USING A COMMON FORM FOR CONSISTENT COLLECTION AND REPORTING OF FASD DATA FROM ACROSS CANADA: A FEASIBILITY STUDY

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ABSTRACT

Background
This study was undertaken to determine the feasibility of collecting information on individuals newly diagnosed with Fetal Alcohol Spectrum Disorder (FASD) in multi-disciplinary diagnostic programs across Canada.

Objective
To determine the frequencies of specific diagnoses within the spectrum, the frequencies and patterns of specific functional deficits, and the range of recommendations made for intervention and management for children and adults.

Methods
All qualifying clinics in Canada were invited to join this project and complete questionnaires on the patients that were seen during the research period.

Results
Over half of all clinics participated (25/45) and submitted the information requested on 307 individuals, ranging in age from 1 to 42 years. Two hundred and eighty-nine individuals had a diagnosis of FASD and were analysed further. The percent of individuals with Fetal Alcohol Syndrome was 2.1% of those with FASD diagnoses, which was lower than expected based on the literature. The level of disability among the entire FASD was always significant with at least 3 domains measured as severely impaired via the criteria for diagnosis but almost one-quarter were extremely disabled with 6 of a possible 9 brain domains measured significantly impaired. No specific patterns of functional disability were found to represent any significant subgroup of the patients. An average of 13 new recommendations for intervention and management were made for each patient in health, mental health, social services, and education.

Conclusion
Although this was a pilot study with a relatively small sample, it is the largest collection of cases of FASD from multiple sites in one country ever published to our knowledge. It illustrates that important patient information can be collected across clinical programs considering the diagnosis of FASD but only with financial support for time and personnel. Using the methodology of a common data form, consistent data collection can be achieved and patterns and trends can be identified that can help with assuring consistency in diagnosis and with planning for improved patient outcomes.

Key Words and Abbreviations: *FASD- Fetal alcohol spectrum disorder, FAS- Fetal alcohol syndrome, pFAS- Partial fetal alcohol syndrome, ARND- Alcohol related neurodevelopmental disorder*

The clinical expressions of Fetal Alcohol Spectrum Disorder (FASD) have been characterized by combinations of specific facial abnormalities, growth deficiencies, and central nervous system (CNS) dysfunction, in the context of prenatal exposure to alcohol since the original descriptions of the condition in the 1970's.[1,2,3] The specific facial features plus the CNS dysfunction are required for the diagnoses of the "Fetal Alcohol Syndrome" (FAS) and "Partial Fetal Alcohol Syndrome" (pFAS). When the same patterns of CNS dysfunction are found in individuals exposed prenatally to alcohol but without any of the physical features of FAS or pFAS, the term "alcohol-related neurodevelopmental disorder" (ARND) has been most commonly used.[2,3] More current nomenclature use FASD as an umbrella term, under which the specific diagnoses lie.[4]

It has been estimated that approximately 1% of Canadians have FASD[5] although a slightly higher prevalence rate of 1.2% was found more recently.[6]

The diagnosis of FASD is complex and involves a multi-disciplinary team of experts; the clinical expressions of CNS abnormalities can vary widely and may require neurological, psychological, speech, neuro-motor or psychiatric assessments for recognition and definition. There are many different clinics with a variety of clinical team compositions across Canada that provide specialized FASD diagnostic services. Two diagnostic systems are most commonly followed: Fetal Alcohol Spectrum Disorder: Canadian Guidelines for Diagnosis[3] and the University of Washington, Diagnostic Guide for Fetal Alcohol Syndrome and Related Conditions – the 4-Digit Diagnostic Code.[2] The Canadian Guidelines were developed as a refinement of the Washington system, but the intent of both was to bring consistency to the diagnostic process and specificity to the diagnoses that were reached.

This extensive differential-diagnostic process identifies when prenatal alcohol exposure ought to be considered a significant contributor to the observed deficits that cannot be explained in whole or in part by other etiologies. The diagnostic evaluation immediately serves as a template for improved interventions and management. However, to date, no clinical study has ever been done across communities in any country to quantify patterns and frequencies of specific dysfunctions, or the commonalities of interventions that are recommended for management within this population. Because of the high prevalence of FASD and its long term impacts on those who are affected, it is important for planning diagnostic services and improving patient care to better understand how FASD is being diagnosed and the diagnostic profile of individuals across the lifespan.

In 2008, the Canada FASD Research Network (then named the Canada Northwest FASD Research Network) identified all the clinics in Canada that purported to routinely carry out interdisciplinary diagnoses for fetal alcohol spectrum disorder. Only 27 clinical programs were identified in seven jurisdictions. Combined, they had clinical appointment slots available to evaluate less than 1000 clients per year country-wide.[7] A requirement for a common data collection tool to provide a national perspective on the frequency of the different diagnoses within the fetal alcohol spectrum, the frequencies and patterns of functional disability and the patterns of management recommendations emerged.

The goals of this study were then to:

1) demonstrate the feasibility of asking busy clinical teams to voluntarily change their procedures so that they could obtain appropriate patient permissions and complete standardized data collection and reporting tools

2) collect data that could provide a national perspective on the frequency of the different diagnoses within FASD, the frequencies and patterns of functional disability and the patterns of management recommendations for children and adults.

