* 1988 [77]	Chile (Cautin, Concepción, Linares, Ranco)	1985–86	Special schools for mentally handicapped children	475	10	21.1	36	75.8	Guidelines established by the Fetal Alcohol Study Group of the RSA (Rosett, 1980 [31])	NA	NA	ACA

(*Continues*)

Table 3 (Continued)

Reference	*Country (State/Province/ Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
Lee *et al*, 2016 [78]	South Korea	NA	Institutions (children with mental retardation)	307	13	42.3	NA	NA	Case definition provided	NA	NA	ACA
Specialized clinical populations												
Grinfeld *et al*. 1999 [79]	Brazil (São Paulo)	1997	Babies referred to genetic clinics	16 640	17	1.0	NA	NA	NA	NA	NA	PS
Bell & Chimata, 2015 [80]	United States (Chicago)	2013–14	Psychiatric care population	611	NA	NA	87	142.4	DSM-5 criteria of ND-PAE (APA, 2013 [81])	43.0	4–78	Clinic-based
Cadle *et al*. 1996 [82]	United States (Kentucky)	1981–95	Patients evaluated at genetic clinics	4212	27	6.4	NA	NA	NA	NA	NA	Clinic-based
O'Connor *et al*. 2006 [83]	United States	NA	Psychiatric care population	122	10	82.0	NA	NA	4-digit diagnostic code (Astley & Clarren, 1999 [23])	81.1	NA	PS
Shanske & Kazi, 1980 [84]	United States (New York)	NA	Developmentally disabled clinical population	905	13	14.4	19	21.0	NA	NA	0–7	Clinic-based
Aboriginal populations												
Fitzpatrick *et al*. 2017 [85]	Australia (northwestern)	2010–11	School-aged children in very remote communities: community sites and local schools	108	1	9.3	21	194.4	Canadian diagnostic guidelines (Chudley *et al*. 2005 [65]) with adaptations to accommodate the cultural context	52.8	7.5–9.6	ACA
Harris & Bucens, 2003 [86]	Australia (northern Territory)	1990–2000	Paediatric wing, hospital	9077	18	2.0	43	4.7	Adapted 4-digit diagnostic code (Astley & Clarren, 1999 [23]) and the criteria by the AAP (2000 [87])	NA	0–10	Mixed methods (PS & Clinic-based)
Mutch *et al*. 2015 [88]	Australia (western)	1980–2010	Children captured in the Western Australian Register of Developmental Anomalies	45 078	NA	NA	188	4.1	NA	NA	0–15	PS

(*Continues*)

Table 3 (Continued)

Reference	*Country (State/Province/Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
Rothstein *et al.* 2007 [89]	Australia (Queensland)	2001–06	Children for specialist paediatric follow-up captured by the FNQ Paediatric Outreach Service	2195	NA	NA	32	14.6	NA	55.0	0–18	PS
Asante & Nelms-Matzke, 1985 [90]	Canada (northwest British Columbia and Yukon)	1983–84	Chronically handicapped children referred for assessment	5065	NA	NA	166	32.8	Guidelines established by the Fetal Alcohol Study Group of the RSA (Rosett, 1980 [31])	63.0	0–16	ACA
Kowlessar, 1997 [91]	Canada (Manitoba)	1981–90	Local school in First Nations community	178	11	61.8	19	101.1	IOM criteria (Stratton *et al.* 1996 [35])	NA	5–15	ACA
Robinson *et al.* 1987 [92]	Canada (British Columbia)	1984–85	Community-based: Native Indian community	116	14	120.7	22	189.7	Guidelines established by the Fetal Alcohol Study Group of the RSA (Rosett, 1980 [31])	49.6	3–18	ACA
Werk *et al.* 2013 [93]	Canada	2006	Canadian census survey catered to Aboriginal children living off-reserve	11 868	NA	NA	83	7.0	NA	NA	0–5	PS (survey)
Williams *et al.* 1999 [94]	Canada (Manitoba)	1994–96	Live births occurring in Thompson General Hospital in 1994	696	5	7.2	NA	NA	IOM criteria (Stratton *et al.* 1996 [35])	NA	NA	Mixed methods (ACA & PS)
Chávez *et al.* 1988 [95]	United States	1981–86	Birth Defects Monitoring Programme: hospitals with obstetric services	19 412	58	3.0	NA	NA	NA	NA	0–1 (newborns)	PS
CDC, 1995 [96]	United States (Iowa, Nebraska, North Dakota, South Dakota)	1981–92	Indian Health Service (IHS) and IHS contract facilities in tribal or American Indian communities	22 222	60	2.7	NA	NA	Criteria by Sokol & Clarren (1989 [97])	NA	0–31	PS
Duimstra *et al.* 1993 [98]	United States (Northern Plains)	1987–90	Indian Health Service facilities; IHS hospital out-patient settings; home visits	1022	4	3.9	NA	NA	Guidelines established by the Fetal Alcohol Study Group of the RSA (Rosett, 1980 [31])	NA	5–18m	ACA
Egeland *et al.* 1998 [99]	United States (Alaska)	1977–92	Paediatric practices that were referral centres for FAS; hospitals; regional native health corporations; state department of health and social services	37 346	114	3.1	NA	NA	Case definition provided	NA	0–16	PS

