

FETAL ALCOHOL SYNDROME & FETAL ALCOHOL SPECTRUM DISORDER AMONG ABORIGINAL CANADIANS: *Knowledge Gaps*

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INTRODUCTION



Fetal Alcohol Spectrum Disorder (FASD) is an umbrella term encompassing a range of outcomes linked by maternal consumption of alcohol during pregnancy. Under the current Canadian diagnostic guidelines, these outcomes include Fetal Alcohol Syndrome (FAS), partial FAS (p-FAS) and Alcohol-Related Neurodevelopmental Disorder (ARND), and are marked by developmental disorders, weight and height deficiencies and a specific set of facial characteristics to varying degrees (Chudley, Conry, Cook, Looock, Rosales, & LeBlanc, 2005). Some of the characteristics of FASD are relatively well understood, but there are at the same time substantial gaps in the knowledge available in the published literature. Summaries of this literature are widely available, but few articulate the broad knowledge gaps, particularly in the context of Aboriginality.

No biomedical research indicates that alcohol teratogenesis differs between Aboriginal and non-Aboriginal

Canadians. Differences in issues surrounding the extent of FASD, the efficacy and delivery of prevention and treatment programs, and adverse life outcomes are instead linked to the social and economic environment of mothers and children. On the population side of the research agenda, Aboriginal-specific studies are necessary to understand and better address FASD in Aboriginal communities.

The impact of FASD has been shown in the literature to be a serious public health concern in some communities. Aboriginal organizations, as well as Federal and Provincial governments, have made Aboriginal FASD a priority and as a result, a number of programs have been established to reduce the incidence of FASD and provide long-term treatment. At the same time, the published academic literature is relatively silent on some dimensions of FASD. Many possible gaps can be identified, and it is partially the intent of this paper to stimulate the dialog



on what future research directions are the priorities. In this context, major gaps are structured along the 'life course' of FASD. This life course begins prenatally, where evaluated best practices for prevention and education appropriate for Aboriginal peoples in Canada are important in preventing FASD. Where FASD occurs, the next stage is birth and childhood, where there are two major gaps. Firstly, basic counts of the number of new cases of FASD – or prevalence – is largely unknown in both the Aboriginal and non-Aboriginal population in Canada. Secondly, while there is a substantial range of programs for children with FASD across the country, little evaluation of these programs has been done. The last stage in the 'life course' discussed here relates to later life, raising questions about the FASD population and the continued demands and resulting costs, both social and economic, on the fabric of Aboriginal communities.

The scope here is the published literature, and it should be emphasized that other forms of knowledge are not included. The epistemology of health-related research relies extensively on the published literature to justify, obviate the need for, and to add a comparative basis for additional work in a particular field. Other kinds of knowledge – including that of communities, individual clinicians and those who deal with people with FASD on a daily basis – are also informative but not included here.

In terms of structure, this paper begins with an introductory section describing the information sources, definitions associated with FASD, and the context of knowledge gaps and transfer. Following this section, the broad knowledge gaps are discussed. A final section suggests possible future directions and provides a short overview of the current FASD research environment.

Methods: Locating Evidence

The studies described in this report were found primarily through searches of Medline and Web of Science databases. The initial searches were conducted for a separate review of Aboriginal FASD prevalence also prepared for the National Collaborating Centre for Aboriginal Health (NCCAHA), and these articles provided an initial source for identifying knowledge gaps. The starting point for these searches was the MeSH heading "fetal alcohol syndrome", which was used to search Medline for articles relating to FAS. No specific MeSH term exists for fetal alcohol spectrum disorder, although FASD was used as a full-text keyword term in other searches. 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; Indigenous 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 Aboriginal-specific population-based studies on FAS/FASD, the historical window of this review is deeper than it might be for other summaries of research papers. These are supplemented with literature within the broad gaps that are not specific to Aboriginal peoples but of potential importance.

Identifying knowledge gaps: The initial searches were made in preparation for a companion report summarizing the literature on the prevalence of FASD among Aboriginal peoples in Canada. In the review of these papers, numerous authors pointed to future research questions and unanswered domains; these provided the basis for further searches of the literature and narrowing of the scope

of potential knowledge gaps. Because of the nature of the literature on FASD and Aboriginal peoples, the review portions of this paper are not systematic but narrative.





THE CONTEXT OF FASD



Fetal Alcohol Spectrum Disorder (FASD) is an umbrella term encompassing a series of related outcomes. All are associated with the intake of alcohol by women during pregnancy. Although FASD is a relatively recent term, Fetal Alcohol Syndrome (FAS) has been identified since the early 1970s and since that time, considerable research on FAS and FASD has been presented in the academic, peer-reviewed literature. A major thread within this literature is the extensive laboratory research on FASD, which includes research on the teratogenic effects of alcohol on the developing fetus, documenting the neurodevelopment of FASD and investigating new diagnostic techniques.

The first reports of the possible effects of prenatal exposure to alcohol were published in the French medical literature by Lemoine (2003) in 1968, followed by more comprehensive and clinically-descriptive case reports by Jones, Smith, Ulleland, and Streissguth in 1973.

