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Canadian clinical capacity for FASD assessment, diagnosis, disclosure and support to children and adolescents: a descriptive study

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Title: Canadian clinical capacity for FASD assessment, diagnosis, disclosure and support to children and adolescents: a descriptive study

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ABBREVIATIONS

- CanDiD - Canadian Interdisciplinary Clinical Capacity to Diagnose FASD study
- CanFASD – Canada FASD Research Network
- FASD – Fetal Alcohol Spectrum Disorder
- NGO - non-governmental organization
- PAE – prenatal alcohol exposure
- SD – standard deviation

ABSTRACT

Objective: Canadian Fetal Alcohol Spectrum Disorder (FASD) guidelines encourage an age-specific interdisciplinary diagnostic approach. However, there is currently no standard-of-care regarding FASD diagnosis disclosure and few studies document Canadian FASD clinical capacity. Our objectives were to describe clinical capacity (defined as skills and resources) for FASD assessment, diagnosis, disclosure, and support in Canada.

Design, setting and participants: Data were drawn from the Candid study, a cross-sectional investigation of Canadian FASD clinical capacity. Data were collected from 41 clinics in 2021 including the proportion of clinics that follow the Canadian interdisciplinary diagnostic guidelines by age group, the presence of a minor patient when the FASD diagnosis is disclosed to parents/guardians, who is responsible for the diagnosis disclosure, use of explanatory tools, and support/counseling services .

Results: Overall, 51%, 32% and 17% of FASD clinics were in Western/Northern, Central and Atlantic Canada, respectively. The number of referrals per year surpassed the number of diagnostic assessments completed in all regions. Approximately 60% of clinics who diagnosed FASD in infants and preschool children followed the interdisciplinary guidelines compared to 80% in clinics who diagnosed school-aged children/adolescents. Diagnostic reporting practices were heterogeneous but most used an explanatory tool with children/adolescents (71%), offered support/counseling (90-95%), and used case-by-case approach (80%) when deciding who would disclose the diagnosis to the child/adolescent and when.

Conclusions: Limited diagnostic capacity and lack of FASD resources across Canada highlights a critical need for continued FASD support. This study identifies gaps in assessment, diagnosis, and reporting practices for FASD in children/adolescents across Canada.

Keywords: FASD; clinical capacity; diagnosis disclosure; children; interdisciplinary team

Strengths and Limitations of this study:

- To our best knowledge, this is the first study that aims to describe Canadian clinical capacity for assessment, diagnosis, disclosure, and support of FASD in children and adolescents.
- Data were drawn from the Canadian Interdisciplinary Clinical Capacity to Diagnose FASD (CanDiD) study – a cross-sectional survey-based study developed by clinicians and researchers at Vitalité Health Network in 2021.
- Data were collected from 41 specialised FASD clinics across Canada (68% of eligible clinics).
- Quantitative data were collected via a telephone/videoconference survey (available in French and English) of clinics who engage in FASD diagnostic assessment of children and adolescent <18 years.

INTRODUCTION

Fetal alcohol spectrum disorder (FASD) is one of the most frequent neurodevelopmental disorders in North America¹ resulting from prenatal alcohol exposure (PAE).² A recent meta-analysis reported that 8% of children exposed to PAE are diagnosed with FASD and that North America has the second highest FASD prevalence after Europe.³ Children exposed to PAE are at high risk of developing FASD regardless of the frequency or amount of alcohol consumed.⁴