(***Continues***)

Table 3 (Continued)

Reference	*Country (State/Province/ Territory)*	*Study year(s)*	*Type of institution(s)/Setting*	*Sample size*	*Number of cases of FAS*	*Prevalence of FAS (per 1000)*	*Number of cases of FASD*	*Prevalence of FASD (per 1000)*	*Diagnostic guidelines/Case definition*	*Sex (% male)*	*Age range (years)*	*Method*
Fox *et al.* 2015 [100]	United States (Arizona, Colorado, New York)	2010	Surveillance site using multiple data sources: genetic/developmental clinics; hospitals; health maintenance organizations; Medicaid; juvenile justice system	13 938	28	2.0	NA	NA	Case definition based on IOM criteria (Stratton *et al.* 1996 [35])	NA	7–9	PS
May *et al.* 1983 [101]	United States (south-western USA: New Mexico, southern Colorado, southern Utah, northern Arizona)	1980–82	Children belonging to Navajo, Pueblo, and Plains culture tribes	22 963	55	2.4	85	3.7	Case definition provided	55.6	0–14	ACA
NBDPN, 2003 [102]	United States (24 States)	1996–2000	State programmes providing surveillance data on birth defects	77 630	32	0.4	NA	NA	NA	NA	0–1 (newborns)	PS
Quaid *et al.* 1993 [103]	United States (central Oregon)	1991	Indian Health Service Clinic and assisting health/social services personnel; dysmorphology clinic	429	4	9.3	8	18.7	Criteria by Sokol & Clarren (1989 [97])	NA	0–3	ACA

ACA = active case ascertainment; DSM = Diagnostic and Statistical Manual of Mental Disorders; FAS = fetal alcohol syndrome; FASD = fetal alcohol spectrum disorder; IOM = Institute of Medicine; m: months; NA = not available; NBDPN = National Birth Defects Prevention Network; ND–PAE = neurodevelopmental disorder associated with prenatal alcohol exposure; PS = passive surveillance; RSA = Research Society on Alcoholism.

the prevalence among adults in the Canadian correctional system (146.7 per 1000; 95% CI = 98.2, 204.9) [70,71] was 19 times higher; and the prevalence among special education populations in Chile (84.2 per 1000; 95% CI = 66.6–103.1) [76,77] was over 10 times higher. Overall, the estimated prevalence of FASD in these special sub-populations was 10-40 times higher compared with the prevalence estimate for the global general population: 7.7 per 1000 (95% confidence interval: 4.9-11.7).