Although knowledge that alcohol may harm the fetus had been reported in different forms for centuries, the Jones and Smith articles laid the framework for the current delineations of FAS (Calhoun & Warren, 2007).

Current knowledge clearly implicates alcohol as a teratogen, causing cell death and inhibiting the cell growth necessary for central nervous system (CNS) development. A probable contributing factor is the oxidative stress placed on the developing fetus by alcohol (Welch-Carre, 2005). Although it appears likely that increasing consumption of alcohol, particularly in the first trimester of gestation, is associated with an increased risk of a child developing FASD, there are no studies that have determined a safe threshold of alcohol consumption during pregnancy. There is some evidence, however, that binge drinking is related to the development of FASD. Animal studies have shown that binge-like drinking results in smaller brain weights



at birth compared to more continuous, lower dose exposure (Maier & West, 2001). In a secondary component of 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 bingeing ($n = 99$) had a smaller proportion of FASD children ($n = 8$; 8.1%) (Barr & Streissguth, 2001). In a meta-analysis of twenty-four studies, Polygenis, Wharton, Malmberg, Sherman, Kennedy, Koren and Einarson (1998) found no evidence of increased fetal malformations at birth with moderate consumption (greater than two drinks per week to a maximum of two per day). Among moderate alcohol users, they found the odds ratio for malformations 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.

The outcomes associated with FASD in the general population have also been documented. The behavioural phenotype associated with FASD and reflected in the current Canadian diagnostic criteria

includes life-long cognitive and functional disabilities, including impairments to intellectual ability; attention and speed of information processing, executive functioning, language, visual perception, learning and memory; and number processing. Behavioural dysfunctions include poorer academic performance, adaptive behaviour and emotional functioning (Kodituwakka, 2007). The result is that individuals with FASD may have substantial later-life difficulties manifested as alcohol or drug problems, trouble with the law, confinements, inappropriate sexual behaviour, and disrupted school experience (Streissguth, Bookstein, Barr, Sampson, O'Malley, & Kogan, 2004).

The current Canadian diagnostic guidelines recognize three separate but related categories under the umbrella of FASD: Fetal Alcohol Syndrome (FAS), Alcohol-Related Neurodevelopmental Disorder (ARND) and partial FAS (p-FAS), which are briefly described in Table 1. Fetal Alcohol Effects (FAE) and Alcohol-Related Birth Defects (ARBDS) are also included in this table because some earlier studies noted here make reference to these terms.

Knowledge Gaps, Knowledge Transfer and Aboriginal Peoples

While the emphasis here is not on research protocols per se, knowledge gaps should be operationalized into research and inevitably action. These gaps should lead to research that will allow the results to be generalized to broader populations beyond the sample used in a particular study.

All research on FAS/FASD eventually contributes to the greater store of knowledge regardless of the population sub-group studied. Some research questions, however, may have more direct relevance to Aboriginal communities, particularly if the research directly involves these communities. For instance, relatively little work has been done directly on the relationship between socio-economic status (SES) of Aboriginal women, their communities, and FAS/FASD despite the consistent observation across studies that women of lower SES are at greater risk (Basford & Thorpe, 2005).

In the traditional scientific context, the academic literature becomes the primary mode of knowledge dissemination. Evidence-based practices complete the loop between individual, researcher, dissemination and public health service

Table 1. Definitions of 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. It is not a clinical diagnosis by itself (Chudley et al., 2005).
Fetal Alcohol Syndrome (FAS)	The most recent Canadian diagnostic criteria for FAS require confirmed exposure to alcohol during pregnancy and three broad anomalies: pre-or-postnatal growth retardation; specific 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).
Fetal Alcohol Effects (FAE)	FAE is a less 'complete expression' of FAS. In one working definition, a person having two of growth deficiencies, facial dysmorphism or central nervous system dysfunction and confirmed in utero exposure is considered to have FAE (Spohr, Willms, & Steinhausen, 2007). The term FAE has been criticized as inappropriately implying a causal link between exposure and outcome and has often been poorly defined (Sampson et al., 1997).
Alcohol-related birth defects (ARBD)	ARBDs generally refer to clinical abnormalities 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 (Jones et al. 1973). ARBD does not constitute a diagnostic category by itself in the Canadian FASD guidelines.

delivery. Traditionally, study participants may not directly be part of the loop, in the sense that they may not either see the finished written results or perhaps directly benefit from evidence-based practices emanating from the research.

Knowledge transfer of FASD research findings becomes more acute for Aboriginal communities because of the historical distance between non-Aboriginal and Aboriginal peoples. As active partners in the research process, the involvement of Aboriginal peoples in the design, implementation, interpretation and development of best practices not only helps alleviate ethical concerns but also helps bridge the gap between worldviews that are often distinct.

Ethical research practices incorporate knowledge transfer, a view echoed by the Royal Commission on Aboriginal Peoples (1996), which specifically included consultations on knowledge, collaboration and access to research results and community benefit as key elements of their guidelines for research. While some organizations have adopted similar frameworks (University of Victoria, n.d.), not all research institutions have embedded ethical practices that include direct knowledge transfer into their research practices.

