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Fetal Alcohol Syndrome & Fetal Alcohol Spectrum Disorder Among Aboriginal Peoples

A REVIEW OF PREVALENCE

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1.0 Summary

In some cases, children born to women who consume alcohol during their pregnancy may have adverse, life-long outcomes. These outcomes include growth deficiencies, particular facial characteristics and a host of central nervous system complications, resulting in learning disabilities, poor socialization, or behavioural difficulties. These outcomes range along a continuum contained under the umbrella of Fetal Alcohol Spectrum Disorder (FASD) and includes Fetal Alcohol Syndrome (FAS).

As Tait (2003) suggests, a large proportion of Canadian research on FAS is focused on Aboriginal peoples and supports a commonly-held belief that substance abuse during pregnancy occurs more frequently among Aboriginal women compared to their non-Aboriginal counterparts. However, the true extent of FAS and FASD in Aboriginal and non-Aboriginal populations is not known and thus no assessment of higher prevalence is possible.

This report focuses exclusively on the published literature on FAS and FASD, and highlights a number of points:

» Published estimates of the prevalence and incidence of FASD and FAS are too methodologically diverse to provide the basis for Aboriginal-specific rates.

- There may be a substantial discord between the estimates available in the literature and the experiential knowledge of Aboriginal communities and clinicians.
- Some of the Canadian Aboriginal-specific published studies focus on higher-risk communities and may promote a perception of higher prevalence or incidence of FASD in the Aboriginal population.
- Because the Aboriginal population constitutes a relatively small proportion of the Canadian population, in absolute terms most cases of FAS/FASD are likely within the non-Aboriginal population.

2.0 Scope of this Report

This report provides an overview of the literature on the prevalence or incidence of Fetal Alcohol Spectrum Disorder (FASD) and Fetal Alcohol Syndrome (FAS) among Aboriginal peoples in Canada. Rather than being a systematic review, which involves a specific clinical research question and exhaustively searches, identifies, and summarizes the available evidence (Montori, Wilczynski, Morgan & Haynes, 2003), this overview is narrative. The rationale for a focus on prevalence was the necessity to begin with the basic epidemiological characteristics of FAS/FASD prior to further work, as well as the continued use of the published literature to substantiate policy and further research.

This is not a comprehensive review of all the available literature on FAS and FASD. It does not summarize the extensive laboratory research that has been conducted on FAS/FASD, since the focus is on prevalence and not necessarily of underlying causation. Additionally, there is a critical literature on FAS that is also not included in this discussion. Epidemiology is not well-equipped to provide explanations that incorporate historical precedent or unmeasurable dimensions of socio-economic status. The gendered nature of FAS, as a preventable outcome linked to women's behaviour, has raised concerns of a 'moral panic' over the intake of any alcohol during pregnancy. As Armstrong and Abel (2000) have argued, this may have diverted attention away from the social inequalities and displaced blame onto individual mothers rather than social circumstances. These concerns become even more relevant when the double jeopardy of gender and Aboriginality suggests that more work is needed on the 'other side' of the methodological bridge. Thus, there is also considerable scope for qualitative work on FAS, although such realms are outside of the scope of this report.

There are multiple audiences for this report. The focus is epidemiological, but for those readers who are not familiar with the language or the terminology of this research, explanation of terms is given where appropriate.

Locating Evidence

The studies described in this report were found through a number of sources, primarily Medline and Web of Science. Initially, the MeSH heading "fetal alcohol syndrome" was used to search Medline for articles relating to FAS. No MeSH term exists for fetal alcohol spectrum disorder. For FASD, an initial keyword search on all fields provided the starting point for the research. Given the large number of biomedical laboratory papers on the teratogenic effects of alcohol, including animal studies, further keywords were used to refine the search to population-based research. This included keywords relating to study design (case-control; cohort); specific geographic areas (Canada; United States); and/or Aboriginal populations (Aboriginal; First Nations; Inuit; Métis; and the term 'Indian', primarily to locate research conducted in the United States) and prevalence. At the suggestion of a reviewer, additional searches were completed including 'prenatal alcohol' and 'alcohol and pregnancy' as keywords.

References within extracted studies were also examined for further articles, and the 'cited reference search' function of the ISI Web of Knowledge database was used to find additional literature citing specific articles found in the first stages of research. 'Grey' literature from federal and provincial health authorities has also been included in this report, although its use has been restricted to background information. Because of the relatively small number of population-based studies on FAS/FASD, the historical window of this review is deeper than it might be for other summaries of clinical papers.

3.0 Prevalence

Since the true prevalence of FASD in the population, Aboriginal or otherwise, is not known, there are a number of paradoxes that occur in reported rates. For example, in the American context, American women have substantially lower rates of alcohol consumption relative to the rest of the world, yet the incidence of Fetal Alcohol Syndrome (FAS) in the United States may be the highest in the world (Abel, 1998a). There is another paradox related to FAS, however. On the one hand, there is widespread recognition among Aboriginal communities and the population health community in Canada that FAS is prevalent and that it represents a serious health threat in many Aboriginal communities (Stout, Kipling & Stout, 2001).[1] Inuit women, in particular, have identified the need for prevalence studies on FAS, along with locally-developed education and prevention materials as a priority health information need (Stout, et al., 2001). On the other hand, the epidemiological evidence is inconclusive not only about the incidence of FAS and the broader umbrella of Fetal Alcohol Spectrum Disorder (FASD) in Aboriginal communities, but also the Canadian population as a whole. Thus, there is an "epidemic of FAS" (Robinson, 1992) and, because of the lack of firm comparative data on the non-native population, a conflicting view that higher prevalence among native peoples is impossible to determine (MacMillan, MacMillan, Offord & Dingle, 1996).

It is likely that both situations are, to some degree, correct. The anecdotal evidence of FAS about the extent of the problem likely reflects an underlying, uncounted prevalence. In an epidemiological context, however, evidence is only available from studies carefully designed to minimize bias or from administrative data that are of known quality. Direct knowledge of FAS fails the epidemiologic criteria; epidemiologic knowledge fails experiential knowledge. Some reconciliation of these forms of knowledge can take place, but it requires more research in both domains before the body of evidence becomes clear and unimpeachable.

This review focuses on the available academic literature on FAS and FASD among Aboriginal peoples in Canada. Beginning with an overview of currently-accepted definitions, the review then moves to diagnostic criteria; basics of incidence and prevalence and difficulties in the measurement of FAS and FASD; prevalence in the general population; and an overview of prevalence in Aboriginal populations. Estimates of the extent of FAS/FASD and the economic costs round out the review.

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1 'Community' is used in this report to refer to people in the aggregate. In reference to Aboriginal populations, there are a number of ways that 'community' can be used. It may refer to a specific First Nation, Inuit or Métis community or it may also be applied to sub-groups within the Aboriginal population in which there may be distinct differences in the nature or extent of FAS/FASD. For instance, there may be substantial differences between urban and rural Aboriginal populations in prevalence of FAS/FASD, although this has not been widely explored. In general, 'community' in this report refers to a First Nation. It should be noted that Aboriginal is a term encompassing not only First Nations but also Métis and Inuit peoples who face many of the same issues regarding FASD.

Definitions

The basic definitions of some terms used in this report are shown in Table 1. Of these, the current Canadian diagnostic criteria (described in more detail in a later section) includes FASD, FAS, ARND and p-FAS. Fetal Alcohol Effects (FAE) and Alcohol- Related Birth Defects (ARBDs) are included because some earlier studies make reference to these conditions.