Canadian FASD evidence-based diagnostic guidelines (first published by the Public Health Agency of Canada in 2005,⁵ and most recently updated by the Canadian FASD Research Network (CanFASD) in 2016^{2,6}) include recommendations on key components for FASD assessment including screening, referral and support; medical assessment; sentinel facial features; neurodevelopmental assessments; nomenclature and diagnostic criteria; and management and follow-up.⁶ Importantly, these guidelines require an age-specific interdisciplinary diagnostic team approach.⁶ Specifically, the guidelines recommend that the diagnostic team for infants (<18 months) should include a pediatrician/physician and a child development specialist able to conduct physical and functional assessments (i.e., speech-language pathologist, physiotherapist, occupational therapist or clinical psychologist) and that the diagnostic team for pre-schoolers (18mo–5yr) and school-aged children (6-18 yrs) should include a physician, psychologist, speech-language pathologist and an occupational therapist.⁶ Therefore, FASD clinical capacity, defined herein as skills and resources to assess, diagnose, disclose and support FASD, remains complex in many jurisdictions. A previous study reviewed Canadian FASD clinical practices in seven provinces and reported that only 46% of Canadian clinics had a complete multidisciplinary team on-site, but 90% used a team approach for diagnosis and treatment plan.⁷ However, the authors

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2
3 did not specify which health professional participated in the diagnostic assessments by age group.⁷
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5 There are no empirical data on the Canadian clinical capacity for interdisciplinary diagnostic
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7 assessment since the guidelines were updated in 2016.
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12 Skillful diagnosis disclosure and psychological support following a medical diagnosis to children
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14 and adolescents is extremely important to limit psychological trauma,⁸ increase adherence to
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16 treatment plans,⁹ and generally improve quality of life. While Canadian FASD guidelines
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18 recommend that individuals with FASD and their caregivers have access to resources to improve
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20 diagnostic outcomes,⁶ there are currently no specific recommendations regarding the presence of
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22 a minor patient (≤ 18 years) when the FASD diagnosis is disclosed to parents/guardians and, to our
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24 knowledge, no known studies describing clinical practices for FASD diagnosis disclosure to
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26 children - who should make the diagnosis disclosure, under which circumstances (e.g., favorable
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28 environment) and how (e.g., use of explanatory tools).
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35 These knowledge gaps compel a research agenda that aims to describe FASD clinical capacity to
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37 inform clinical and diagnostic protocols to children and adolescents. Specific objectives of this
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39 study were to describe: (i) diagnostic interdisciplinary team composition (< 18 months; ≥ 18
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41 months- ≤ 5 years; $\geq 6-18$ years) and the number of clinics who follow the Canadian
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43 interdisciplinary diagnostic team guidelines by age group; (ii) FASD assessment and diagnosis
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45 reporting practices for children and adolescents including if the minor patient is present during the
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47 diagnosis disclosure, who discloses the diagnosis to the child/adolescent patient, and use of
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49 explanatory tools to facilitate the FASD diagnosis disclosure; and (iii) FASD support and
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counseling provided to patients and families during the diagnosis disclosure and in the 3-month period following the diagnosis.

METHODS

Data were drawn from the Canadian Interdisciplinary Clinical Capacity to Diagnose FASD (CanDiD) study – a cross-sectional survey-based study developed by clinicians and researchers at Vitalité Health Network in 2021. CanDiD is the first investigation that aims to describe Canadian clinical capacity for assessment, diagnosis, disclosure, and support of FASD in children and adolescents. Quantitative data were collected via a telephone/videoconference survey (available in French and English) of Canadian specialty clinics who engage in FASD diagnostic assessment of children and adolescent <18 years. Clinics were identified through membership in the Canadian FASD Research Network (CanFASD), and through an exhaustive Internet search using a purposive sampling (i.e., snowball or network sampling). Introductory recruitment emails were sent to all identified clinics in spring 2021, followed by a phone call one week later to schedule an interview. To increase response proportion, up to 3 follow-up emails (or phone calls) were sent. Semi-structured 20-minutes surveys were completed with a key respondent in each clinic identified by the clinic manager or lead pediatrician who is most knowledgeable with clinical capacity. A copy of the questionnaire was sent to respondents prior to survey completion to allow preparation and consultation with clinic staff. The study was approved by the ethics committee of Vitalité Health Network. Participants provided verbal informed consent.