Knowledge transfer is more than the publication of study results and happens along various points in a continuum from the choice of research questions to the

application of knowledge in the form of concrete, best-practice interventions and programs. Participation in all phases of the research process may encourage the development of an Aboriginal-led research agenda and the transfer of methodological knowledge.¹ It is important to emphasize that this is not a 'one-way' process and that there are benefits to the research community as well. The trust engendered by active partnerships may allow greater access to communities and contact with key informants; knowledge of community or Nation-specific approaches to the interpretation of the data; the informing of new techniques for the collection and analysis of data; and better understanding of the meaning and relevance of a broad research agenda within Aboriginal culture.

¹ While knowledge transfer has received substantial attention in the relationship between researchers and Aboriginal communities, there are tensions. In particular, there may be substantial differences of opinion over the ownership of data and access to it during the research process between researchers and community partners. In addition, the traditional model of research promotes distance between participants and researchers to minimize bias.





KNOWLEDGE GAPS

There is no dimension of FASD where the published literature indicates strong understanding regarding Aboriginal peoples² in Canada. One means of identifying knowledge gaps is to view important intersections along the 'life course' of FASD. The first of these is the early pre-FASD stage, focusing on prevention and treatment for women at risk. The second point is the inception of FASD, from which prevalence and incidence are important markers but not well understood for Aboriginal peoples in Canada. Additionally, program evaluation for treatment programs for children with FASD is largely absent from the literature. The third stage along FASD's 'life course' is later life, since much of the literature focuses on childhood but is relatively silent on the impact of FASD on communities and individuals at older ages.

The Pre-FASD Stage: Prevention and Treatment Programs for Women

FAS/FASD is widely perceived to be "100% preventable." Because it is a birth outcome linked to behaviours, programs to reduce or eliminate alcohol consumption by women during pregnancy have clear potential benefit in reducing the incidence of FAS/FASD. Prevention has been bundled into primary, secondary and tertiary strategies (Stockburger, 2003). Primary prevention is broad, promoting the health and well-being of a community through evidence-based practice. Community-based programs tend to be multidisciplinary in nature, focusing not only on individual behaviours but also on modifying systemic practices within the

²Throughout this document, when not referring to specific studies found in the literature, the term "Aboriginal" is used to refer to First Nations, Inuit, and Métis peoples inclusively. However, use of this term by other researchers and organizations may not be as inclusive and remains their own.



community (Basford & Thorpe, 2005). Secondary prevention is directed to women who are at risk, in this case women with substance abuse problems who may become pregnant. In tertiary strategies, 'prevention' does not focus on the mother but on harm reduction for the already-exposed fetus.

A wide range of prevention strategies and risk reduction programs for women – both Aboriginal and non-Aboriginal – has been implemented across Canada and the United States. Extensive reviews of such programs are available in Stockburger (2003), Basford and Thorpe (2005), and Legge, Roberts and Butler (2001). A key component of future programs is the modification of women-centred and culturally appropriate programs for smaller and rural communities (Stockburger, 2003). However, a modified systematic review of the grey and academic literature found that a lack of empirical knowledge makes it difficult to sufficiently inform policy, practice and education (Basford & Thorpe, 2005). For pregnant women, the literature is sparse and, similar

to Aboriginal-specific studies, hampered by small sample sizes and few comparison groups (Howell, 1999; cited in Dell and Roberts, 2005).

Program evaluations of Aboriginal-specific prevention programs have not been widely reported in the published literature. A recent study by Glik, Prelip, Myerson & Eilers (2008) may, however, provide some insight into the design and evaluation of FASD prevention programs in lower-SES communities of women. In this example, messages were 'narrowcast' to two disadvantaged communities in southern California and designed in conjunction with community advisory committees, segmenting the audience by language and ethnicity. Focus groups were used to define the messages to be used in the information campaigns and used to pre-test materials. Rather than using fear-based messages, primary interventions were based on language and ethnic-specific social norms and distributed within the communities. To assess exposure to the materials, two-wave repeated cross-sectional survey data were collected through women's health

and physician clinics and, more broadly, through a community-wide random digit dialed survey of women aged 18 to 35. Since the two communities designed and distributed the message materials differently, substantial differences in the exposure to the prevention messages emerged, ranging from 11.3% of women surveyed in clinic settings in one community versus 54.2% in the second. Predictors based on a backwards stepwise regression of the pooled data suggests, in addition to living in the latter community, that having a high school education (OR: 1.63, 95% CI: 1.01-2.68) and being a drinker (OR: 2.16, 95% CI: 1.08-4.33) resulted in greater likelihood of having been exposed to the messages. While this study does not provide assessment of changes in attitudes or behaviours associated with the program's messages, it does provide an example of design and evaluation of narrowcasting techniques designed in conjunction with communities that may be relevant to FASD programs in other contexts (Glik et al., 2008).