METHOD

This study was funded through a contribution agreement with the Public Health Agency of Canada in May 2011 and all data collection was concluded in March of 2014. Forty-five clinical programs were now identified in 2010 as performing regular diagnostic assessments for FASD using the multi-disciplinary team approach that is considered the standard of care in Canada. These programs varied by province in the size of their gen-

eral referral areas and in their capacities to see new individuals. Use of either the Canadian Guidelines or the Washington Guide permitted the same data elements to be measured and collected. The few clinics that did not use either system were excluded from the study.

Participating clinics met in person and by teleconference to determine the approach and data elements to be standardized for collections. A questionnaire was designed to reflect the clinical work and information that was already being commonly collected by diagnostic clinics with respect to the diagnoses made, the functional diagnoses, and the specific management recommendations that were offered. Participants reviewed the data forms for flow and content and every eligible clinic was represented, either directly or through clinical groups.

Data forms and instruction manuals were developed for children (up to age 17) and for adults ($\geq$ age 18) separately and included many similar components as well as a few age dependent questions. Manuals were developed to clarify each potential entry to avoid needless errors or misinterpretations; thereby streamlining data to be collected more consistently. The information in the forms included:

- Demographic information
- The specific alcohol-related diagnosis
- The four–digit code that was associated with the diagnosis
- The diagnostic scheme that was used.
- Brain evaluations according to the domains of function as defined in the Canadian Guidelines (and easily converted from the Washington Guide)
- The functional levels of ability within key subdomains, according to psychometric tests.
- The management recommendations that were made in health, mental health, education and social services.
- Other brain diagnoses that were noted (via history or examination).
- Secondary disabilities and other deficits/problems (adult form only).

Descriptive analyses were used to characterize demographic information, brain domains and subdomains of impairments, management profiles and secondary disability or associated problems. Association rule learning technique[8] was used from R analytic software to find patterns of subdomain impairments that occur together and patterns of management recommendations/referrals in respective analyses that are given together. This statistical technique is commonly used to find items (e.g. impairment types) that tend to occur together in a population, such as, which items are commonly purchased together in a grocery store. Associations are quantified by support (proportion of the sample that have the particular domain/referral single/pair/set) and confidence (strength of the rule or how often it is true). Support was set to a minimum of 0.4 (40% of individuals) and confidence was 0.6 (at least 60% of the time) for pattern investigations. Support and confidence are identical for single values (one item in a group) thus only support is reported for significant single conditions. Support is reported simply as percent of the sample along with confidence values.

Diagnostic information was only filled in for domains and subdomains of impairments as tested. Others were marked as “not assessed” or were simply left blank. There is no clinical reason for all individuals to be tested in every domain for diagnostic purposes. For example, only very low scores in three domains are required for a diagnosis of CNS impairment in the systems used, so a clinician may have only completed testing in the first three that were found to be severe or profoundly impaired; thereby not requiring to progress through the remaining tests or domains. The form did not include space to explain why data was missing. It is possible that there was no clinical concern in those areas that would have warranted testing. It is also possible that for time and cost constraints, the professionals just stopped when they believed that they had adequate information regardless of achieving a full or complete picture. Nevertheless, no one domain had a substantial proportion of the missing values. A majority of subjects had at least one missing data point across the nine domains. When analyzing across types of impairments or management referrals, it

was thus necessary to include missing values in the denominator. Missing/unknown values were excluded from all other analyses (demographic and secondary disabilities). The additional sections for secondary disabilities in the adult form were optional for completion and it was assumed that the missing values had a similar distribution as the responses that were complete.

Ethics permission was obtained by each site to participate. Families and clients were asked to give permission for their data to be collected and forms were completed by clinical staff.

Clinforma Data Systems and Project Management, Fig P Software Incorporated provided the FASDnet data system for data entry and data management by the participating clinical sites. Completed forms were submitted confidentially to ClinForma via email or fax. Unique patient codes were provided for the clinics' use. The references that linked the individuals to the codes were held by the clinics and only select demographic data (age, sex, living situation, and the clinical site) were submitted with the outcome data. Data were transferred to the Alberta Centre for Child, Family, and Community Research for analysis and management of the data.

RESULTS

Of the 45 clinical programs that were identified and initially show interest in the project, only 25 clinics received administrative permission to participate and successfully obtained appropriate ethics committee approval. Each of these clinics developed an organizational pathway to incorporate the FASData form into their clinical work, and submitted data after caregiver consent and/or patient assent.

Of the remaining 20 programs, 15 were still in the continuum of becoming prepared to participate by the end of the study. The remaining five programs had formally declined to participate.

During the 15.5 months of open submission, 307 data forms were submitted. Of these, 231 were submitted on the child form and 76 were submitted on the adult form. Of these 307 individuals, 289 had confirmed diagnoses within the FASD spectrum, 218 children (94%) and 71 adults (93%). Diagnosis was deferred for eighteen (13 children and 5 adults) who were removed from the dataset before all other analyses were conducted.