Study variables

Questionnaire items were developed *de novo* or adapted from the literature.⁶ All items were extensively pre-tested in both French and English with healthcare professionals who work closely with children and families affected by FASD. Variables measured included questions pertaining to: i) general information on the clinic (e.g., location, source(s) of funding, services offered); ii) number of referrals and assessments done per year (used to calculate diagnostic capacity defined as the proportion of assessment completed in a year amongst the mean number of referrals received); iii) interdisciplinary team composition for FASD diagnosis by age group; iv) current diagnosis reporting practices for children/adolescents and use of explanatory tools; and v) immediate and post-diagnosis support and counseling following the diagnosis disclosure. Supplementary Table 1 describes each variable investigated including the item used in the survey, response options, and re-coding of response options for analysis.

Data analysis

Of 78 clinics identified, 18 were excluded (i.e., did not diagnose FASD in children ≤18 years, or had permanently closed), 6 refused to participate, and 13 did not return our recruitment efforts. Forty-one clinics completed the study questionnaire (68% of eligible clinics (41/60)).

To protect the clinics confidentiality, select characteristics were compared in three provincial groupings: *Western and Northern Canada* included clinics located in British Columbia, Alberta, Saskatchewan, Manitoba and Yukon; *Central Canada* included clinics located in Ontario and Quebec; and *Atlantic Canada* included clinics located in New Brunswick, Nova Scotia, Prince Edward Island and Newfoundland. Because of the small sample size, FASD clinical capacity characteristics were reported by age group for all clinics. Means for continuous variables and

frequency distributions for categorical variables were compared. Analyses were performed using SPSS, Version 26.0 (IBM Corp. Released 2012. IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp.).

PATIENT AND PUBLIC INVOLVEMENT

Patients or the public were not involved.

RESULTS

In this study, more than half of respondents were clinic administrators (56%), 15% were physicians/pediatricians, and 29% were other health professionals.

Socio-demographic and clinic characteristics

Half of the FASD clinics were in Western/Northern Canada (51%), and 32% and 17% of clinics were in Central and Atlantic Canada, respectively (compared to 58%, 37% and 5% of non-participating clinics (n=19), respectively). Western/Northern Canada had the highest proportion of clinics that were operational before 2006 (including the oldest clinic in the sample (i.e., operational since 1999)) and Atlantic Canada had the highest proportion of clinics that were operational in the last decade (Table 1). No clinic located in Central or Atlantic Canada was operational before 2005.

Table 1. Socio-demographic and clinical characteristics of Canadian clinics offering FASD diagnosis to children ≤18. CanDiD study, 2021 (n=41).

	FASD Clinics in Canada			
	Western and Northern Canada	Central Canada	Atlantic Canada	Total (all clinics)
Number of clinics, n	21	13	7	41
Year clinic became operational, %				
≤2005	40	25	17	32
2006-2011	40	42	33	39
≥2012	20	33	50	29
Number of referrals per year, mean (SD)	84 (111)	65 (74)	36 (39)	69 (90)
Number of assessments per year, mean (SD)	63 (64)	36 (33)	16 (13)	46 (52)
Source of funding for clinic services, %				
Federal funding	5	15	43	15
Provincial funding	95	85	86	90
Research grant	14	0	14	10
Non-governmental organization (NGO)	33	15	0	22
Other (donations, private funding)	14	46	57	32
Services offered, %				
Prevention	38	31	29	34
Screening	43	69	57	54
Diagnosis	100*	100*	100*	100*
Support to families	86	100	71	88
Specialized FASD training for health professionals	71	54	57	63

*Participating clinics were selected because they engaged in FASD diagnostic assessment of children/adolescent <18 years (i.e., inclusion criteria).

Most regions received provincial funding (85-95%). Proportion of in-kind donations and private funding were highest in Central (46%) and Atlantic Canada (57%). Atlantic clinics reported the highest proportion of federal funding (43%), while Central and Western/Northern clinics reported receiving non-governmental organization (NGO) funding (15% and 33%, respectively; 0 Atlantic clinics reported NGO funding) (Table 1).