Although most provinces and territories have some programming in place for high-risk women, routine and uniform screening to identify them for secondary and tertiary prevention is less common and is only beginning to be implemented in some jurisdictions (Health Canada, 2001). The most appropriate points of contact for such screening are likely physicians and nurses engaged in prenatal care, although women who are likely to drink heavily during pregnancy may not have access to or utilize these services. Aboriginal women, particularly in urban areas, may be at higher risk of not receiving prenatal services. In a study of 652 postpartum women in Winnipeg, Manitoba, Heaman, Gupton, and Moffatt, (2005) found that a significantly higher proportion of Aboriginal women received inadequate prenatal care compared to non-Aboriginal women (15.7% versus 3.6%). In a model controlling for both personal characteristics (i.e., stress and low self-esteem) and socio-economic factors, the authors found that Aboriginal identity remained a significant predictor of inadequate care.

Screening tools for identifying high-risk women may be useful for pre-conceptual education or, for women who are pregnant, ensure that early treatment for alcohol-exposed children is available. Table 2 provides an overview of these instruments. There is moderate evidence to suggest that T-ACE and TWEAK may help identify women who would benefit from further interventions (Tait, 2003). Russell, Martier, Sokol, Mudar, Jacobson, and Jacobson (1996) measured the efficiency and predictive power of these tests in a sample of disadvantaged African-American periconceptual women in Detroit, Michigan. In this sample, the sensitivity – which captures the probability that a risk drinker is positive on a test – of T-ACE and TWEAK was 88% and 91%, respectively.³ Specificity,

which measures the probability of a non-drinker scoring negatively, ranged from 79% and 77% for T-ACE and TWEAK, respectively. In a Washington State study involving inner-city women, the sensitivity and specificity of TWEAK administered during pregnancy was assessed as 70.6% and 73.2% (Dawson, Das, Faden, Bhaskar, Krulewitch, & Wesley, 2001). Currently, only the T-ACE tool has been compared to other methods of alcohol-use detection in an Aboriginal context; Gale (cited in Roberts and Nanson, 2000) found

that T-ACE significantly increased the detection of ‘high risk’ patients compared to clinician’s assessments alone in a North Plains reservation population. Such screening tools are, however, likely of less value if access to early prenatal care or treatment programs are not available.

The underlying reality of programming for Aboriginal women is that even without proper evaluation studies, interventions and treatments are certainly better than no treatment and that the greater harm is



³Russell et al. (1996) present a variety of specificity and sensitivity values for these tests based on particular cut-points along receiver-operating curves. The values shown here represent the second cut-point in the data.

Table 2. Overview of Alcohol Screening Tools



Instrument	Features	Strengths	Concerns
CAGE Cut down Annoyed Guilty Eye opener	4 Questions. Not specifically designed for screening pregnant women, has served as a source of items for questionnaires designed to screen for risk drinking during pregnancy.	Assess lifetime rather than current alcohol related problems.	Does not identify heavy drinkers who have not experienced alcohol related problems. More effective in screening men than women.
*T-ACE Tolerance-Annoyed Cut down Eye opener	4 Questions. One question regarding how many drinks to feel high, and three questions from the CAGE.	Developed for use in obstetric gynecological practice. More sensitive to risk drinking than the CAGE.	
*TWEAK Tolerance Worry Eye Opener Amnesia Cut-down	5 Questions. Combines questions from the MAST, CAGE, & T-ACE.	More sensitive and less specific than the T-ACE. Outperforms the MAST or CAGE.	
MAST Michigan Alcoholism Screening Test	25 Questions. Not specifically designed for screening pregnant women, has served as a source of items for questionnaires designed to screen for risk drinking during pregnancy.		Does not identify heavy drinkers who have not experienced alcohol related problems. More effective in screening men than women.
*AUDIT Alcohol Use Disorder Identification Test	10 Questions. Combines questions about alcohol use directly and on consequences of alcohol use.	Early identification of harmful drinking rather than alcohol disorders such as alcohol abuse.	Not been evaluated in obstetric populations. Longer and more complicated to score.
*4 "P"s	4 Questions. Questions about alcohol or drug use during current pregnancy, in her past, in her partner, and in her parents.	Yes or No format. Easy to administer and score.	Potential lack of specificity and the possibility that women would answer direct questions about alcohol before questions about problems with alcohol.
Modified 5 "P"s	5 Questions. Questions about alcohol or drug use during this pregnancy, in her parents, in her partner, in her past, in her previous pregnancy. The 5 "P"s is an adaptation of the 4 "P"s.	Question about alcohol use during previous pregnancy may help to diagnose FAS in woman's other child(ren). One predictor of a FAS is being born to a mother with a child with FAS.	Potential lack of specificity and the possibility that women would answer direct questions about alcohol before questions about problems with alcohol.
*TQDH Ten-Question Drinking History	10 Questions. Focuses on type and amount of alcohol consumed.	Does not differentiate between beer, wine and liquor when determining at-risk drinking.	More than 4 drinks per week is considered risk drinking. Best for women not yet pregnant.

*Available at: www.ncemch.org/pubs/PDFs/SubAbuse.pdf Source: Baldwin, 2003



in doing nothing. As noted by Dell and Roberts (2005), one important conclusion can be drawn from the literature through the 1990's: women who remain in treatment fare far better than those who do not. However, more extensive program evaluations for Aboriginal-specific programs – and an evaluation of their scope and resources – may play key roles in further reducing the incidence and impact of FASD.