Table 1. Definitions of Some Terms Used in This Report

Fetal Alcohol Spectrum Disorder (FASD)	FASD is an umbrella term encompassing the range of effects that can occur to an individual whose mother drank alcohol during pregnancy (Chudley, Conry, Cook, Loock, Rosales & LeBlanc, 2005). It is not a clinical diagnosis by itself.
Fetal Alcohol Syndrome (FAS)	FAS falls within the umbrella of FASD. The most recent Canadian diagnostic criteria for FAS require confirmed exposure to alcohol during pregnancy and three broad anomalies: pre-or-postnatal growth retardation; dysmorphic characteristics, including a distinct facial appearance; and some evidence of central nervous system (CNS) impairment (Chudley, et. al., 2005). Although various diagnostic criteria have been developed to better quantify these relationships, the basic components of FAS have not changed since the first criteria were developed in the 1970's (Riley & McGee, 2005).
Partial FAS (p-FAS)	p-FAS diagnoses require the same CNS impairments as FAS, but there are no criteria for growth impairment and fewer facial anomalies need be identified.
Alcohol-Related Neurodevelopmental Disorder (ARND)	ARND narrows the list of criteria further, requiring similar CNS impairments to FAS and p-FAS and confirmed maternal exposure to alcohol but without the growth impairment or facial anomalies (Chudley, et. al., 2005). Diagnostic criteria for FAS, p-FAS and ARND are described in more detail below and included in Appendix A.

Fetal Alcohol Effects (FAE)	FAE is a less 'complete expression' of FAS. In one working definition, a person having two of growth deficiencies, facial dysmorphology or central nervous system dysfunction (Spohr, Willms & Steinhausen, 2007). The term FAE has been criticized as inappropriately implying a causal link between exposure and outcome and often poorly defined (Sampson, et al., 1997).
Alcohol-related birth defects (ARBD)	ARBDs generally refer to clinical conditions where clinical or animal research has linked maternal consumption of alcohol and an observed outcome and there is a history of exposure (Chudley, et al., 2005). While ARND refers to CNS or behavioural abnormalities, ARBDs are physical outcomes. ARBD does not, however, constitute a diagnostic category by itself in the Canadian FASD guidelines.

The first suggestion of case reports of a cluster of birth defects associated with in utero exposure to alcohol was presented by Lemoine in 1968 (Lemoine, 2003) and subsequently by Jones, Smith, Ulleland & Streissguth in 1973. Lemoine's paper was seminal and contained substantially more cases (n = 127), but was published in the French literature and did not receive wide attention. It also drew few conclusions about the observed syndrome and did not present diagnostic criteria (Hoyme, et al., 2005). The paper by Jones et al. summarized case histories of eight children of alcoholic mothers living in Seattle, Washington who presented a similar pattern of effects including craniofacial, limb and cardiovascular defects linked to prenatal-onset growth deficiency and developmental delay.

Binge drinking appears to have an important role in the development of FASD. In a secondary study by Barr and Streissguth (2001) linked to a larger longitudinal study involving 1,439 singleton births in Washington State during 1974-75, 38.4% of women who reported drinking five or more alcoholic drinks per month and binge drinking (n = 73) had FASD children (n = 28). Women who reported daily or near daily drinking without binging (n= 99) had smaller proportions of FASD children (n = 8; 8.1%), suggesting that binge drinking as well as total volume may be implicated in FAS. There is debate, however, over whether 'safe' levels of alcohol consumption exist.[2] In a meta-analysis of twenty-four studies, Polygenis, Wharton, Malmberg, Sherman, Kennedy, Koren, and Einarson (1998) found no evidence of increased fetal malformations with moderate (greater than two drinks per week to a maximum of two per day) alcohol consumption and a broad group of physical malformations at birth. The

2 Medical practitioners, for example, face conflicting advice about whether moderate or low consumption is harmful. In a survey of obstetrical textbooks published after 1990, 24% consistently recommended abstinence during pregnancy, and 52% were contradictory in that they condoned some level of consumption in sections while advocating abstinence in others (Loop, K,. & Nettleman, M. (2002), cited in Clarke et al., 2005).

odds ratio for malformations among moderate alcohol users was 1.01 (95% CI: 0.94 – 1.08). Malformations at birth do not equate to FAS, but these results are suggestive of a potential threshold for alcohol, after which the dose-response relationship takes effect. There is some evidence of a dose-response relationship without threshold for neurobehavioural outcomes. Rather than using FAE or FAS diagnosis as the key variable, Sampson, Streissuth, Bookstein and Barr (2000) examined the continuous relationship between a variety of exposure measures and outcomes using data from the longitudinal Seattle, Washington study. In their analysis, average daily volume of alcohol shows the greatest variation, but for average drinks per occasion, the results consistently show steadily decreasing neurobehavioural outcomes with increased consumption.

For infant mental development, a similar meta-analysis by Testa, Quigley & Eiden (2003) found a linear negative effect between level of exposure and Mental Development Index scores on 12-13 month old infants, but not at other ages. The magnitude of effects across studies decreased when adjusted for covariates, which varied according to the studies included. From the author's perspective, the most striking conclusion from this meta-analysis is that the body of relevant research is neither as large nor as conclusive as might be expected, particularly in light of the effect that the literature on drinking during pregnancy has had on social policy and norms.

Diagnostic Criteria

Appendix 1 presents the set of suggested diagnostic criteria recently proposed by a subcommittee of the Public Health Agency of Canada's National Advisory Committee on Fetal Alcohol Spectrum Disorder (Chudley, et al., 2005). Separate sections are included for FAS, p-FAS and ARND. Under these criteria, FAS is characterised by maternal exposure to alcohol and, in the absence of any other diagnoses:

- A distinct dysmorphology, or set of physical abnormalities associated with FAS, including facial features such as a flattened area between the upper lip and nose (philtrum), thin upper lip, epicanthal folds, and a narrow palpebral fissure, or the length of the space between the margins of the eyelids;
- Central nervous system (CNS) effects, expressed as a particular set of potential behaviours described in more detail in the Appendix;
- and evidence of pre-or-post natal growth impairment, with either birth weight or length; current height or weight; or weight-to-height ratios at or below the 10th percentile for age.

Partial FAS is similar to the previous definition for full FAS with fewer specific facial anomalies and no growth impairment criteria. As with FAS and p-FAS, a diagnosis of

Alcohol-related Neurodevelopmental Disorder (ARND) requires the presence of three CNS deficits as described in Appendix A, but without the dysmorphological or growth impairments. Although alcohol-related birth defects (ARBD) has also been used to describe a wide variety of outcomes related to prenatal alcohol exposure, these guidelines suggest that it should be used with caution and not used as an umbrella or diagnostic term.

These criteria are similar, but not identical, to other criteria developed for the purpose of identifying individuals with FASD. In the United States, the Institute of Medicine (IOM) published diagnostic guidelines for children with FAS and alcohol-related effects in 1996 (Stratton, Howe & Battaglia, 1996). These guidelines, however, are not specific enough to ensure diagnostic accuracy and include categories that do not require confirmed maternal consumption of alcohol. While they do contain the broad elements of FAS, they do not include specific, quantitative benchmarks for its assessment. To address this, Astley and Clarren (2000) published a set of criteria using 4-digit diagnostic codes as an aid to clinicians. These criteria have been incorporated into the most recent Centers for Disease Control (2005) guidelines for identification of persons with FAS,[3] and a harmonized version is promoted in the 2005 Canadian guidelines.