The absolute number of individuals diagnosed with FASD in the clinics that participated cannot be determined due to a number of factors. An agreement was reached with the primary ethics panel at the University of British Columbia (that was subsequently followed by all jurisdictions) that stipulated:

1) Data on individuals who were assessed for FASD and found to have an alternate condition was not allowed to be collected;

2) The number of individuals/families that declined to have their information included were not to be counted; and

3) The number of cases that could have been submitted but were not because of administrative reasons was unknown. Thus, the full number of FASD diagnoses cannot be determined in this study.

Even with these limitations, this remains the largest multi-site/multi-jurisdictional clinical sample of individuals with FASD ever assembled, to our knowledge, in Canada.

Demographic Distributions

Four Canadian provinces provided data: British Columbia, Alberta, Manitoba, and Ontario Almost half of the children were aged 6 to 10 years of age at time of assessment (45%) and another third were under age 15 (29%). Adults ranged from 18 to 42 years of age with 64% being 18 to 26 years of age. There were equal numbers of males and females. Over half of the children were living with a parent or other guardian (58%) and a substantial proportion were living in in foster care (39%). The remainder of the children lived in supported living, in custody, or unknown living situations. Almost half of the adults lived independently (45%), another 38% lived with a caregiver, parent, or other guardian, and the remaining 16% were living in either supported living, in custody, were homeless, or in unknown living situations (Table 1).

TABLE 1 Demographic characteristics of individuals with FASD

	Child patients			Adult patients		
Characteristic	**N**	**Total**	**%**	**N**	**Total**	**%**
Age group (years)						
1 to 5	24	218	11.0%			
6 to 10	97	218	44.5%			
11 to 15	63	218	28.9%			
16 to 19	34	218	15.6%			
18 to 20				24	70	34.3%
21 to 25				21	70	30.0%
26 to 42				25	70	35.7%
Gender						
Male	116	218	53.2%	36	71	50.7%
Female	102	218	46.8%	35	71	49.3%
Province						
British Columbia	36	218	16.5%	low n	71	< 5%
Alberta	102	218	46.8%	46	71	64.8%
Manitoba	45	218	20.6%	low n	71	< 5%
Ontario	48	218	22.0%	23	71	32.4%
Living arrangement						
Caregiver, parent or other guardian	126	218	57.8%	27	71	38.0%
Foster care	86	218	39.4%			
Supported living, custody, homeless or unknown	6		2.8%	11	71	15.5%
Independent				32	71	45.1%

Specific Diagnoses within the FASD Spectrum

Diagnoses were generally recorded using terminology as suggested by the Canadian Guidelines, however, at times the University of Washington system was also in use. Among the completed child forms, the Canadian Guidelines were used for 166 cases and the Washington Guide for 48 cases. Among adult forms, the Canadian Guidelines were used for 40 cases, whereas, the Washington Guide was used for 30 cases. The guideline selection was not filled out for five individuals across forms (child or adult). Overall, the Canadian Guidelines were used in 73 percent of the assessments (where reported).

Among those assessed using the child or the adult forms (N= 289), only 6 FAS diagnoses were made. This accounts for 2.1% of those with a diagnosis of any FASD. The majority of individuals received a diagnosis of Alcohol-Related Neurodevelopmental Disorder (ARND; 181 children and 61 adults) followed by partial FAS (pFAS; 31 children and 8 adults).

Physical Characteristics: Growth Deficiencies and Facial Anomalies

Among children, 2.3% were less than or equal to the 3rd percentile on both height (3.8%) and weight (5.1%), 7.4% were less than or equal to the 10th percentile on both growth measures, and another 7.4% had one criteria less than or equal to the 10th percentile. All these measures are less than what would be expected in an average population (e.g. 3% should be below 3rd percentile for average children by definition). Individuals with FASD were no more likely to fall below the average growth estimates. There were not enough adults to report the number below the 3rd percentile for both height (7.1%) and weight (7.1%) but 8.6% had both criteria less than or equal to the 10th percentile (Table 2).

Of the three cardinal facial features of FAS, the majority of individuals had none (56% child; 48% adult), or only one (27% child; 32% adult). Among children, 13% had two facial features and 4% had all three. There were very few adults with all three features to report; therefore, the cases with either two or three facial features was combined (20%; Table 4).

Among the children with some or all of the sentinel facial features; philtral smoothness and upper lip thinness were more likely to occur together (14.4%) than philtrum smoothness and palpebral fissure length (6.3%), or upper lip thinness and palpebral fissure length (5.3%). Fewer adults had significant facial features, although 14.4% had both philtrum smoothness and palpebral fissure length and 7.2% had both philtrum smoothness and upper lip thinness. There were not enough adults with both upper lip thinness and palpebral fissure length to report (n< 5; Table 2).