The Birth and Youth Stage: Prevalence Estimates of FASD and Treatment Programs for Children

In the case of FAS/FASD, basic measures of prevalence are lacking for Aboriginal and non-Aboriginal peoples alike. Among a small number of 'high risk' Aboriginal communities, the prevalence of FAS has been measured, but questions remain about its extent in other kinds of communities. A potentially important

direction would be to capture rates of FAS/FASD across Canada in a variety of settings to provide a more realistic estimate of prevalence for Aboriginal populations. Improved Aboriginal estimates would not address the issue of the lack of estimates in the general population, although such studies should be encouraged to provide comparisons.

No Canadian population-wide estimates of FAS and FASD exist. The worldwide incidence of FAS was estimated to be 0.97 cases per 1,000 live births by Abel in 1995, based on a pooling of international studies. Using more stringent criteria for case ascertainment and study design, the review by Sampson et al. (1997) reported incidence of FAS of approximately 1.4 per 1,000 live births in the United States, a figure that was adjusted upwards to 2.8 per 1,000 after adjustment for incomplete screening. For FAS and ARND combined, prevalence rates were estimated to be 8.3 per 1,000 (Sampson et al., 1997).

Published prevalence studies for Aboriginal peoples in North America have focussed almost exclusively on First Nations populations and FAS. In the United States, the FASSNET program captured cases from multiple sources and found rates of FAS ranging from 0.7 in New York State to 5.0 per 1,000 among Alaskan Indians in the 1995 to 1999 period (Meany, Miller, & FASSNET Team, 2003). In Canada, Aboriginal-specific studies have often occurred in response to clear public health concerns in specific communities. In a screening of children in one such community in the 1980's, Robinson (1992) found that prevalence of FAS and FAE to be 121 per 1,000 and 69 per 1,000, respectively. In a study of 36 communities in Yukon and northwest British Columbia, Asante (reported in Burd and Moffat, 1994) found high rates of combined FAS and FAE of 46 per 1,000 (Yukon) and 25 per 1,000 (northwest British Columbia). Between 1973 and 1993, 207 cases of FAS in Saskatchewan



were identified in a study by Habbick, Nanson and Snyder (1996). Of these, 178 cases were Aboriginal. Over the twenty-year period, the authors found that rate of FAS was consistent, averaging 0.59 per 1,000 live births. In all cases, maternal use of alcohol during pregnancy was confirmed. In the 1990's, Williams and Odaibo (1999) found through a screening study rates of 61 per 1,000 for FAS and 33 per 1,000 for FAE in Manitoba. In a cohort of live births in Thompson, Manitoba, children were followed up if they were suspected of having been exposed to alcohol during gestation or had birth characteristics indicative of exposure. In the follow-up, the majority of suspected cases were Aboriginal. Of the 90 children identified, 49 were not seen by the paediatricians because of the remoteness of the communities, they were not locatable, or because the home

communities would not give permission for the visits. Overall prevalence, using the available cases, resulted in a rate of 7.2 per 1,000 (Williams & Odaibo, 1999).

Typically, non-Aboriginal prevalence studies have largely relied on data from clinics or hospitals based in urban areas, which may result in under-estimates of rates. In comparison, screening studies of small communities will lead to higher estimates because of the near-complete study populations (May, 1991).

Currently available prevalence estimates from the published literature are not usable as proxies for FASD in the Aboriginal population. Clear gaps in the knowledge of the distribution of FASD are not exclusive to Aboriginal peoples; estimates also do not exist for the non-Aboriginal Canadian population.

There are no published estimates for communities in eastern Canada, among Métis or Inuit peoples, nor for urban Aboriginal populations. Given the continued attention given to FASD and Aboriginal peoples, more diverse estimates are necessary, not only to more accurately quantify the extent of FASD but also to better inform planning in different contexts.

While more complete estimates are necessary, there are substantial difficulties. Because FASD diagnoses are multi-faceted, guidelines strongly suggest that a team approach is necessary for proper diagnosis. However, the expense associated with a team approach means that providing diagnostic services to remote communities is generally not feasible. Screening tools are available that rely on a picture of FAS-related physical characteristics and allow non-clinicians to quickly identify potential cases (Burd & Juelson, 2003). However, the diagnostic criteria for the facial dysmorphism associated with FAS have not been validated in Aboriginal populations, and there are concerns that the accepted cluster of facial features may be similar to those normally present in some Aboriginal peoples. These obstacles are challenging since early diagnosis is necessary to facilitate meaningful deficit-based treatment programs (Basford & Thorpe, 2005). Furthermore, quick screening tools focussed on facial characteristics may not capture other dimensions of FASD where the 'face' of FAS is not apparent. Future research on prevalence should include prior validation of the facial dysmorphism of FAS, as well as validation of the tools used to assess CNS deficits for Aboriginal peoples.

Little data are available on the early-life home environments of Aboriginal FASD children in Canada. In the United States, a limited number of studies suggest that a large proportion of children with FAS live in institutional or foster care. Barth



estimates that up to 80% of FAS children live in such arrangements (Barth, 2001). Other American research has suggested that up to 80% of children living in institutions or foster care do not return to their family home. In Saskatchewan, Habbick, Nanson, Snyder, Casey and Schulman (1996) found that in their study group, which was 86.0% Aboriginal, fully 72% of children had been placed in foster care at some point and, at the time of the study, only 25.6% were living with a biological parent. It is not clear what proportions of FASD children in the wider Canadian population enter foster care, and, for Aboriginal children, whether this involves the children leaving their home communities.