The diagnostic criteria for FASD-related conditions have substantial ambiguities that may potentially lead to differing assessments depending upon the population studied, and the time period in which the study takes place. Although full FAS may be at the more severe end of the continuum of potential effects emanating from maternal alcohol consumption (Canadian Perinatal Surveillance System, 1998), the growth and CNS effects likely have greater costs to the individual or to their communities. From an Aboriginal perspective, the facial characteristics associated with FAS may not be applicable (Bray & Anderson, 1989). While the phenotype may be applicable to Caucasian and African-American populations, no standards exist for other populations (Chudley, et al., 2005). Partial presentations of FAS without these markers are, however, more difficult to classify and individuals not diagnosed with FAS as a result may be at increased risk for inadequate care (Astley, 1997).

Although the basic characteristics of FAS are delineated in diagnostic criteria such as those indicated above, certain features of FAS/FASD lead to ambiguously broad definitions. In part, this is because the features associated with FAS, including facial, growth, and CNS outcomes, vary over the normal course of development (Abel, 1998b). Facial characteristics may also be similar to facial ethno-specific phenotypes, which may result in over-estimates of prevalence in some populations (Abel, 1995). Behavioural characteristics may not be apparent at birth and may develop gradually from infancy and into the first few years of grade school (Aase,

3 The CDC guidelines do not, however, note that the typical facial dysmorphia associated with FAS may not be applicable to Aboriginal populations. Instead, the diagnosis criteria suggest that the clinician make assessments based "on racial norms (i.e. those appropriate for a person's race)" without discussion about what those norms may be (CDC, 2005).

1994). Additionally, guidelines for FASD rely on the skill of the observer and may be subject to bias. Use of diagnostic coding schemes may, however, help to reduce inter-rater variability.

Construct validity and reliability are essential components of study design. Validity in this context is the extent to which an instrument measures what it purports to measure; reliability is the degree to which results of a measurement procedure can be replicated (Kelsey, Whittemore, Evans & Thompson, 1996). In the case of FAS, Abel (1998a) has noted that no studies of the reliability of FAS diagnostic paradigms have been published. At the time of this writing, only one paper appears to test the reliability of FAS diagnoses.

In the initial presentation of their 4-digit FASD classification criteria, Astley and Clarren (2000) re-tested 454 patients of a University of Washington FAS clinic. Initially, all subjects had been classified according to qualitative assessments based on morphological, growth and CNS criteria – a 'gestalt'[4] approach rather than a systematic one. In comparison, the same individuals were re-assessed using the 4-digit diagnostic code. This code reflects the magnitude of expression of growth deficiency, exposure to alcohol during gestation, brain dysfunction, and the typical FAS facial features. In the original data, 69 individuals were diagnosed with full FAS; in the re-test, eleven individuals met the criteria for full FAS under the 4-digit code. Possible fetal alcohol effects, a substantially broader category characterized primarily by no growth deficiency, mild facial phenotype and 'possible' brain damage, was captured similarly between the two diagnostic criteria (gestalt = 344; 4-digit = 365) (Astley & Clarren, 2000).

In a test of internal reliability, Astley and Clarren (2000) re-assessed 20 randomly selected case histories from the group. Each of these cases had previously been assessed using the 4-digit scale. With no knowledge of the original coding, the authors replicated the original coding for all 20 subjects. In this instance, although both criteria shared broad categories, there was little reproducibility between the two measures. Although the 4-digit code is internally reliable, the teams involved in the test-retest were directly involved in the development of the 4-digit code. Further reliability tests involving outside clinicians are necessary. Furthermore, the study does not validate the 4-digit system. While it may be more internally consistent, it is difficult to evaluate whether it truly captures FAS more completely than the less-structured approach.

Hoyme, et al. (2005) note that the neurobehavioural and encephalopathic characteristics of the 4-digit code are not specifically defined in these criteria, and are not unique to the prenatal effects of alcohol on fetal development. Family and genetic backgrounds are also not adequately incorporated into these criteria. Hoyme et al. found that the possibility for false

4 Astley and Clarren and Abel (1998) use diagnostic gestalt in the sense of the sum being greater than the whole of its parts; as Abel (1998) explains, each part on its own may be slightly unusual, but it is only when meshed together that these disparate elements take on the shape of a syndrome.

positives under this system is greater, since any child with a disability who has been exposed to alcohol prenatally may be assigned a diagnostic classification, even if the cause of the disability is genetic (Hoyme, 2005).

In practice, physicians may feel qualified to assess FASD but may vary in their ability or understanding of diagnostic criteria. Clarke, Tough, Hicks and Clarren (2005) surveyed medical practitioners in Canada to assess their understanding and attitudes towards the diagnosis of FASD. The authors randomly selected a sample of family physicians (n = 2,378), paediatricians (1,396), psychiatrists (1,439), midwives (197), and obstetricians (539). The response rate for their mailed questionnaire was low (41.3%).[5] Other than midwives, the majority (75%) of medical practitioners felt that diagnoses of FAS or FAE were within the scope of their practice, and a substantial majority (94%) felt that FAS is an identifiable syndrome. A small majority (56.4%) felt that a lack of training was a barrier to diagnosis. A majority also felt that diagnostic supports, including specialists available for consultation (61.8%), and clinical practice guidelines for FAS (60.8%) would be helpful. Less than 60% of the respondents in this study recognized that the combination of growth, brain and facial abnormalities provided the most accurate assessment of FAS.

Although there is consistency in terms of the components of the syndrome, small variations in definitions may lead to substantial over-and-under-ascertainment of cases, which in turn affect estimates of prevalence of FAS. Research studies may have a greater degree of control over how criteria are applied. For Aboriginal peoples, diagnostic criteria may have to be revised if misclassification results from the application of phenotypical criteria not validated for these populations. Although not widely discussed in the literature, validation of the CNS criteria for Aboriginal populations may also be important.

Incidence and Prevalence

Incidence and prevalence are basic epidemiological building blocks. Incidence refers to the occurrence of new disease within a particular time frame. Dividing the number of new occurrences in, for example, a year by the population-at-risk for the condition results in an incidence rate. Prevalence captures the proportion of people at risk who have the condition, including both newly-developed and pre-existing cases. Like incidence, prevalence is expressed as a rate (Rothman & Greenland, 1998).

The distinction made here between incidence and prevalence may be somewhat artefactual in the context of FAS/FASD. Rothman and Greenland (1998) argue that the

5 The potential bias may be that non-respondents may have different attitudes about the importance of FAS/FAE, their scope of practice, or knowledge of diagnostic criteria.

occurrence of congenital malformations cannot be counted as incidence since the reference point is birth. They are not 'new cases' streaming into a population, since the population-at-risk is newborns. Unless incidence changes over time, rates of both prevalence and incidence will be similar for congenital conditions. In the case of FAS/FASD, it is likely that increased public awareness of the effects of consuming alcohol during pregnancy may lead to reductions over time, but as of yet, there is little direct evidence of this. For FAS/FASD, the rest of the discussion will refer to such rates as prevalence unless explicitly described in the literature as incidence.

A number of study designs have been used to estimate rates of FAS and FASD. Passive surveillance studies may use registry or administrative data sources to identify cases. A second form of study design is clinic (or hospital) based, which may be retrospective or prospective and rely on attendance at specialized clinics or hospital admissions for case ascertainment. Thirdly, population-based studies may focus on a particular community or population and actively seek cases through screening or other means. May and Gossage (2001) have found that, when reviewing prevalence estimates by study design, passive surveillance studies consistently report lower rates of FAS, particularly in comparison to active studies targeting known higher-risk populations. In the studies examined by May and Gossage, FAS rates produced by passive studies range from 0.26 to 2.29 per 1,000 live births versus 1.4 to 9.8 per 1,000 for active case ascertainment studies.