TABLE 2 Growth features associated with FAS

	Child patients			Adult patients		
Characteristic	**N**	**Total**	**%**	**N**	**Total**	**%**
Growth height						
< 3rd percentile	8	213	3.8%	5	70	7.1%
> 3rd & < 10th percentile	18	213	8.5%	6	70	8.6%
> 10th percentile	187	213	87.8%	59	70	84.3%
Growth weight						
< 3rd percentile	11	214	5.1%	5	70	7.1%
> 3rd & < 10th percentile	18	214	8.4%	5	70	7.1%
> 10th percentile	185	214	86.4%	60	70	85.7%
Growth both height and weight						
Both < 3rd percentile	5	215	2.3%			
Both > 3rd and < 10th percentile	16	215	7.4%			
One < 10th percentile	16	215	7.4%	9	70	12.9%
Both > 10th percentile	178	215	82.8%	55	70	78.6%
Both < 10th percentile[1]				6	70	8.6%

1. Combined values for adults due to low n

TABLE 3 Sentinel facial features associated with FAS

	Child patients			Adult patients		
Characteristic	**N**	**Total**	**%**	**N**	**Total**	**%**
Number of face features						
None	116	208	55.8%	33	69	47.8%
One	56	208	26.9%	22	69	31.9%
Two	27	208	13.0%			
Three	9	208	4.3%			
Two or three[1]				14	69	20.3%
Palpebral fissure length						
< -2 SD	37	213	17.4%	25	69	36.2%
> -2 SD & < -1 SD	49	213	23.0%	11	69	15.9%
> -1 SD	127	213	59.6%	33	69	47.8%
Philtrum smoothness						
4 or 5	55	209	26.3%	20	69	29.0%
3	64	209	30.6%	23	69	33.3%
1 or 2	90	209	43.1%	26	69	37.7%
Upper lip thinness						
4 or 5	45	208	21.6%	7	69	10.1%
3	58	208	27.9%	32	69	46.4%
1 or 2	105	208	50.5%	30	69	43.5%
Pairs of face criteria						
Palpebral fissure length and philtrum smoothness	13	208	6.3%	10	69	14.5%
Palpebral fissure length and upper lip thinness	11	208	5.3%	low n	69	< 5%
Philtrum smoothness and upper lip thinness	30	208	14.4%	5	69	7.2%

1. Combined values for adults due to low n

Central Nervous System Dysfunction

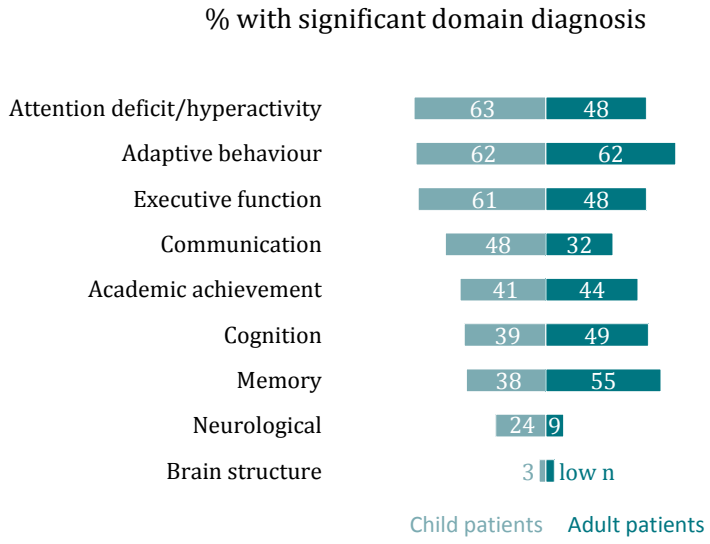
In most circumstances, the Canadian Guidelines and the Washington Guide define CNS dysfunction as a component of FASD when these problems are not likely due to other specific prenatal causes or postnatal life experiences, but is due to persistent diffuse brain damage that can be detected through physical evaluation of the brain (images or physical findings) and/or finding significant abnormalities in at least 3 domains of brain function (out of a possible 9). A significant abnormality is further defined as a score at or below 2 standard deviations from the mean. This is equivalent to performance within the lowest 2 to 3 % of same-aged individuals in that domain. In some cases, the sub-diagnosis of brain damage/dysfunction is made through neuroimaging or neurological findings of abnormality. When using the Washington Guide, a diagnosis of an FASD may still be given in the presence of positive neurological findings and less extreme neuropsychological scores, since the neurological findings are considered to be other clear evidence of structural brain damage.[2]

The performance deficits for the FASD population in this study as a whole varied widely in the specific areas and severity of deficit as well as the impact on the individuals' lives. Twenty-two percent of children and 24% of adults had 6 to 9 of the 9 possible brain domains significantly impaired. Approximately half of the children (53%) and half of the adults (42%) had 3 to 5 significantly impaired brain domains. The remaining 25% of children (n= 54) and 34% of adult s (n= 24) had less than 3 significantly impaired brain domains.

As to the question of whether there was a profile or profiles of functional deficits common to the majority of the cases diagnosed, association rule learning procedure revealed that no pattern(s) of either impaired domains or of impairment on functional tests were common among more than half of the individuals. With minimum support and confidence set to 0.4 and 0.6 respectively, the brain domains impaired in a significant number of children were attention deficit/hyperactivity (63% of individuals), adaptive behaviour (61%), executive function/abstract reasoning (61%), communication (48%), and academic achievement (42%). Three patterns of combinations of domains were also significant for children; executive function and adaptive behaviour (44%, confidence = 0.73), adaptive behaviour and attention deficit/hyperactivity (44%, confidence = 0.71), and executive function and attention deficit/hyperactivity (42%, confidence = 0.70). Sixty-five percent of children had substantial impairments in one of these three sets of domains (Figure 1).