Western/Northern clinics received the highest number of FASD referrals per year (mean(sd): 84(111); total of 1503/2537 (59%)) and their diagnostic capacity was highest (75%). Diagnostic capacity in Central and Atlantic clinics was approximately 50% (55% in Central and 44% in Atlantic Canada) (Table 1).

All participating clinics offered diagnosis services (i.e., inclusion criteria for the CanDiD study). Approximately one third of Canadian clinics were involved in prevention efforts (34%), 43-69% of regions were involved in FASD screening, almost two thirds of clinics offered FASD training for health professionals (63%), and most offered support to families (88%) (Table 1).

Assessment and diagnosis by age group

Requirement of the confirmation of PAE or the presence of 3 sentinel facial features for FASD diagnosis assessment was variable among regions: 77%, 67% and 43% of clinics located in Western/Northern, Central and Atlantic regions required this confirmation, respectively.

Only 17% (n=7/41) of clinics diagnosed FASD in infants <18 months; among them, 57% (n=4/7) followed the Canadian interdisciplinary team guidelines (Figure 1). Similarly, 60% (n=15/25) of

clinics who diagnosed FASD in preschool children aged 18 months-5 years followed the interdisciplinary team guidelines. Most clinics (n=40/41) diagnosed FASD in school-aged children (i.e., ≥ 6 -18 years), of which 80% (n=32/40) followed the Canadian guidelines (Figure 1).

Diagnosis disclosure

Designated individuals in charge of the FASD diagnosis disclosure to children/adolescents varied considerably across clinics. In 40% of clinics, pediatricians/physicians were responsible for the disclosure. Other health professionals and clinic administrators were responsible for diagnosis disclosure in 15% and 5% of clinics, respectively. One quarter of clinics used a multidisciplinary team for the disclosure (20% used more than one professional including a physician and 5% used more than one professional excluding a physician). Finally, in 15% of clinics the person responsible for the disclosure varied and often included parents in the diagnosis delivery.

Only 5% of clinics always disclose the diagnosis to children/adolescents at the same time as the parents/guardian disclosure, 15% never included children when first disclosing the diagnosis to parents/guardian, and 80% included the child sometimes, depending on certain factors. Most frequently cited determining factors included parents/guardian request (88%), child developmental age (78%), child chronological age (59%), clinical judgement (78%) and clinic's policy (9%).

Most clinics reported using at least one type of explanatory tools when announcing the diagnosis with parents/guardian (85%) or children/adolescents (71%) (Table 2). In both parents and children, visual tools (pictures, graphs) or communication tools (analogies) were used most often (Table 2).

Table 2. Description of explanatory tools used when announcing the FASD diagnosis to parents/guardian and children/adolescents. CanDiD study, 2021 (n=41).

	Parent/ guardian	Child*
Use of tools when announcing the FASD diagnosis (%)		
Visual tools (pictures, graphs)	73	53
Electronic tools (ipads, computers)	17	13
Communication tools (analogies)	63	53
Use at least one tool	85	71

*Among 38 clinics who disclose the FASD diagnostic to child

Immediate and post-diagnosis support/counseling following diagnosis disclosure

Almost all clinics offered support and counseling to parents/guardian at the time of disclosure (95%) and in the 3-month period following the disclosure (90%). Seventy-one percent and 76% of clinics offered support and counseling to children when announcing the diagnosis and in the 3-month following the disclosure, respectively. While only one third of clinics offered support/counseling to other family members (i.e., siblings, other close family members) at the time of FASD diagnosis disclosure, 65% offered them support/counseling in the 3-month period following the disclosure.

All clinics provided information to families in need post-diagnosis. The information requested most often included information on financial aid programs (95%), mental health programs outside the clinic (98%), support group programs (85%) and information pertaining to Jordan's principal Implementation Act (i.e., ensures equality in health, social and educational services to all First Nations children) (85%).