Difficulties in Estimating Prevalence

In principle, prevalence rates rely on two pieces of information: a count of cases, and a denominator of the population-at-risk. In practice, estimating prevalence is hampered by difficulty in defining cases and inherent biases in study designs. The following list of points is not comprehensive, but includes issues that have been identified in the literature regarding the estimation of FAS/FASD rates in particular. Some issues are more general, while others relate specifically to FAS/FASD prevalence in Aboriginal populations.

CASE ASCERTAINMENT (OVER AND UNDER-ESTIMATION): The issue of how FAS and FASD are defined are crucial in estimating incidence. As the comparison of gestalt and 4-digit code in Astley and Clarren (2000) suggests, wide variation in the number of children diagnosed with FAS/FASD can result from different methods of inclusion. In the case of FASD, the range of possible effects may result in some true cases never being diagnosed with the condition. In some instances, over-estimates of FAS may result from clinical ambiguity, in that there is some evidence that clinicians may interpret the signs of FAS irrespective of maternal exposure to alcohol.

MISSCLASSIFICATION OF EXPOSURE AND RISK: Under the CMAJ guidelines described above, FAS can only be diagnosed with confirmed maternal consumption of alcohol during pregnancy. Mothers may not readily provide this information, particularly if they are aware of social stigma associated with drinking during pregnancy, which will undercount incidence. Biomarkers for exposure to alcohol in infants are under development, but ethical issues surrounding screening at birth and potentially different levels of consent for such testing among women who may have consumed alcohol during pregnancy may bias results by also underestimating exposure. Misclassification may also be greater in ARND studies because the associated effects are not specific to FAS and may be related to other exposures. As a reference, the full text of the CMAJ guidelines lists nine other conditions that may overlap with FAS in terms of their clinical presentation. Although none are identical to FAS, clinical expertise is required to differentiate these conditions from FAS.

DENOMINATORS OR POPULATION AT RISK (OVER AND UNDER-ESTIMATION): Studies that focus on particular geographic populations may not generate prevalence rates that can be generalized to broader populations. This is an issue particularly for First Nation-specific estimates, since a number of studies use specific Reserves as the focus of their research. Individual Reserves are not likely representative of the First Nations population as a whole and may have substantially different experiences with alcohol consumption and socio-economic status, which may be associated with maternal intake of alcohol. Thus, application of these rates to the general Aboriginal population may over-estimate the risk of FAS.

SOURCE OF DATA: Although some studies have used registry or administrative records retrospectively to study FAS children, such studies cannot be used to estimate true incidence in a population (Fox & Druschel, 2003). Specialized registries may not be representative of the population and, as previously discussed, may underestimate rates relative to population-based studies.

TIMING OF CASE ASCERTAINMENT: While birth may be a convenient time to identify cases, only the most severely-affected children will be diagnosed with FAS. Rates based on birth data will thus underestimate the degree of FAS in a population. In the general population, FAS is most readily identified in children between the ages of two and eleven (Fox & Druschel, 2003). In the FASSNet system (described later in this report), the large majority of children in the 1995 to 1997 cohort were not of school-age at the time of publication. Both CNS effects and learning difficulties become more evident once children enter school,

so timing studies too early may result in under-counts of cases (Center for Disease Control, 2002). Conversely, there is some evidence that the phenotypical criteria for FAS/FASD become less pronounced as children age, particularly past puberty. Studies restricted to older children may potentially introduce some misclassification of exposure because of the difficulties of maternal recall of alcohol intake during pregnancy.

SAMPLE SIZE: estimates of FAS are sensitive to the relatively small sample sizes of some studies. Some studies, based on registry or administrative data, including Lumley (1985); Sokol (1980); and Sokol (1993) (all cited in Abel, 1995) have had sample sizes exceeding 10,000.[6] Others, however, have relied on much smaller numbers of cases, which becomes problematic as FAS and FASD are still relatively rare events under most circumstances. In one of the studies cited by Sampson, et al. (1997), a Boston inner-city hospital examined 322 infants. Of the 58 infants exposed to alcohol in utero, 42 were examined. None of the infants were diagnosed with FAS, and thus the incidence was zero. After one year, one child in the study was found to have FAS, and thus incidence rose to 3.1 per 1,000 (Sampson, et. al., 1997). In studies with small sample sizes, the emergence of one case can have a substantial effect on estimates, although confidence intervals on estimates will reflect the underlying sample size.

PUBLICATION AND GEOGRAPHIC BIASES, AND THE ASSUMPTION OF HOMOGENEITY: Some studies of FAS in Canada have concentrated on Aboriginal communities where there is a clear public health concern stemming from knowledge of potentially high FAS rates. The result is that higher prevalences from these studies might be misinterpreted as representative of Aboriginal communities in general when in reality there is substantial variation and diversity.

Additionally, there is a geographic bias evident in Canadian Aboriginal FAS studies in the published literature. There do not appear to be many, if any, published studies of Aboriginal communities outside of the Western or Prairie provinces. From a cultural perspective, the lack of research in Eastern Canada may mean that many Nations and cultural groups, including Algonquin, Ojibway, Micmac and Haudenosaunee, are under-represented in broad estimates of FAS and FASD available in the medical literature.

6 Note that these studies do not derive estimates from population screening but by review of administrative data. Thus, the sample size, while large, may be less sensitive to prevalent cases compared to proactive case ascertainment.

Prevalence of FAS: General Population

The worldwide incidence of FAS was estimated by Abel (1995) as 0.97 cases per 1000 live births.[7] This estimate is based on pooling the results of 29 international studies published between 1977 and 1994. There are clear differences between the countries in which these studies took place. Among American studies, the rate of FAS was 1.95 per 1000 live births, compared to 0.08 per 1,000 for other countries. Abel's overview does not include details on the design of the studies included. For example, Tait (2003) has noted that the study by Sokol et al., included in Abel's (2005) review, did not provide specific diagnostic criteria nor detailed information on alcohol exposure. Abel also does not provide assessment of the variance of the individual estimates, or meta-analytical techniques beyond estimates of average, median and modal rates for the pooled studies.

A more complex assessment of FAS and FASD prevalence was undertaken by Sampson et al. (1997), who critiqued previous FAS studies on the basis of case ascertainment and study design. Only two American studies fit their criteria for determining proper prevalence. Both are prospective cohort studies, where a population of prenatal patients was screened and followed until birth outcomes could be assessed. Participants included a group of exposed infants and a group of unexposed infants. Clinicians were blinded to exposure status, and assessed infants soon after birth for indications of FAS. One of these studies, retained because of its large sample size and extensive neurobehavioural data, collected data from a Seattle, Washington hospital over a one year period. Of the 1,439 births, two were diagnosed with FAS, for an incidence of ~1.4/1,000 live births. Because not all exposed children were screened, Sampson et al. calculated an adjusted rate controlling for incomplete screening of 2.8/1,000. By the age of seven, five waves of psychometric assessments were given to 581 of these children with various levels of exposure derived from the initial study. The psychometric assessments included attention, neuromotor, mental and learning disability domains. From the original data, thirteen levels of exposure were defined based on timing and amount of alcohol consumption of the mothers. With these data, the authors were able to link dose and outcomes from birth through to seven years of age. Children who showed continuous deficits in the outcome measures across the period in combination with high alcohol exposure were regarded as "true ARND" cases. The adjusted rate based on these 12 cases is 8.3/1,000 (12/1439).