Brain domains impaired in a significant number of adults were as follows: adaptive (62%), memory (55%), cognition (IQ; 49), attention deficit/hyperactivity (48%), executive function/abstract reasoning (48%), and academic achievement (44%; Figure 1). There were also three sets of significant domains among adults: cognition and memory (42%, confidence = 0.86), memory and adaptive behaviour (42%, confidence = 0.77) and attention deficit/hyperactivity and adaptive behaviour (41%, confidence = 0.85). Sixty-one percent of adults had substantial impairments in one of these 3 sets of domains (Figure 1).

FIG. 1 Percentage with severely or profoundly impaired brain domains by domain type and age group



Psychometric tests of function were used to determine severity of brain domain impairments. There were 10 significant subdomains in children: adaptive behaviour (60%), inattention (54%), planning/problem solving (51%), working memory (50%), attention deficit/hyperactivity (49%), inhibition (48%), flexibility (47%), and math (42%). There was only one pair of psychometric tests that accounted for at least 40% of the population: attention deficit/hyperactivity and inattention (46%, confidence = 0.93).

Adults had a slightly different set of common subdomains: auditory memory (56%), working memory (53%), adaptive behaviour (53%), verbal cognition (50%), overall cognition (46%), overall memory (44%), math (44%), and inhibition (43%). Two pairs of psychometric tests were also significant: overall memory and auditory memory (43%, confidence = 0.97), and working memory and auditory memory (40%, confidence = 0.76). Forty-nine percent of adults were profound or severely impaired on one of these two pairs of psychometric tests.

There were no sets of greater than two domains or pairs of psychometric tests that accounted for more than a small fraction of the population. The top 3 sets of 3 domains (the minimum number usually required for diagnosis) each accounted for 40 to 44% of children and only 65% had one of these top 3 sets. Among adults, the top 3 sets of 3 domains accounted for 41 to 42% each but only 61% of adults had one of the 3 sets of domains significantly impaired.

Recommendations For Management Made By the Clinical Teams

The forms included the itemization of recommendations that were made to health systems, support programs and services, mental health counseling, legal system service, and directions for new supports or services for care givers after the diagnostic procedures were completed. In general, clinicians made many recommendations for management, suggesting a very high level of functional disability and need in the population that was not being met prior to assessment. Sixty-nine percent of children and 76% of adults had 10 or more specific recommendations while the remainder received fewer. There was an option for clinicians to select “would have given a recommendation but that the service was not available.” One or more other recommendations was rated as “Would

have recommended but not available" for 33% of children and 39% of adults.

There was little variability between individuals (of each age group) in management recommendations despite wide range in specific patterns of disability or severity of that disability. The most frequent recommendations for children were educational supports (88%), educational modifications (90%), speech-language therapy (61%), school-based learning (61%), mental health counseling (57%), social/life skill programs (57%), recreational services (57%), occupational therapy (56%), FASD advocacy (55%), and long-term, stable, enriched home environment (55%). There were many significant sets of recommendations and the highest three sets of recommendations were educational supports and educational modifications (82%, confidence= 0.94), school-based learning and educational supports (59%, confidence= 0.97), and school-based learning and educational modifications (60%, confidence= 0.98). Eighty-four percent of children were referred to one of these top three pairs of disorder management types. Of these recommendations 59% were for new interventions or approaches while 30% endorsed ongoing programing, and 11% were for new programming that was known to be unavailable.

Sets of 3 recommendations that at least half of children received were educational modifications, school-based learning, and educational supports, (58%, confidence = 0.98); educational modifications, speech-language therapy, and educational supports (53%, confidence = 0.94); educational modifications, recreational services, and educational supports (54%, confidence = 0.98); and educational supports, long-term stable enriched home environment, and educational modifications (50%, confidence = 0.98). Seventy-nine percent of children received one of these 4 sets of referrals/recommendations.

The top recommendations or referrals for a significant number of adults were: disability income support (82%), recreational services (58%), FASD advocacy (58%), employment accommodations (56%), life skills program (56%), family physician (54%), FASD mentorship (52%), individual mental health counselling (52%), supported employment (52%) and stable safe living situation (51%). The top sets of recommendations were: disability income support and recreational services (54%, confidence= 0.66), disability income support and FASD advocacy (54%, confidence= 0.66), and individual mental health counseling and disability income support (46%, confidence= 0.89). Seventy-six percent of adults had one of the top pairs of management referrals/recommendations. There were no sets of three referrals/recommendations that at least half of the adults received. Of these recommendations 55% were for new interventions or approaches while 22% endorsed ongoing programing, and 23% were for new programming that was known to be unavailable. The final category was twice as high as it had been for the children and adolescents.