INTERPRETATION

Early diagnosis, skillful delivery and diagnostic treatment plan and support of FASD is of utmost importance to improve child development¹⁰ and mitigate negative factors associated with its neurodevelopmental impairments including academic failure, substance abuse, poor mental health, problems with law enforcement and maintaining employment.¹⁰⁻¹³ Data from this first Canadian investigation on the clinical capacity for FASD assessment, diagnosis, disclosure and support to children and adolescents revealed that clinics across Canada were not able to respond to FASD referral demands. Health inequality include differences in quality and access to healthcare services.¹⁴ In this study, although the majority (53%) of eligible specialized FASD clinics were situated in Western Canada, this region deserved the largest population of youth <19 years (i.e., approximately 4.9 million compared to 2.7 million in Central Canada and 467K in Atlantic Canada).¹⁵ Consequently, 59% of total requests for FASD consultations were received in Western Canadian clinics. However, it is important to note that their diagnostic capacity was higher than eastern clinics. Equality and equity in health services access is essential to improve health condition, support youth and families and improve quality of life of individuals with FASD. Future studies should investigate FASD prevention and potential FASD health inequality in Canada.

Few clinics assessed and diagnosed FASD in infants and preschool-aged children. Although FASD can be diagnosed at various ages, it is commonly diagnosed when children enter the school-system (>5 years) and fail to attain behavioural and developmental milestones (e.g., motor skills, social skills, language development).^{16,17} Most clinics followed the Canadian interdisciplinary guidelines for infants, preschool and school age children and adolescents (57%, 60% and 80%, respectively). It is noteworthy that following the survey completion, most clinics who did not have a complete

interdisciplinary diagnostic team acknowledged that they did not meet the Canadian guidelines because of limited human and/or financial resources – they were simply doing the “best they could”.

Diagnostic reporting practices were heterogeneous across Canada with the exception that most used some type of explanatory tool when announcing the diagnosis to both parents/caregivers and patients. While physicians were involved in diagnosis disclosure to minor patients in most clinics (60%), approximately one-fifth of clinics entrusted other health professionals with the disclosure, and remaining clinics used administrators, parents, or a variable approach depending on the clinical judgment of the healthcare team. Most clinics (80%) did not systematically include or exclude children/adolescents when announcing the diagnosis to parents/caregivers and used a case-by-case approach when delivering the diagnosis. In a recent viewpoint article, Nunn¹⁸ stated that medical news delivery should be age-appropriate, and in most cases, interaction and delivery is more important than the information itself. Because a child’s chronological age does not necessarily correspond to their developmental age, when and how a diagnosis should be disclosed is also unclear.⁹ Other factors such as cognitive impairments or maturity levels may represent additional barriers when delivering a neurodevelopmental diagnosis.⁹ While existing medical news delivery protocols such as BREAKS¹⁹ or SPIKES²⁰ can offer some guidance, they are not specific to neurodevelopment disorders and do not offer insight for medical news delivery to minors. Although recommendations on reporting practices of neurodevelopmental disorders cannot take a one-size-fits all approach, high heterogeneity in Canadian reporting practices highlight the need for more research to better understand which tools should be used and which health professionals

should make the disclosure to improve clinical and diagnostic delivery protocols for children/adolescents.

Finally, our results indicate that most Canadian clinics supported families during and after the diagnosis disclosure. This is of utmost importance as receiving a life-altering diagnosis can be traumatizing for patients and families.⁹ Prompt and easy access to counseling following a medical diagnosis can impact adherence to treatment plans⁸ and, in turn, impact FASD-related outcomes.