Further, the prevalence reported in the individual studies is even more alarming. For instance, the prevalence of FASD among children in care with mental deficiencies in Chile was reported to be 620 per 1000 [32], among adoptees from eastern Europe it was more than 520 per 1000 [50] and among children residing in orphanages in Lithuania it was approximately 400 per 1000 [36]. The highest prevalence of FAS, between 460 and 680 per 1000, was reported in Russia in orphanages for children with developmental abnormalities [48]. Additionally, the prevalence of FASD among youth in correctional services was reported to be more than 230 per 1000 in Canada [69] and more than 140 per 1000 among psychiatric care populations in the United States [80].

DISCUSSION

This study demonstrates that the prevalence of FASD is highly variable, and disproportionately impacts some special subpopulations, and this is not unexpected given the context of the origin populations and the life-course of individuals with FASD. In general, children are often placed in care due to a number of unfavourable circumstances, such as parental alcohol and/or other drug problems, abuse and/or neglect, abandonment and young maternal age. These circumstances are associated with an increased probability that a child had been exposed to alcohol *in utero* [104]. If appropriate diagnosis, interventions and support services are not put in place early in life and maintained throughout their life, many youth and adults with FASD are at a high risk for becoming involved in the legal system, either as offenders or as victims. It was estimated that youth with FASD are 19 times more likely to be incarcerated than youth without FASD on any given day in a specific year [105]. Lastly, individuals with FASD are likely to suffer from developmental delay, learning problems and mental health problems [4]; therefore, a high prevalence among special education populations (e.g. in special schools for mentally handicapped children) and specialized clinical populations (e.g. in psychiatric care) is not surprising.

Several factors contribute to the prevalence of FASD in Aboriginal populations. For example, the prevalence of alcohol use during pregnancy in the Aboriginal populations of the United States and Canada were found to be approximately three to four times higher, respectively, compared to the general population [106]. Even more alarmingly, approximately 20% of women who consume alcohol during pregnancy engage in binge drinking in the Aboriginal populations compared to 3% in the general population in both countries [106]. The high prevalence of alcohol consumption and FASD in some Aboriginal populations must be understood within the historical and social context of colonization and the socio-demographic realities. Intergenerational impacts of colonial history, including trauma, residential school experiences and economic and social marginalization, contribute to alcohol use in Aboriginal communities [107,108].

While all these subpopulations share many risk markers, it is not clear whether FASD results in a common risk factor or impairment that increases risk for contact with certain service systems. It is also unclear whether the variation in the prevalence of FASD among the special subpopulations identified is due to differences in rates or patterns of prenatal alcohol exposure, dosimetry or increased susceptibility to alcohol exposure prenatally. Both missed diagnoses and underdiagnoses of FASD confound efforts to better understand these differences [54]. What is clear, however, is that exposure to alcohol prenatally that leads to a diagnosis of FASD has predictive implications with respect to adversity. In the past, it could be argued that we had insufficient information on FASD to make public policy recommendations. We now have convincing evidence that FASD is a relatively prevalent alcohol-related disorder that greatly increases the risk of long-term adversity. As such, public policy and clinical care for people with FASD needs to change to respond to such predictable outcomes. The data presented in this study have important implications for health-care providers, psychiatrists, psychologists, social workers, individuals working within the justice and child welfare systems, policymakers and, most importantly, for people affected with FASD and their families. These prevalence estimates are crucial for promoting early identification of FASD and provision of prevention and care interventions as well as for informing policymakers and service providers about the overall impact of FASD on population health. In addition, these prevalence estimates will help to generate policy and programme support for services required by people with FASD. Routine screening protocols should be established for identification of children, youth and adults in different settings such as child welfare, special education, justice system and others in order to provide them with appropriate support and early interventions. Service providers should be trained on FASD awareness, identification and interventions of people with higher risk for prenatal alcohol exposure and FASD.