The type of management referred did not vary much by the type of domains impaired. Tables 4 and 5 show the range of percentage impaired in seven of the nine brain domains (neurological signs and brain structure omitted due to low numbers) by the top 10 management referrals per age group. For example 88% of children with significant impairment in attention deficit/hyperactivity were referred to educational supports compared to 92% of individuals with academic impairments. The rest of the individuals in other impaired domains were referred at a rate between 88 and 92%. Note that significant brain domain impairments are not mutually exclusive (individuals more often than not have impairments in more than one domain).

TABLE 4 Percentage of child patients referred to management by type of management and the top 7 domains.

	Domains						
Management	Attention deficit/ hyperactivity	Adaptive behaviour	Executive function	Communication	Academic achievement	Memory	Cognition
Educational supports	88%	89%	92%	93%	92%	90%	93%
Educational modifications	79%	80%	82%	84%	87%	84%	84%
Speech-language therapy	61%	58%	60%	71%	59%	54%	65%
School-based learning	56%	61%	61%	66%	64%	65%	60%
Recreational services	55%	53%	60%	54%	63%	63%	58%
Long-term, stable, enriched home environment	55%	57%	54%	51%	51%	53%	48%
FASD advocacy	48%	54%	50%	43%	46%	47%	48%
Occupational therapy	42%	45%	46%	50%	39%	42%	47%
Social and life skill programs	38%	43%	47%	43%	40%	51%	47%
Individual MH	43%	44%	44%	35%	44%	41%	34%

TABLE 5 Percentage of adult patients referred to management by type of management and the top 7 domains.

	Domains						
Management	Attention deficit/ hyperactivity	Adaptive behaviour	Executive function	Communication	Academic achievement	Memory	Cognition
Disability income support	85%	84%	94%	96%	90%	90%	97%
Recreational services	53%	55%	68%	65%	61%	62%	69%
FASD advocacy	65%	59%	68%	78%	71%	67%	69%
Individual Mental health	44%	48%	50%	30%	35%	41%	46%
Stable, safe living situation	53%	50%	56%	48%	55%	46%	54%
Supported employment	38%	45%	47%	43%	45%	46%	51%
FASD mentorship	44%	48%	53%	57%	61%	54%	60%
Family physician	32%	39%	29%	35%	32%	38%	43%
Employment accomodations	35%	43%	35%	35%	35%	44%	49%
Life skill programs	35%	36%	47%	48%	52%	44%	54%

"Other Brain Diagnoses"

Most developmental diagnoses made within the CNS are referred to by their specific patterns of functional abnormality. This is different than FASD which is an etiologic diagnosis that may include any and all evidence of brain dysfunction. It would not be unexpected that other specific brain based diagnoses would be found in individuals with FASD and therefore, these should be seen as expressions of FASD component parts in most cases and not co-morbid conditions.

Children had a number of other brain-based diagnoses present or suspected. Eighty percent had ADHD, 68% had a language disorder, 52% had a learning disorder, 45% had an intellectual disability, 43% had a mood disorder, 34% had a sleep disorder, 14% had a developmental coordination disorder and 18% had another brain diagnosis. Six other brain diagnoses were reported for adults (present or suspected). Sixty-two percent of adults had an intellectual disability, 55% had ADHD, 49% had a learning disability, 43%

had a language disorder, 20% had another brain diagnosis, and less than five individuals had a developmental coordination disorder.

Life Challenges

There were a number of additional questions in the adult survey to further assess challenges specific to adults including; substance use, secondary disabilities, daily living skills and social skills deficits. Other sections (health and mental health) were included in the survey, but responses were not filled in or were unknown for too many individuals to reliably assess the results.

Forty-six percent of adults had an IQ less than 70, another 44% had an IQ of 70 to 84, and 10% had an IQ of 85 to 100. Three out of every 4 adult females had one or more pregnancies. Alcohol problems were the most common substance use problem with 86% having ongoing or past use followed by 73% with marijuana use, 59% with tobacco use, and 48% using other drugs. Almost all adults had past or current trouble getting or keeping a job (less than five individuals had no job troubles). Most individuals (89%) required past or current help living on their own or required assisted or sheltered housing (75%). Over one in 4 (28%) of those requiring housing assistance were homeless at the time of assessment or in the past. Eighty-five percent had no high school diploma and 80% lacked functional literacy and numeracy skills. Sixty-three percent of adults were a legal offender and half were a legal victim (50%). Many of the individuals were institutionalized at the time of assessment or in the past (57% hospital, and 40% jail).

All adults had one or more challenges in daily living skills; managing medications (89%), managing finances (86%), daily schedule (83%), managing shopping (81%), managing meals (78%), managing cleaning (74%), managing laundry (74%), transportation (68%), and/or hygiene/grooming (60%). Almost with information recorded had social skills deficits in work, personal, and/or self (less than five patients had no deficits). A number of individuals experienced family abuse problems as a victim (83%), aggressor (53%), both victim and aggressor (50%) or other (57%). Parenting problems were observed in the following areas: discipline (64%), neglect (35%), and children removed (25%).

DISCUSSION

Although this was an initial feasibility pilot study with a relatively low sample size, the outcomes and the data are still important to report and lend valuable information to the field. Future efforts will strive to refine processes and approaches, based on these results and experiences.