Implications and Future Directions

The CanDiD study provides evidence-base data to identify outstanding knowledge gaps in FASD research and clinical capacity. In addition, our work highlights the imminent need for the development of a National FASD Strategy in Canada. Lack of resources appear to be a serious impediment to healthcare for youth with FASD. Specialized multidisciplinary FASD clinics have the potential to: (i) develop and implement prevention programs to reduce alcohol consumption in pregnant women and in women of child-bearing age; (ii) offer timely resources to individuals and families afflicted by FASD; (iii) provide continued training to health care professionals to ensure proper screening and diagnoses; and (iv) improve health care utilization (and indirectly improve justice and education services) by providing early accurate diagnoses. Consequently, continued national support for FASD clinical capacity including funding for development, training and maintenance of interdisciplinary teams is imperative to improve health services for children living with this life-long difficult condition.⁵

LIMITATIONS

Limitations of this study include that it was impossible to describe inter-provincial differences to protect clinic confidentiality. Because we had no clinical capacity data for non-participating clinics, results may not be generalizable to all clinics in Canada. Finally, although key respondents were appointed by clinic managers or lead pediatricians and represented the person “most knowledgeable about the clinic’s clinical capacities”, data were provided by a single person and may not adequately reflect clinical capacities of each clinic.

CONCLUSION

Results from this study indicate that several clinics across Canada are faced with a lack of FASD resources to meet Canadian interdisciplinary diagnostic guidelines highlighting a need for continued FASD support.

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COMPETING INTERESTS STATEMENT

The authors declare no conflict.

ETHICS APPROVAL

The study was approved by the ethics committee of Vitalité Health Network. Participants provided verbal informed consent.

AUTHORS' CONTRIBUTIONS

ED, DB and NL developed the survey instruments and contributed to conceptualization of the study. DB conducted the interviews. ED and NL supervised the data collection. DB and LL entered the data. ED conducted data analysis. ED, MP and NB coordinated drafting the article. All authors reviewed the literature, contributed to the design of the analysis and interpretation of data, drafted sections of the article, reviewed the article critically, approved the final version, and are responsible for the reported research.

DATA SHARING STATEMENT

Data are available upon reasonable request

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Figure 1. Proportion of Canadian clinics that follow the Canadian interdisciplinary team composition guidelines by age group. CanDiD study, 2021 (n=41).