Fox and Druschel (2003) compared rates derived from different registry systems by comparing New York State's passive birth defects registry to a registry designed specifically to capture new cases of FAS. The FAS registry, the Fetal Alcohol Syndrome Surveillance Network (FASSNet) was a population-based system where children with FAS or suspected

7 To calculate this rate, Abel summed the observed cases of FAS (95) divided by the total sample size. The denominator in Abel's published table is 97,576, but when summed, the actual data from the table totals 97,536. Use of either denominator leads to the same rounded estimate of 0.97 per 1,000 live births.

prenatal exposure to alcohol were actively identified from diagnostic and service programs in the state. By contrast, the New York State Congenital Malformation Registry (CMR) is a mandatory reporting system where children with birth defects, including FAS, are reported to the system up to the age of two. Case definition for FASSNet is based on the Institute of Medicine's criteria, while the birth CMR uses the non-specific International Classification of Diseases version 9 (ICD-9) code 760.71.[8] The use of these codes in the CMR data may explain to some degree why these data resulted in a substantial number of false positives when these records were compared to FASSNet data (14/33 = 42%). In total, Fox and Duschel found that the 33 cases of FAS in the CMR during the study period from 1995 to 1998 were substantially fewer than the 57 cases captured by FASSNet. Using birth data from the same period, the estimated prevalence ranged from 0.28/1,000 for CMR versus 0.37/1,000 for FASSNet data.

The use of non-specific ICD-9 codes in earlier studies may be problematic for precise case definition. In the early 1980's, the American Center for Disease Control studied abstracts of cases classified under ICD-9 760.71 in eight native communities in Iowa, Nebraska, North Dakota and South Dakota (Center for Disease Control, 1995). In their summary of those abstracts, 74 of 251 identified cases (29.5%) had no information on prenatal alcohol exposure or maternal history of alcohol consumption in their records. The CDC suggests that only a small proportion of cases identified in this period using ICD-9 codes met a rigorous definition of FAS, but recommended continued surveillance using the method since it likely captures more general patterns of adverse effects of maternal alcohol consumption during pregnancy.

Washington State is one of the few jurisdictions to have tracked maternal consumption of alcohol, and, in the same period, collected data on the prevalence of FAS. The Pregnancy Risk Assessment Monitoring System (PRAMS) is an initiative of the Centers for Disease Control focusing on maternal attitudes before, during, and after pregnancy. Since its development in 1987, PRAMS had extended to 29 states and New York City by 2005. Astley (2004) has reported that for Washington State, consumption of alcohol in the three months prior to pregnancy and in the third trimester declined significantly ($p < 0.001$) between 1993 and 1998. Astley correlates this decline with a similar significant reduction in the prevalence of FAS in a screened population of children entering foster care in Kings County, Washington State. Ascertainment within this population is near-complete in this county,

8 ICD-9 and its more recent replacement ICD-10 are standardized coding schemes for diseases and treatments. ICD-10, which is a more complex revision of the earlier standard, contains two alcohol/birth-related codes: P04.3 [Fetus and newborn affected by maternal use of alcohol], excluding fetal alcohol syndrome; and Q86.0 [Fetal Alcohol Syndrome (dysmorphic)]. Kvinge, Leonardson, Neff-Smith, Brock, Brozelleca & Welty (2004) also used ICD-10 760.71 to initially select cases for a study in Northern Plains Indian communities. Of the 142 such cases, only 43 (30%) met a more stringent definition of FAS.

In Ontario, for example, hospitalization and ambulatory care data are abstracted with ICD-10-CA (a Canadian revision of ICD-10). Some sense of the distribution of FAS can be gleaned from these records, but since they only include ambulatory and inpatient records they will underestimate prevalence substantially. Primary care visits to physicians in Ontario are coded with OHIP-specific diagnostic codes that do not include FAS or FASD.

but the actual number of cases used to determine the reduction in FAS is small. Over the six years of the study, 264 children were screened and five cases of FAS were identified in these children using FAS facial photographic analysis software and the 4-digit diagnostic code. These five cases form the basis for Astley's assessment of a declining trend over six years. Other difficulties lie with the PRAMS data. Since it is based on self reports, there is likely potential for under-reporting of certain behaviours, particularly those with strong social stigma. If the awareness of the effects of exposure to alcohol during pregnancy has become more widespread, we might expect reporting of this behaviour to decline irrespective of true declines in behaviour. Additionally, some women at very high risk for consuming alcohol during pregnancy may not have fixed addresses or may have higher rates of mobility, reducing the representativeness of respondents.

4.0 Prevalence in Aboriginal Populations

This section provides a summary of prevalence estimates of FAS and/or FASD that have appeared in the literature and refer to Aboriginal populations. These studies are summarized in Table 4. Both Canadian and American studies are included in this section. Other reviews that provide comprehensive overviews of the epidemiology of FAS among Aboriginal populations and the methodological limitations of these studies include Burd and Moffat (1994) and Bray and Anderson (1989). Burd and Moffat's article also contains information on two unpublished studies referred to below. Methodological limitations of these studies cannot be ascertained beyond the description provided by Burd and Moffat. Note that a number of studies use the term Fetal Alcohol Effects (FAE) to refer to more subtle expressions of FASD.

Generally, urban and clinic-based studies may result in undercounts of true cases. On the other hand, studies focussing on small communities with high risk may screen near-complete populations, potentially leading to higher observed rates of FAS, as in the study by Robinson, Conry & Conry (1987).

Sandor, Smith, MacLeod, Tredwell, Wood & Newman (1981) examined 76 diagnosed cases of FAS in 1981. Participants were observed in Vancouver hospitals and were drawn from British Columbia and the Yukon. Of the 76 cases, 69 were of Aboriginal descent and seven were non-Aboriginal. Although not able to determine rates of FAS/FAE directly, they estimate rates to be between 1 and 5 per 1,000 births. Based on the higher proportion of Aboriginal children with FAS, the authors suggest a possible, but untested, racial susceptibility for the teratogenic effects of alcohol, although no control for socio-economic status was included.

May, Hymbaugh, Aase and Samet (1983) studied Native communities in the southwestern United States in the early 1980's. Rates of FAS/FAE varied over time, between tribes, and between cultural groups. Between 1969 and 1982, rates of FAS/FAE among southwestern Indians (from birth to 14 years) ranged from 2.5 and 2.7 per 1,000 births for Navajo and Pueblo cultures respectively to 19.5 for Southwest Plains (including Apache and Ute) cultural groups. These findings suggest that a blanket assessment of greater prevalence among Aboriginal peoples does not take into account large potential differences between groups. May et al. (1983) suggest that the differences in rates are not only the result of differences in alcohol consumption but also to differences in social regulation between these groups. They also report multiple parity of women with FAS children; 85 of the affected children were born to 65 mothers, a finding that has also been reported in other studies. Based on 300 case reports where the status of siblings was available, Abel (1998) found that 27 out of 35 younger siblings also had FAS, a rate of 771 out of 1000.

Robinson, Conry and Conry (1987) reported on a 1984 survey of a British Columbia First Nations reserve in Canim Lake. This community had 350 members. Of the 123 children aged 18 or less living in the community, 116 participated in the study. Twenty-two (18.9%) of these children were given a preliminary diagnosis of FAS or FAE, which was ascertained prior to knowledge of maternal alcohol consumption. Prevalence for this community for FAS/FAE combined was estimated at 190 per 1,000 live births. Eight children were diagnosed with FAS (prevalence = 69 per 1,000) and 14 with FAE (prevalence = 121 per 1,000). Of the forty-five birth mothers of these children, 14 had given birth to one or more of the children identified with FAS/FAE; 12 of these children (54%) had been born to five women.