Canada's Diagnostic Context

Given the estimated demand for diagnoses, the capacity of clinics were surprisingly low. In 2008, only 27 clinical programs were identified in 7 jurisdictions with enough combined clinical appointment slots available to evaluate less than 1,000 clients per year across Canada.[7]

It was estimated, based on informal discussions with clinical leads that between 1,500 and 2,000 clinical opportunities for FASD diagnosis were then available by 2010 in 45 clinical programs. This was a substantial increase in 2 years based on the same discussions. Of course, not all individuals evaluated for FASD are found to have FASD, and not all available clinical opportunities are used for FASD diagnosis but may instead have been used for individuals requiring assessment for another neurodevelopmental disorder. It remains uncertain as to how many new diagnoses of an FASD are made annually in Canada.

Some provinces have concentrated FASD diagnostics in a few large regional centres, (i.e. Manitoba and British Columbia) or multiple small community based programs (i.e. Alberta). Diagnostic capacity has also been fluid across the country; new program sites open and others close, often due to funding cycles. The precise number of active program sites at any given moment is challenging to determine. The most consistent FASD diagnostic services have been and remain in British Columbia, Alberta, Manitoba, and Ontario. Saskatchewan has maintained a smaller diagnostic presence. New Brunswick has been in the process of development of an FASD clinical service for the last several years. The Yukon has had clinical services in Whitehorse that have been staffed primarily by traveling clinicians from the

southern provinces. The Northwest Territories and Nunavut tend to refer individuals south for diagnoses when that has been deemed necessary.

FASD Diagnosis in General

Unfortunately, the sample size for those who were assessed but did not receive an FASD-related diagnosis was too small to analyze; comparing and contrasting characteristics and patterns of this with a confirmed PAE-related diagnosis with those who did not meet criteria would have been fascinating. Nor had any of the adults being assessed previously received a diagnosis; determining if diagnoses remained impervious to time is another important quality assurance factor.

It has been estimated that the diagnosis of FAS is made in about 10 percent of diagnoses of the full fetal alcohol spectrum.[9,10] In this study, FAS was diagnosed in only 2.1% of individuals. Furthermore, it was surprising that over one quarter of the individuals did not meet what is usually considered the 'standard' threshold for diagnosis. Of these 54 children and 24 adults, 22 children and 10 adults fell within the guidelines due to exceptions to the standard rule as indicated in the guidelines; being under age 6 (children only), or having structural or neurological impairments (which counts as one domain in the Canadian Guidelines and considered sufficient for a diagnosis in the Washington Guide as "Neurobehavioral Disorder: Alcohol Exposed"). The Washington Guide allows for a lower threshold of functional impairment than the category Alcohol-Related Neurodevelopmental Disorder in the Canadian Guidelines. Remaining are 32 children (15%) and 14 adults (20%) with discrepancies between the diagnosis given and the testing results that justified the diagnosis. To clarify, the testing as presented often placed the subject within the lowest 10% of individuals using the Washington Guide rather than the lowest 2 to 3 % that is required for an FASD diagnosis using the Canadian Guidelines. Although many clinicians accept that there is occasionally a need for judgment in the process of diagnosis, the rate of such non-standard cases in the results appeared to be high. This raises the possibility that clinicians misunderstand or misremember the stated diagnostic rules or routinely stretched them to cover borderline cases. If this is the case, it is possible that diagnostic guidelines and policy could require some refinement better accommodate individuals in need of services.

CNS

Data from this study suggest that gestational alcohol exposure is associated with diffuse brain dysfunction alone much more often than with the inclusion of the growth and facial stigmata. Growth deficiencies did not appear to be a predictor of FASD in this sample. At the same time, the CNS performance deficits varied widely. A quarter of the individuals had very severe performance deficits in 6 to 9 of the brain domains – significantly more impairment than is requited for a diagnosis of FASD and more disability than usually thought typical of those with an FASD.

Animal experimentation and modern brain imaging techniques have demonstrated that the most typical forms of brain damage, or alteration associated with ethanol teratogenesis, are at a microcellular or microstructural level and occur throughout the brain.[11,12,13,14,15,16,17] It is not surprising that diffuse damage caused at different times during brain development would result in variable patterns of decreased performances. Based on this large volume of research, a single pattern of performance deficit was not expected. However it was thought to be possible that a small number of patterns might emerge that would be representative of a large part of the population. This was not the case; the individuals with FASD showed high variability in the specific functional problems that led to their overall disability.

These two findings of very infrequent facial stigmata and high variability in specific performance deficits together emphasize why FASD is such a hidden disability. But that hardly suggests that these individuals do not have unmet needs. Management recommendations were remarkably similar across different domains of impairment. Multiple academic interventions were recommended for most children, suggesting that this relatively common group of individuals is also extremely needy in school-based settings. In adults, a lack of funds and appropriate supports lead to high rates of adaptive dysfunction. Adults had a significant number of secondary disabilities that did not appear to be reflected in the types of

management referrals recommended perhaps due to the availability of specific services. These findings suggest that intervention/management programs do not seem to require great specificity to brain functional profiles for implementation.