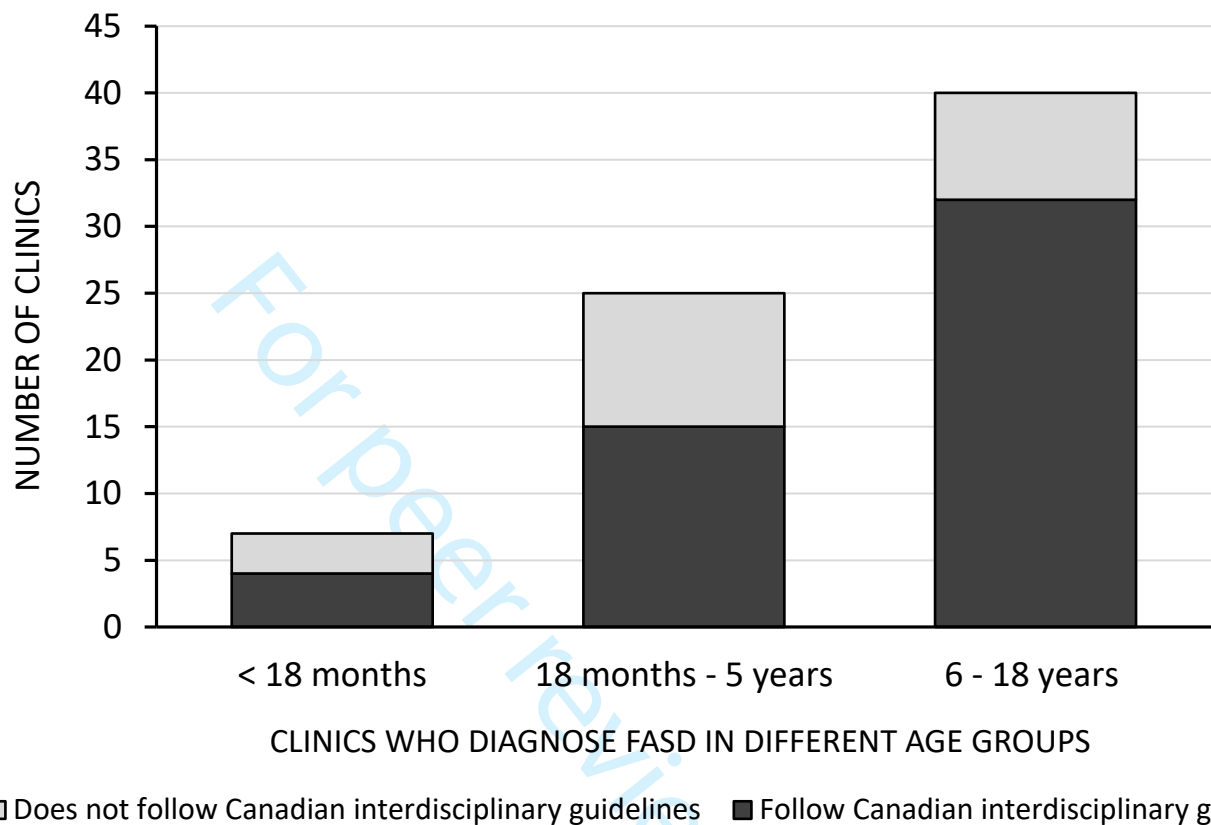


Figure legend: Canadian FASD diagnostic guidelines recommend that the diagnostic team for infants (<18 months) be composed of a pediatrician/physician and a child development specialist able to conduct physical and functional assessments (i.e., speech-language pathologist, physiotherapist, occupational therapist or clinical psychologist). Recommendations for the diagnostic core team for pre-schoolers (18mo–5yr) and school-aged children (6-18 yrs) include a physician, psychologist, a speech-language pathologist, and an occupational therapist (Cook et al., 2016).

Supplementary Table 1. Description of CanDiD study variables.

Study variable	Response choices	Recoded for analysis
General information on the clinic		
What best describes your role at the clinic?	Pediatrician; Family physician; Occupational therapist; Speech-language pathologist; Psychologist; Clinic manager; Community coordinator; Other (specify)	-
Where is your clinic located (province)	As is	Atlantic provinces (NB, NS, NFLD, PEI); Central Canada (ON, QC); Western and Northern Canada (MB, SK, AB, BC, YK)
What year did your clinic become operational?	As is	-
What are your sources of funding (check all that apply)?	Federal funding; Provincial funding; Research grant; Non-governmental organization (NGO); Other	-
What services does your clinic offer (check all that apply)?	Prevention; Screening; Diagnosis; Support to families; Training for health professionals; Other (specify)	Prevention; Screening; Diagnosis; Support to families; Training for health professionals
In a typical year (<i>please think about a normal year prior to COVID-19 pandemic</i>), how many new FASD referrals does your clinic receive?	As is	-
In a typical year (<i>please think about a normal year prior to the COVID-19 pandemic</i>), how many FASD assessments are completed?	As is	-
Does your clinic require a confirmation of prenatal alcohol exposure OR the presence of the 3 sentinel facial features for a FASD diagnostic assessment?	Yes; No	-
Interdisciplinary team composition		
Does your clinic diagnose FASD in infants <18 months?	Yes; No	-