An unpublished study by Asante and Nelms-Matzke in 1985, cited in Bray and Anderson (1989), found 82 cases of FAS and 94 of FAE within a group of 781 Yukon and north-western British Columbia children aged 0 to 16 referred for assessment. Of these children, 586 were Native and 195 non-Native. Among Aboriginal children in the Yukon, the rate of FAS was estimated at 46 per 1,000, substantially greater than the 0.4 per 1,000 for non-Aboriginal children. A FAE rate of 26 per 1,000 was also estimated for Aboriginal children in north-western British Columbia. Since the children were identified as chronically handicapped prior to the study, the rates likely do not reflect the population as a whole (Burd & Moffat, 1994).[9] No controls for socio-economic status or data on the degree of maternal drinking are included in this study.

Kvinge, Leonardson, Neff-Smith, Brock, Brozelleca and Welty (2004) studied the characteristics of FAS-identified children in four Northern Plain Indian Health Services hospitals or clinics in the mid-western United States. In two separate retrospective case-control studies, Kvinge et al. used medical records to compare clinical features and hospitalizations of FAS/non-FAS children (study 1), and partial FAS/non-FAS children (study 2) born between 1981 and 1993. The inclusion criteria for FAS were confirmed prenatal alcohol consumption, FAS diagnosed by a physician, growth deficiency, and CNS impairment. Children meeting the broad definition of one to four of these criteria were classified as having incomplete FAS. In each study, cases were matched with two controls, although the selection criteria for the controls are not mentioned in the paper. This study is particularly relevant because the unaffected controls are also from the Northern Plains cultural group, and thus the differences in the facial traits associated with FAS children can be compared to a non-Caucasian 'norm.' All dysmorphic facial features occurred significantly more often in cases than in controls, in particular low nasal bridges (OR=30.0; 95% CI: 4.62-1263). The largest substantial difference in CNS dysfunction between cases and controls was developmental delay, which was evident in 74.4% of cases and 2.3% of controls (OR=122.1; 95% CI: 22.97-872.53). For both FAS and partial FAS, Kvinge et al. found that gross and

9 The rate of FAS is 77% higher than it is for FAE, which is the reverse of what one would expect, providing a further indication of potential bias.

fine motor developmental delays and hospitalizations were significantly higher for FAS and incomplete FAS cases than controls.

Between 1973 and 1993, 207 cases of FAS were identified in a study by Habbick, et al. (1996) in the province of Saskatchewan. Of these, 178 cases were Aboriginal. Over the twenty-year period, the rate of FAS was consistent, averaging 0.59 per 1,000 live births. In all cases, maternal use of alcohol during pregnancy was confirmed.

Egeland, Perham-Hester, Gessner, Ingle, Berner and Middaugh et al. (2003) undertook a comprehensive study of Alaskan FAS between 1977 and 1982. The case definition required FAS suspected or diagnosed by a physician, prenatal alcohol exposure or a maternal history of alcohol abuse, characteristics of fetal alcohol syndrome facial features, growth deficiency, and central nervous system impairment. Using a variety of sources to identify potential cases, including hospitals, pediatricians, Alaskan state programs, rural nursing stations, health services case files, administrative data on Medicaid claims, birth and death certificates, and Native health services, Egeland et al. assembled a large cohort (n= 630) of potential FAS cases. Of these cases, 90% (568) had medical charts, and prenatal alcohol exposure was confirmed in 462 (81%). For 248 (44%) individuals, diagnosed or suspected FAS was noted in their medical charts; 145 met all five of the criteria in the case definition.

Table 2 shows the prevalence rates estimated from the study by Egeland et al. for Aboriginals and non-Aboriginals by period, using the total number of live births in Alaska between 1977 and 1982, when the majority of cases identified were born as the denominator. The table differentiates between the rates of individuals who had notations of FAS in their medical records ("FAS Noted") in addition to the rate of meeting all five of the diagnostic criteria ("FAS Cases"). Egeland et al. note that the greater number of cases identified in the Aboriginal population may in part be due to the active case-finding on the part of the Indian Health Services, as well as corresponding under-ascertainment in the non-Native population. Not expressly discussed by Egeland et al. is the proportionally larger difference between 'noted' and 'cases' for Native persons, suggesting that there may be elevated pre-identification of children having suspected FAS for Native Alaskan children.[10]

10 For Native persons, total cases over the period totalled 114 and noted cases 195. For non-Natives, the totals were 35 and 23. Over-estimates from medical records are 71% and 52%, respectively.

Table 2. Rates for FAS-Noted Individuals and FAS Case Patients per 100 Live Births, Alaska Natives and non-Natives: 1977-1992.

	Native		Non-Native	
	FAS Noted	FAS Cases	FAS Noted	FAS Cases
1989-92	5.7	2.5	0.4	0.3
1985-88	6.7	4.1	0.2	0.2
1981-84	5.9	3.8	0.2	0.1
1977-80	2.4	1.4	0.2	0.1
Total	**5.2**	**3.0**	**0.3**	**0.2**

Source: Egeland, et al. 1998

An updated report from the CDC's FASSNet suggest that prevalence rates are approximately 0.4 per 1,000 births for the total population between 1995 and 1997 (Center for Disease Control, 2002). For American Indian populations, rates were found to be substantially higher than other populations but not uniformly so. For example, the rate for Alaskan Natives (5.6 per 1,000 births) exceeded the rates for non-Natives eighteen-fold; by contrast, in New York and Colorado only one case of FAS was reported in the Native population. A subsequent release of FASSNet data for the period 1995 to 1999 period (Table 3) suggests that rates of FAS are relatively stable, with a slight decline among Alaska natives from 5.6 to 5.0 (Meany, Miller & FASSNet Team, 2003).

Table 3. FAS Prevalence by Selected Ethnicity, 1995-99, FAS Surveillance Network

	Alaska		Arizona		Colorado		New York	
	Cases	Rate per 1,000	Cases	Rate per 1,000	Cases	Rate per 1,000	Cases	Rate per 1,000
Non-Hispanic White	10	0.3	16	0.1	24	0.2	33	0.3
American Indian	60	5.0	53	2.0	1	0.7	2	0.7
All Cases	**70**	**1.4**	**113**	**0.3**	**51**	**0.3**	**79**	**0.5**

Source: Meany, et. al. 2003

Williams and Odaibo (1999) estimated incidence in northeastern Manitoba by examining all records for live births in Thompson, Manitoba in 1994. From this birth cohort, all

children with suspected FAS were selected for follow-up two years after birth. These cases were selected if one of the following characteristics were found: maternal abuse of alcohol during pregnancy, low birthweight, head circumference of 33 cm or less (10th percentile), and/or maternal use of alcohol and birth or pregnancy complications associated with FAS. Of the 745 live births, 90 children were identified for follow-up. Forty-nine of the ninety cases were not seen after identification: sixteen of the children because of the remoteness of their communities; eight because they were not locatable; and twenty-five because their home communities would not give permission for the pediatricians to visit.[11] Of the forty-one children available for the study, five were diagnosed with FAS, for a prevalence of 7.2 per 1,000 live births. If the non-participants lost to follow-up differ in FAS status relative to those studied, the results may be biased. The authors suggest that the rate would be higher if these children were included in the study.

Table 4. Summary of Prevalence from Aboriginal FAS, FAE or FASD Studies, 1983-2003

Authors	Publication Year	Location	Study Type		Estimate (1000 births)	Denom.	Notes
May, et. al.	1983	Navajo, SW US	active	FAS	1.4	N/A	0-18
		Pueblo, SW US	active	FAS	2	N/A	0-19
		Plains, SW US	active	FAS	10.3	N/A	0-20
Wong^	1983	BC	active	FAS	6.6	N/A	births
Asante, et al.^	1985	YK, NW BC; 36 communities	active	FAS+FAE	46 (YK)	586	0-16
				FAS+FAE	25 (NW BC)		
Robinson, Conry and Conry	1987	BC Aboriginal community	active (screening)	FAS	190	116	
Chavez, Cordero & Becerrant	1988	United States	surveillance, birth records	ICD-9	2.9	19,412	live births
Christensen^	1990	Alaska (1986-1986)	active	FAS	5.1	N/A	
				FAE	1.7	N/A	
		Alaska (1986-1988)	active	FAS	2.7	N/A	
				FAE	1.7	N/A	

CONTINUED ON PAGE 19

11 Community consent was not sought by the researchers, but in one instance a community became aware of the work and expressly asked the pediatricians not to visit. Tait (2003) presents a discussion of the implications for research in First Nations communities that discusses this study in more detail.