In addition to the difficulties in measuring the number of people who are affected, there is a relative lack of high-quality research on the effectiveness of treatment programs for children with FASD. In a systematic review of the literature on interventions for children and youth, Premji, Benzie, Serret, and Hayden (2006) found only ten studies that met their criteria for methodology and scope. Of these, only three were studied in greater detail because the balance were case studies or could not be acquired. The authors conclude that given the evidence in these studies, the efficacy of FASD interventions for children and youth is

not scientifically substantiated and no conclusions can be drawn. The authors also suggest that the focus of intervention studies should be on reducing vulnerability, modifying environmental stressors and increasing protection, including positive, stable and supportive care-giving environments (Premji et al., 2006). Other research has suggested that child-centred school environment may be important to positive outcomes for FASD children. Low staff-child ratios, access to professionals including social workers, psychologists, nurses and speech specialists, and self-contained classrooms may all be important to improved outcomes for FASD children (Roberts & Nanson, 2000).

The Later-Life Stage: Aging and FASD

The fundamental focus of most research on FAS/FASD has been on children with less research on identification and treatment for older adolescents and adults. This is possibly attributable to the difficulty in identifying individuals with FAS/FASD past puberty, when growth catch-up and changes in facial morphology occur (Chudley et al., 2005). History of pre-natal exposure to alcohol may also be difficult or impossible to obtain later in life (Clark, Lutke, Minnes, & Ouellette-

Kuntz, 2004). Additionally, identification of people with FAS only began in the early 1970's and, for other dimensions of FASD, not until diagnostic criteria were widely disseminated much later.

British Columbia is the only province currently providing identification of FAS or FAE for adults. In a Health Canada survey of key informants,⁴ respondents recognized the lack of adult programs and research on FAS/FAE. Of the challenges and gaps noted in a larger service-provider survey, the lack of a range of services for adults was the third most-commonly cited after the lack of professional's knowledge of FAS/FAE and the lack of diagnostic services. Additionally, the few FAS/FAE-specific centres offering comprehensive diagnostic services are not available to adults (Health Canada, 2001).

A small number of longitudinal studies tracking individuals identified early in life have been conducted. Howell, Lynch, Platzman, Smith, and Coles (2006) reported on a longitudinal cohort recruited between 1980 and 1985 in Atlanta, Georgia. Women were recruited prenatally for the study from a largely low-SES African-American population if they reported at least two drinks per week during pregnancy. One-hundred and twenty eight children were studied from this group. Two sets of controls

were used: to control for socio-economic status (SES), 53 higher SES, non-exposed children were recruited for the study, and an additional control group of 84 adolescents from local school special education programs were selected to control for the effects of disability status on behaviour and academic functioning.

Of the exposed children, 46 were dysmorphic, while 82 were not. At an average age of 15.1 years (SD=0.94), the children were evaluated on cognitive, academic and physical dimensions (Howell et al., 2006). In this sample, prenatal alcohol exposure was associated with significant effects on cognition and academic performance but behaviour and adaptive functioning were not affected,

contrary to other published literature. The authors suggest that these differences may be the result of neurological damage unfolding in a compromised environment rather than a teratogenic effect per se.

In a German study by Löser, Bierstedt and Blum (1999), children with FAS or FAE who were identified between 1974 and 1997 in Munster hospitals were tracked into adulthood. By the time of the study (1997), 67 had reached adulthood. Fifty-two were available for study after loss to follow-up, refusal or incomplete diagnostic data. At the time of study, the constellation of familiar markers in childhood had abated but some characteristics were still noticeable, including low weight (29%), reduced

height (39%) or microcephaly (56%). The characteristic craniofacial markers of FAS in this sample were less apparent in adulthood. There was some evidence of asocial behaviour among the sample, but no comparison to a non-FAS set of controls. In comparison to the broader population of Munster, substantially few FAS children attended secondary or grammar schools and, as they moved into adulthood, their occupational profiles did not require substantial education.

A pivotal study on later-life outcomes by Streissguth et al. (2004) involved 415 subjects enrolled in the University of Washington's Fetal Alcohol Follow-up Study. Diagnoses of FAS and FAE were originally completed between 1972



⁴Of the 28 key informants, six were Aboriginal.

and 1995 and the criteria used in this period included a positive history of maternal alcohol consumption during the pregnancy, growth deficiency, specific facial characteristics, and CNS complications. Informants such as parents or guardians were interviewed about the individual's life history using a standardized tool; subjects were administered age-appropriate IQ, achievement and adaptive behaviour tests. The participant's median age was 14, and 25% were Native American. Streissguth et al. identified five adverse life outcomes (inappropriate sexual behaviour, disrupted school experience, trouble with the law, confinements (i.e., psychiatric or alcohol / drug hospitalization; incarcerations) and alcohol or drug problems) and calculated odds ratios for these outcomes in relation to a host of individual and environmental characteristics. Comparisons to non-FAS/FAE children are not made in terms of outcomes, but for those aged 21 or older each adverse outcome had been experienced by more than half of the study group with the exception of alcohol or drug problems.