There are several limitations in this study. First, FASD prevalence estimates were derived over an approximately 40-year time-span, so the prevalence of FASD, for example, in an American Indian community in the 1980s may not be relevant at all to current prevalence in that community, nor comparable to the prevalence in an aboriginal community in Australia captured 30 years later. Specifically, the majority of the studies reporting prevalence of FASD among Aboriginal populations in Canada are 2–3 decades old and suffer from many methodological limitations [90–92,94], and thus those existing data are not applicable for decision-making purposes and rigorous active case ascertainment studies are urgently needed in Canada. Further, outdated studies from Australia, which are based on PS, report an unrealistically low prevalence of FASD (lower or slightly above 1%) among Aboriginal populations [86,89]. However, a recent ACA study reported the prevalence of FASD among Aboriginal populations of Australia to be over 19% [85].

Further, existing studies suffer from variability in the quality and inconsistency in the methods used among them. Specifically, studies used 12 different diagnostic criteria to classify children or adults as FAS or FASD (all of which have substantial lack of overlap [109], not to mention that these studies had widely varying criteria for documenting quantity and frequency of alcohol consumption required. It is also possible that some prevalence studies were initiated due to the suspected high rate of FASD in these settings, demonstrated by an increased demand to service providers or increased health-care cost, which may lead to overestimated results.

There are multiple other special subpopulations impacted by increased rates of FASD—two examples are children whose mothers are in treatment for substance use disorder(s) and infants requiring neonatal intensive care. However, there are no studies that examined the prevalence of FASD in these special subpopulations. Further, 45 years after discovering FAS, we found that it was not possible to conduct meta-analyses among low socio-economic populations and specialized clinical populations due to insufficient data; thus, rigorous research is urgently needed to appreciate those populations most impacted by FASD.

It appears that prenatal alcohol exposure defines a high-risk population in need of long-term monitoring [110]. Our ability to develop enhanced care and monitoring of this high-risk population (individuals with FASD) is limited by the very low rates of diagnosis for all age groups. For adults, diagnosis is often limited by difficulty determining prenatal alcohol exposure status (especially in cases where the biological mother is unknown) and uncertainty about the adult phenotype of FASD. This is even more problematic in elderly people. For correctional populations in particular, the setting may also result in a limited diagnostic capacity for FASD. Providing FASD diagnoses is further limited by a lack of resources, an impacted health-care referral system and stigmatization of maternal alcohol consumption. In addition, current diagnostic guidelines have limited agreement [110,111]. Diagnostic screening and staff training on FASD in the respective systems/institutions are crucial in order to ensure that FASD-affected individuals are receiving the appropriate care and treatment.

The results indicate that there is a critical need for ACA prevalence studies to be conducted among these populations/within these service systems in almost all countries throughout the world. Measuring and monitoring the prevalence of FASD and alcohol consumption during pregnancy over time in both the general population and population subgroups are crucial for understanding and identifying vulnerable populations, targeting prevention and treatment resources and establishing baselines to evaluate the effectiveness and cost-effectiveness of prevention and treatment strategies. A comprehensive surveillance system could also allow for a better understanding of the associated morbidity and mortality rates, quality-of-life indicators and service utilization rates of affected individuals. This will reduce the risk of the development of other common adverse outcomes that often occur in individuals with FASD later in life, such as school failure and dropout, mental health problems, inappropriate sexual behaviour, alcohol and other drug problems, unemployment, dependent living and homelessness, as well as involvement with the law and incarceration [112].

Prenatal alcohol exposure is preventable through public health messaging and treatment of substance use disorder(s) in mothers. It is absolutely necessary to continue to improve prevention of alcohol consumption during pregnancy, screening strategies, targeted interventions for women of childbearing age with substance use problems, diagnosis-informed care and the provision of support for people with FASD and their families, especially in these special sub-populations.

Declaration of interests

None.

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Supporting Information

Additional supporting information may be found online in the Supporting Information section at the end of the article.

Appendix S1 PRISMA 2009 Checklist.
Appendix S2 Quality appraisal of the identified studies reporting on the prevalence of FASD among special sub-populations and reference list.
Appendix S3 Measures of heterogeneity and potential publication bias.