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Authors	Publication Year	Location	Study Type		Estimate (1000 births)	Denom.	Notes
Burd^	1991	ND, United States	active	FAS	3.1	15,531	0-18
Duimstra, et. al.	1993	4 Reserves, Plains, S Dakota	surveillance + active	FAS	8.5	1,022	<2 years
Bergeson	1993	Alaska	active	FAS+FAE	2.1	32,932	0-19
Habbick, et. al. *	1996	Saskatchewan (1973-77)	surveillance, case exam	FAS	0.52	77,670	
		Saskatchewan (1978-82)	surveillance, case exam	FAS	0.62	84,580	
		Saskatchewan (1983-87)	surveillance, case exam	FAS	0.61	88,520	
		Saskatchewan (1988-92)	surveillance, case exam	FAS	0.59	79,800	live births
Chudley~	1997	Manitoba	active (screening)	FAS	61	179	5-15
				FAE	33	179	5-15
Egeland, et. Al.	1998	Alaska (1977-80)	mulitple sources	FAS	1.4	7,160	live births
		Alaska (1981-84)	mulitple sources	FAS	3.8	8,971	live births
		Alaska (1985-88)	multiple sources	FAS	4.1	10,150	live births
		Alaska (1989-92)	multiple sources	FAS	2.5	11,065	live births
Williams and Odaibo *	1999	NE Manitoba	active (screening)	FAS	7.2	696	Adjusted
Miller et al.	2002	Alaska (1995-97)	multiple sources	FAS	5.6	7,117	
		Arizona (1995-97)	multiple sources	FAS	2.5	15,685	
Meaney, et al.	2003	Alaska (1995-99)	surveillance	FAS	5.01	11,974	live births
		Arizona	surveillance	FAS	8.38	26,440	live births
		Colorado	surveillance	FAS	18.2	1,535	live births
		New York	surveillance	FAS	0.7	2,856	

^ Reported in Burd and Moffat, 1994

* Study included a large number of Aboriginal cases but proportion not specified

~ Reported in Square, 1997

5.0 Estimates of the Extent and Costs of FAS/FASD in the Aboriginal Population

A general sense of the extent and cost of FAS/FASD can be gained from applying prevalence and per capita cost estimates to the Aboriginal population. Since specific costs and prevalences are not available for Aboriginal populations, however, such estimates are inherently flawed and only included to provide rough estimates.

Table 5 shows the estimated number of FASD/ARND cases using the prevalences from Sampson, et al. (1997), since these are of higher quality. A major assumption is that population-based rates used here are applicable to both Aboriginal and non-Aboriginal populations without regard for geographic variation, social and economic differences between Nations, or that the rate is constant across the age spectrum. These assumptions may not be tenable and the estimated numbers should be seen in that light. What is appropriate to note, however, is that the number of potential cases – if prevalence is indeed similar between Aboriginal and non-Aboriginal peoples in Canada – is fewer in the Aboriginal population. In absolute terms, even if prevalence is potentially higher among Aboriginal people in Canada, the proportional difference in population size means that the numeric 'burden' of FAS or FAS/ARND is greater in the non-Aboriginal population.

Table 5. Estimated FAS/ARND Cases, Canadian Aboriginal Population, 2001

	Populations					
	Total	Aboriginal	North American Indian	Métis	Inuit	Non-Aboriginal
	29,639,030	976,305	608,850	292,305	45,070	28,662,725
FAS Prevalence						
2.8 / 1,000	82,989	2,734	1,705	818	126	80,256
4.8 / 1,000	142,267	4,686	2,922	1,403	216	137,581
Combined FAS and ARND						
9.1 / 1,000	269,715	8,884	5,541	2,660	410	260,831

The potential economic costs associated with FASD are likely substantial. In a recent study, Stade, Ungar, Stevens, Beyene, and Koren (2006)[12] interviewed 148 parents or caregivers of children with FASD[13] and estimated annual costs including productivity losses because of care and estimates for societal costs from such dimensions as externalizing behaviour. Of the sample, 45% (67) of both children in the study and their caregivers were Aboriginal persons. The adjusted annual cost associated with FAS/FAE in Canada for individuals aged 1 to 21 is $14,342 (95% CI: $12,986 - $15,698). Geographic location, age of the child, and severity of illness were all significant determinants of cost. Over the age span from early childhood to age 21, the largest annual resource outlays were earlier in life, characterized by higher health care utilization. As the children in the study aged, the annual costs decreased and predominantly reflected education and external behavioural needs.

While the study was designed to measure individual-level costs and not provide population estimates, the authors do suggest that based on a conservative FAS/FAE prevalence in the general population of 3/1000, the potential annual costs for Canadian children in this age range could be around $344,208,000 (Stade, et al., 2006). Applying the adjusted cost estimates per child to the 2001 Census Aboriginal population aged 0 to 19 (the years available in Statistics Canada's tabular data) for the regions shown in the Stade et al. paper and the same rate of FAS/FAE of 3/1000, the annual costs are potentially around $18,056,000 for Aboriginal children in Canada. These costs are highly sensitive to the choice of prevalence estimate. Using the rate of 9.1 from Sampson, et al. for combined FAS/ARND, the costs associated with Aboriginal children aged 0 to 19 increase to $54,770,000.

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12 An additional note about this study is that it is one of the few to be national in scope, with study participants nearly equally divided between eastern, central, and western Canada.

13 Study participants were elicited through FASWorld, a national parent support organization. No detail was provided in the article on how parents were actually selected. Some potential bias may result from using FASWorld members, since members of a support organization may be more cognizant of the costs associated with the condition.

6.0 Conclusions

In 1996, MacMillan, MacMillan, Offord and Dingle (citing Bray and Anderson, 1989) wrote that although:

> *... recent studies suggest that fetal alcohol syndrome (FAS) is more prevalent among Canadian native children than among non-native children, the evidence is inconclusive .... Because there is insufficient information about the prevalence of FAS in the non-native population, it is impossible to conclude that there is a higher prevalence among native people.* (p. 1576)

From an epidemiological standpoint, the view of MacMillan et al. that higher prevalence of FAS among Aboriginal peoples cannot be assessed is still valid and applies also to FASD in general. As this review has suggested, the variety of methods used to produce incidence and prevalence of FASD among Aboriginal peoples prevents pooling the data together to produce single, over-arching estimates. At the same time, relatively little work has been done in the general Canadian population to set baseline prevalence that might be used to compare rates observed in studies of particular populations.

Yet this may be too narrow a view and risks dismissing a clear public health concern among Aboriginal peoples. It is unlikely that all of the observed differences between

Aboriginal and non-Aboriginal populations is driven entirely by selection or study biases. No baseline Aboriginal prevalence rate can be estimated; the range is too extreme for an average, as per Abel's global estimates, to act as a surrogate. At the same time, to discard all reports of excess rates in Aboriginal communities is to risk ignoring what may potentially be a serious public health risk with long-term consequences and may run counter to the experiential knowledge of communities themselves.