The patients in this study were referred and thus may not be representative of the larger population of FAS/FAE cases. The findings do, however, provide some evidence of the potentially strong effect of environment on adverse life outcomes. Adjusted odds ratios suggest that a low percentage of life spent in a stable or nurturing home; being a victim of physical, sexual or domestic violence; and later age at diagnosis are associated with a greater likelihood of exhibiting adverse life outcomes to varying degrees. In this

group, the odds ratio for inappropriate sexual behaviour for those who had a low percentage of life in a stable or nurturing home versus those that did was 4.06 ($p = 0.0006$). Similarly high odds ratios were also found for disrupted school experience (OR: 4.67; $p = 0.0003$), trouble with the law (OR: 2.69; $p = 0.01$) and alcohol / drug problems (OR: 4.10; $p = 0.001$) (Streissguth et al., 2004).

In a 20-year follow-up of a cohort of 60 FAS/FAE patients in Germany, 37 participating individuals were assessed for physical characteristics; an interview assessed academic and occupational careers, domestic arrangements and independent living; and a standard questionnaire captured emotional and behavioural problems in young adults (Spohr, Willms, & Steinhausen, 2007). Parents, foster parents or an institutional caretaker provided responses for the interview and questionnaire.

In this cohort, BMI measured at follow-up largely normalized (Spohr et al, 2007). Some catch-up growth occurred, although a large proportion still had considerable growth deficiencies. In terms of occupation and education, the cohort had relatively poor outcomes, with only five individuals (5%) having held an 'ordinary' job and the same proportion having a secondary school education. Just over ¼ of the cohort lived in institutions and 35% were in a dependent-living situation with assistance from others. Despite having positive, more stable environments relative to the other cohort reported in Streissguth et al. (2004), outcomes were considered to be significantly impaired and the

authors conclude that their results do not corroborate the earlier findings, which they attribute to differences between German and American differences in patient selection, health care, FASD treatments and social systems (Spohr et al, 2007).

In one of the few examples of Canadian research on adult FAS, Clark et al. (2004) used a convenience sample of 62 individuals⁵ identified through the FAS/E Support Network of British Columbia, 24 of which (39%) were Aboriginal. The mean age of the sample was 22 years. Because of the small sample size, multivariate analyses of the data were not possible and the results were reported as bivariate odds ratios (ORs).⁶ Few relationships were statistically significant. The study does provide some suggestive evidence on secondary disabilities among adults with FAS/E, including the large percentage (91.3%) having other mental health diagnoses,⁷ and a history of confinement (eg. hospital or prison) and trouble with the law.

While the outcomes of persons with FASD in late adolescence and early adulthood have been documented, later life-course trajectories are not known (Clark et al., 2004). For Aboriginal peoples, there are multiple issues associated with aging. The economic and social impact of older persons with FASD in Aboriginal communities is not known. For those who may have faced incarceration, best practices for healing and reintegration appropriate for FASD also needs to be better understood (Clark et al., 2004).

⁵The study participation rate is reported as 55%, but this does not include a further 62 individuals who were excluded because they were incarcerated, could not be found, or 'deemed unable to participate', which is not defined in the paper. Including these cases shifts the potential response rate to 35%.

⁶An odds ratio is the ratio of probabilities between two groups, normally exposed versus non-exposed. When no difference exists between the odds of an outcome in the two groups, the OR is 1.0. Confidence intervals are generally supplied with ORs. While not a true hypothesis test, a confidence interval that spans 1.0 (for example, an interval of 0.8 – 1.6 around an OR of 1.2) suggests that the OR is not significant.

⁷Some bias may be introduced into this sample because all individuals are part of a structured clinical program. It is likely, then, that being in the 'system' makes it more likely that these individuals will receive diagnostic services for other conditions as well, elevating prevalence.





CONCLUSION

The three broad knowledge gaps – relating to evaluations of prevention and treatment programs, prevalence, and aging – have relevance to both Aboriginal and non-Aboriginal peoples in Canada alike, since FASD is not exclusive to either population. However, the attention paid to FASD and Aboriginal peoples in Canada supports a commonly-held belief that substance abuse during pregnancy occurs more frequently among Aboriginal women compared to their non-Aboriginal counterparts (Tait, 2003). While there may be considerable knowledge of best practices and FASD in Aboriginal communities, little of it has distilled to the published literature.

Each of the identified knowledge gaps relates to a separate rationale for expanded study among Aboriginal peoples. Future prevention and treatment program design needs a fundamentally more robust evaluation framework, and in turn the dissemination of knowledge through the academic literature is necessary to

ensure the promotion of best practices. Prevalence studies need to be undertaken in a wider range of communities and settings to provide better estimates of the extent of FASD across diverse peoples. Aging studies are necessary to better understand the life course trajectories of people with FASD, as well as the broader implications for communities. In the same sense that there is no single Aboriginal population, there is no single Aboriginal FASD experience, a common denominator to the gaps noted above. Future research on FASD among Aboriginal peoples to fill these gaps thus needs to reflect diversity, whether geographic, cultural or between urban and rural.