It is clear that the legal, ethical, and administrative processing that is necessary to conduct this type of clinical study across jurisdictional lines is possible but arduous and may limit the questions asked. Without funding for the sites however, it is necessary to rely on good will and enthusiasm to gain collaboration. Delays are to be expected and there is a limit to how hard researchers can push their time-strapped clinical colleagues. A longer time line should be planned for further work of this type. Clearly funding support to the programs would have also permitted clinical time to attend to the form completion and submission.

This study demonstrates that the experience of evaluating individuals for FASD is similar across diagnostic programs across provincial boundaries. Data collection, while highly valuable, is constrained by ethical considerations time constraints and financial concerns at the clinic sites. Still, these constraints are not impossible to work through and the results are obviously helpful. The medical world is shifting as best it can to an evidence-based approach. Evidence for many common conditions must be, as in this case, actively sought. The continued collection of multisite data would obviously be useful in demonstrating the disabilities and needs of this large population and help to improve service delivery.

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Conflict Of Interest Guidelines

All authors have no commercial associations or other arrangements (e.g., financial compensation, potential to profit, consultancy, stock ownership, honoraria, patent-licensing arrangement, etc.) that might pose a conflict of interest in connection with the submitted article.

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APPENDIX 1

FASD CLINICS IN CANADA WHO SUBMITTED DATA FORMS

Diagnostic Clinic Name	Location
Sunny Hill Health Centre for Children; Complex Developmental Behavioural Conditions Team	*Vancouver , BC*
Asante Centre; Diagnostic and Assessment Services, Fetal Alcohol Spectrum Disorder (FASD) Society for British Columbia	Maple Ridge, BC
Fraser Developmental Clinic; Harper and Associates	*New Westminster, BC*
Lakeland Centre for FASD	*Cold Lake, AB*
NWC Alberta FASD Services Network	*Barrhea, AB*
FASD Clinical Services, Glenrose Rehabilitation Hospital	*Edmonton, AB*
Central Alberta FASD Clinic, Family Services of Central Alberta	*Red Deer, AB*
FAS Diagnostic Clinic, MediGene Service Inc.	*Calgary, AB*
FASD Assessment and Diagnoses Clinic, Renfrew Educational Services	*Calgary, AB*
FASD Program: Diagnostic Services Accredited Supportive Living Services	*Grimshaw, AB*
Pediatric Specialty Clinic, Alberta Health Services	*Camrose, AB*
South Alberta FASD Assessment and Diagnostic Clinic, South Alberta FASD Network	*Lethbridge, AB*
Fetal Alcohol Spectrum Disorders Diagnostic Clinic Alberta Children's Hospital Child Development Centre	*Calgary, AB*
Northwest Regional FASD Society-Mackenzie Network,	*High Level, AB*
Children's Fetal Alcohol Services and First Steps, Bridges Family Programs	*Medicine Hat, AB*
Children's Hospital of Winnipeg, Manitoba Developmental Pediatrics University of Manitoba Children's Hospital of Winnipeg	*Winnipeg, MB*
Surrey Place Centre FASD Adult Diagnostic Clinic	*Toronto, ON*
FAS Clinic, Motherisk, the Hospital for Sick Children	*Toronto, ON*
Child Development Centre Hotel Dieu Hospital	*Kingston, ON*
Ramsey Lake Health Centre	*Sudbury, ON*
Guelph-Wellington FASD Team, Guelph and Wellington	*Guelph, ON*
Center for children and families in the justice system in the London Family Court Clinic	*London, ON*
NorWest Commodity Health Centre – FASD Program, NorWest Community Health Centres	*Thunder Bay, ON*
Waterloo Region FASD Diagnostic Team	*Waterloo, ON*
Grande River Hospital	*Kitchner, ON*

FASD DIAGNOSTIC CLINICS PARTICIPATING BUT WHO DID NOT SUBMIT DATA FORMS

Diagnostic Clinic Name	Location
Child Development Centre; Complex Developmental and Behavioural Conditions (CDBC) Clinic,	Nanaimo, BC
Queen Alexander Centre for Children's Health; Complex Developmental and Behavioural Conditions (CDBC) Clinic	Victoria, BC
Fraser Valley Child Development Centre	Abbotsford, BC
Northern Health Assessment Network (NHAN) Provincial Health Services Authority	Prince George, B.C.
Centrepoint Young Offender Program,	Edmonton, AB
Northwest Primary Care Network Children and Youth FASD Diagnostic Clinic	High Level, AB
Northern Association for FASD	High Prairie, AB
NEAFAN (North East Assessments) FAS Diagnostic Clinic	Fort McMurray, AB
Regina Community Clinic – FASD Centre	*Regina, SK*
St. Michael's Hospital; La Ka Shing Knowledge Institute	Toronto, ON
Mothercraft/ Breaking the Cycle FASD Assessment and Diagnostic Clinic	Toronto, ON
Fetal Alcohol Spectrum Disorder Associated Youth Services of Peel	Mississauga, ON.
Grandview Children's Centre	Oshawa, ON
Eastern Door Centre – Diagnostic Team, Elsipogtog First Nations – Elsipogtog Health and Wellness Centre	Elsipogtog, NB
New Brunswick Centre of Excellence for FASD/Centre d'excellence sur l'ETCAF au N-B	Moncton, NB