This suggests that more research on prevalence is necessary. Given the methodological issues described in this section of the report, Burd and Moffat's (1994) advice on the design of new studies is relevant:

> Future studies should include four features: (a) the cohorts should include people with both FAS and developmental disorders other than FAS; (b) the cohorts should be stratified by ethnic status; (c) the blinding of diagnosticians to the history of maternal alcohol use during pregnancy; and (d) the expansion of study designs to allow for identification of sensitivity and specificity of both screening methods and diagnostic criteria. (p. 692)

Tait (2003) adds that study designs should also examine the socio-economic status of women and their offspring. To this we can add other features, including stratified sampling of communities by Nation and investigation of Aboriginal-specific diagnostic criteria. Other issues relating to FAS/FASD beyond prevalence have not been extensively explored. There has been silence, for instance, on what the long-term effects of FAS have been on Aboriginal communities as individuals with the syndrome age. There is no clear sense of how historically-deep the syndrome may be, and thus First Nations reserves may have been dealing with its unobserved consequences for decades. FAS, according to Abel (1995), is not an 'equal opportunity' birth defect because it disproportionately affects individuals of low socio-economic status. Another dimension of 'unequal opportunity', however, may be the pooling of FAS in small Aboriginal communities.

7.0 Appendices

Appendix 1: Recommended Diagnostic Criteria for FAS, Partial FAS and ARND, Public Health Agency of Canada, National Advisory Council on Fetal Alcohol Spectrum Disorder

The criteria for the diagnosis of fetal alcohol syndrome, after excluding all other diagnoses, are:

1. Evidence of prenatal or postnatal growth impairment, as in at least 1 of the following:
   i. birth weight or length at or below the 10th percentile for gestational age
   ii. height or weight at or below the 10th percentile for age
   iii. disproportionately low weight-to-height ratio (= 10th percentile)
2. Simultaneous presentation of all 3 of the following facial anomalies at any age:
   i. short palpebral fissure length (2 or more standard deviations below the mean)
   ii. smooth or flattened philtrum (rank 4 or 5 on the lip-philtrum guide)
   iii. thin upper lip rank
3. Evidence of impairment in 3 or more of the following central nervous system domains: hard and soft neurologic signs; brain structure; cognition; communication; academic achievement; memory; executive functioning and abstract reasoning; attention deficit/hyperactivity; adaptive behaviour, social skills, social communication.
4. Confirmed (or unconfirmed) maternal alcohol exposure.

The diagnostic criteria for partial fetal alcohol syndrome, after excluding other diagnoses, are:

1. Simultaneous presentation of 2 of the following facial anomalies at any age:
   i. short palpebral fissure length (2 or more standard deviations below the mean)
   ii. smooth or flattened philtrum (rank 4 or 5 on the lip-philtrum guide)
   iii. thin upper lip rank
2. Evidence of impairment in 3 or more of the following central nervous system domains: hard and soft neurologic signs; brain structure; cognition; communication; academic achievement; memory; executive functioning and abstract reasoning; attention deficit/hyperactivity; adaptive behaviour, social skills, social communication.
3. Confirmed maternal alcohol exposure.

The diagnostic criteria for alcohol-related neurodevelopmental disorder, after excluding other diagnoses, are:

1. Evidence of impairment in 3 or more of the following central nervous system domains: hard and soft neurologic signs; brain structure; cognition; communication; academic achievement; memory; executive functioning and abstract reasoning; attention deficit/hyperactivity; adaptive behaviour, social skills, social communication.
2. Confirmed maternal alcohol exposure.

The term alcohol-related birth defects (ARBD) should not be used as an umbrella or diagnostic term, for the spectrum of alcohol effects. ARBD constitutes a list of congenital anomalies, including malformations and dysplasias and should be used with caution.

Source: Chudley, et al. 2005 (S11-S12).

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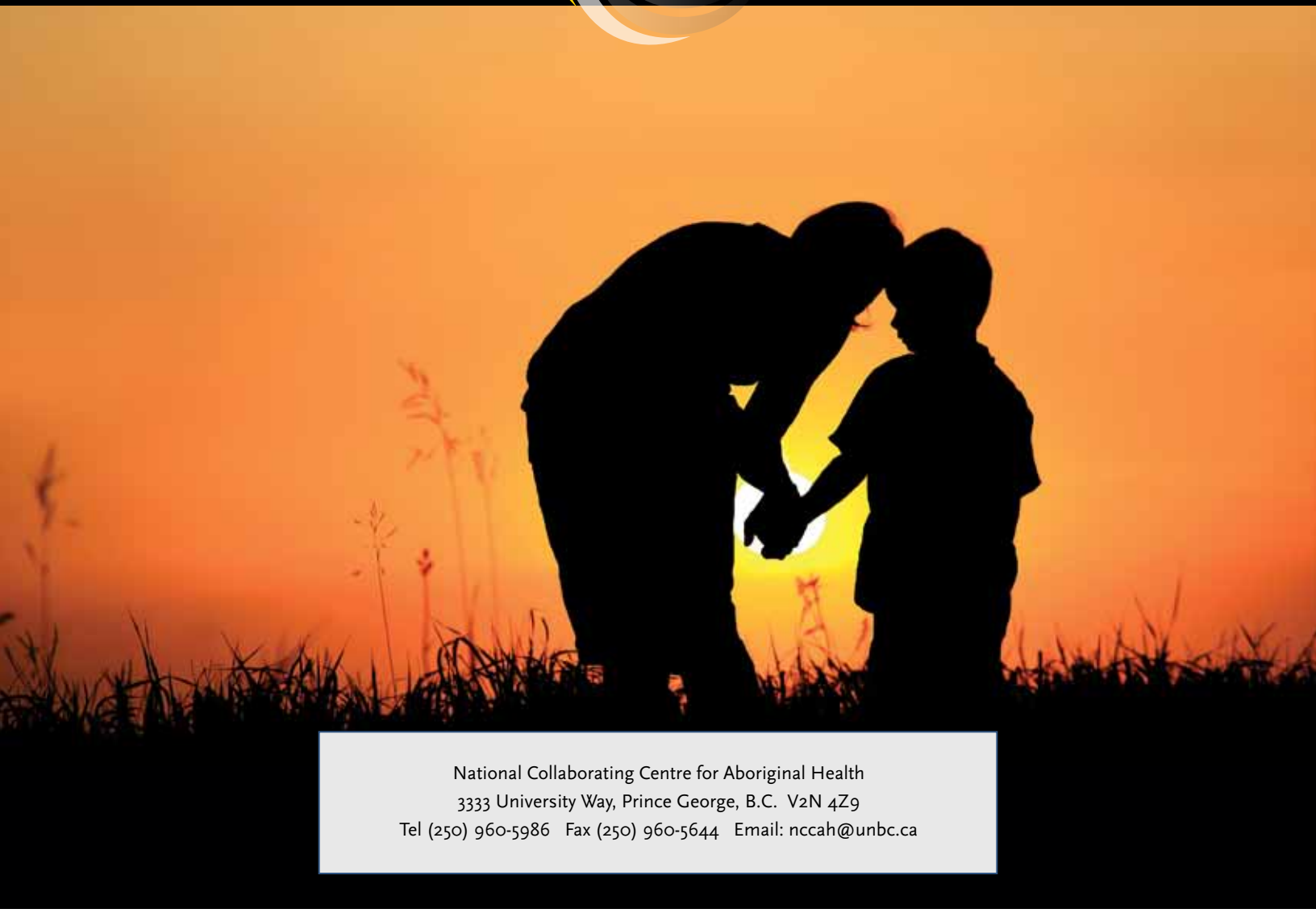

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