In Canada there is a substantial research community revolving around FASD and Aboriginal health that may provide synergy. While the entire scope of this community is not included here, selected examples provide a sense of the breadth of research currently underway. At the federal scale, Health Canada's FAS

Initiative includes a First Nations and Inuit Steering Committee. The Public Health Agency of Canada's *A Framework for Action* describes five broad goals that are fundamental to an FASD reduction strategy. These goals include increased public awareness of FASD and the impact of alcohol consumption during pregnancy; the development of increased capacity; effective national screening, diagnostic and data reporting tools and programs; expansion of the knowledge base and the exchange of information; and increased commitment and support for action on FASD (PHAC, 2005). While mention is made of Aboriginal groups as one partner, the focus in the Framework is non-specific and population-wide, in keeping with the pervasive nature of FASD across the entire Canadian population. Correctional Services of Canada has also been active in developing a protocol for screening in adult populations.

Provincially, British Columbia has been active in the development of co-ordinated research initiatives on FAS/FASD. Direct research has been initiated in the FAS Research Network of BC, as well as by the BC Centre of Excellence for Women's Health and the Centre of Excellence for Children and Adolescents with Special Needs: University of Northern British Columbia Task Force on Substance Abuse. British Columbia also includes FASD within its Health Status Registry (HSR), although diagnosis and reporting have been problematic (BC Vital Statistics Agency, nd). Cases in the HSR are collected and registered from hospitals and the Asante Centre, a multi-disciplinary diagnostic, assessment and family support service located in the province.

Research networks have also been formed specifically to support FASD research. The Canadian Northwest FASD Research

Partnership represents the collaboration of seven provincial / territorial jurisdictions with the goal of developing a common approach to prevention, intervention, care and support.⁸ One initiative of the Partnership is the Canadian Northwest FASD Research Network,⁹ whose purpose is to build relationships, maximize opportunities for FASD research capacity, and to engage in knowledge transfer. The Network's FASD project inventory provides summaries of current projects and proposals.¹⁰

A current example of collaborative partnership between Aboriginal communities and academic research is the work being undertaken by Masotti et al. (2006). In this instance, the communities involved are two First Nations in British Columbia and two in Ontario, and the interventions are drinking reduction programs focussed



⁸<http://www.cnfasdpartnership.ca/index.cfm>

⁹<http://www.fasdnetwork.ca/default.cfm>

¹⁰<http://www.fasdnetwork.ca/admin/contentx/default.cfm?PageId=4257>



on women who have previously given birth, since parity is a known risk factor for subsequent FAS-affected children. Three 'partners' participated in the design of the interventions, which as of this writing have not been implemented: the academic research group, an opinion leader workgroup, and a community oversight committee. Initially, research questions for this project were proposed by the Canadian Institute for Health Research's Institute for Aboriginal People's Health (IAPH). The academic researchers then identified local opinion leaders in the selected communities through local surveys of women of childbearing age. Workgroups were struck in each community comprised of these opinion leaders and a community research facilitator, who defined questions and strategies. In addition, an oversight advisory committee was struck in each community to provide "checks and balances" to the working group.

Aboriginal organizations have also been developing research capacity. The National Aboriginal Health Organization (NAHO) is an Aboriginal-run body whose objectives include the promotion of health through knowledge-based activities, the promotion of research, and

the development of research partnerships, including the development of principles of ownership, control, access and possession (OCAP) of Aboriginal research. A major undertaking by the FNC is the First Nations Regional Longitudinal Health Survey (RHS), an on-going survey of health behaviours and beliefs of on-reserve and some non-reserve communities taking place on a four year cycle. As of July, 2006, the RHS has moved to the Health and Social Development Secretariat of the Assembly of First Nations. Additionally, First Nations Statistics, an agency designed in part to strengthen First Nations analysis and dissemination of financial and census data, might represent an additional partner in the design and analysis of FAS/FASD research initiatives.

The development of new collaboration models for FASD research with Aboriginal peoples requires a different perspective on epidemiological method. One of the basic tenets of epidemiology is its universality; the basic techniques and study designs are transportable across populations and conditions, and reduce 'background noise' by focussing on precisely defined exposure and outcomes at the individual level. A population-specific epidemiology is in many ways a contradiction in terms.

Some dimensions of context, including historical patterns and processes, need to be removed to some extent because they are not easily controlled for and cannot be measured directly, yet these are implicated in diseases that are behaviourally-driven, such as FASD. If the importance of community in Aboriginal contexts is extended to the research process, the involvement of Aboriginal groups at all steps in the chain from conception to conclusion is necessary not only from an ethical standpoint, but also from the common endpoint, which is a reduction in incidence and more comprehensive care for those with FASD. It may not be fully possible to reconcile epidemiological and Aboriginal knowledge in the same context. Instead, it may involve small steps, including the recognition of the 'biases' that each approach introduces into the interpretation of the research findings.

In conclusion, the gaps noted in this paper are the starting point for further discussion on the road ahead. The 'life course' of FASD provides one lens to illuminating those gaps, but other approaches and other experiences may also play important roles.

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