Is the FASD diagnosis of infants <18 months done by an interdisciplinary team?	Yes; No	-
Which healthcare professionals are included in the interdisciplinary team for the diagnosis of infants <18 months (check all that apply)?	Pediatrician; Family physician; Occupational Therapist; Speech-language pathologist; Psychologist; Other (specify)	-
Does your clinic diagnose FASD in children 18 months – 5 years?	Yes; No	-
Is the FASD diagnosis of children 18 months – 5 years done by an interdisciplinary team?	Yes; No	-
Which healthcare professionals are included in the interdisciplinary team for the diagnosis of children 18 months – 5 years (check all that apply)?	Pediatrician; Family physician; Occupational Therapist; Speech-language pathologist; Psychologist; Other (specify)	-
Does your clinic diagnose FASD in children/adolescents aged 6 – 18 years?	Yes; No	-
Is the FASD diagnosis of children/adolescents aged 6 – 18 years done by an interdisciplinary team?	Yes; No	-
Which healthcare professionals are included in the interdisciplinary team for the diagnosis of children/adolescents aged 6 – 18 years (check all that apply)?	Pediatrician; Family physician; Occupational Therapist; Speech-language pathologist; Psychologist; Other (specify)	-
Current diagnosis reporting practices		
Who is responsible for announcing the FASD diagnosis to parent(s)/legal guardian(s)?	Pediatrician; Family physician; Occupational Therapist; Speech-language pathologist; Psychologist; Other (specify)	-
When announcing the FASD diagnosis to parent(s)/legal guardian(s), is the diagnosis reported to the child/adolescent at the same time?	No; Yes, always; Yes, sometimes	-
Who is responsible for announcing the FASD diagnosis to the child/adolescent assessed?	Pediatrician / Family physician at the clinic; Another member of the interdisciplinary team at the clinic; Parent(s)/legal guardian(s); Family doctor (outside of the clinic); Other (specify)	-

What factor(s) determine(s) whether the child/adolescent is present during the FASD disclosure (check all that apply)?	Parents’/legal guardians’ request; Child/adolescent developmental age; Child/adolescent chronological age; Clinical judgement of possible impact of the FASD diagnosis on the child/adolescent; The clinic’s policy; Other (specify)	-
Does your clinic use any of the following explanatory tools when announcing the FASD diagnosis to parent(s)/legal guardian(s) (check all that apply)...? Please think of tools you use to facilitate the disclosure, not tools used as a mode of delivery, such as Zoom or Skype.	(i) Visual tools (pictures, graphs); (ii) Electronic tools (ipads, computers); (iii) Communication tools (analogies); (iv) Other (specify) (Yes, No) for each item	-
Does your clinic use any of the following explanatory tools when announcing the FASD diagnosis to children/adolescents (check all that apply)...? Please think of tools you use to facilitate the announce disclosure ment, not tools used as a mode of delivery, such as Zoom or Skype.	(i) Visual tools (pictures, graphs); (ii) Electronic tools (ipads, computers); (iii) Communication tools (analogies); (iv) Other (specify) (Yes, No, Clinic does not disclose the FASD diagnosis to the child/adolescent) for each item	-
Immediate and post-diagnosis support and counseling		
Does your clinic provide any support/counseling to families during the FASD disclosure?	(i) Support/counseling to the parents/caregivers; (ii) Support/counseling to the child/adolescent; (iii) Support/counseling to sibling/other family member (Yes, No) for each item	-
Does your clinic provide any support/counseling to families in the 3-month period following the disclosure?	(i) Support/counseling to the parents/caregivers; (ii) Support/counseling to the child/adolescent; (iii) Support/counseling to sibling/other family member	-

	(Yes, No) for each item	
During the FASD disclosure or in the following 3-month period, does your clinic provide information on any of the following to families in need (check all that apply)?	FASD (pamphlets, brochures); FASD support groups for families; Financial aid programs; Respite care programs; Support worker programs; Housing programs; Mental health programs (other than those offered at the clinic); Jordan's principle; Other (specify)	-

STROBE Statement—Checklist of items that should be included in reports of *cross-sectional studies*

	Item No	Recommendation	Page No
Title and abstract	1	(a) Indicate the study's design with a commonly used term in the title or the abstract	1,3
		(b) Provide in the abstract an informative and balanced summary of what was done and what was found	3
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	5,6
Objectives	3	State specific objectives, including any prespecified hypotheses	6
Methods			
Study design	4	Present key elements of study design early in the paper	7
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	7
Participants	6	(a) Give the eligibility criteria, and the sources and methods of selection of participants	7
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	7
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	8
Bias	9	Describe any efforts to address potential sources of bias	17
Study size	10	Explain how the study size was arrived at	8
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	8
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding	8
		(b) Describe any methods used to examine subgroups and interactions	8
		(c) Explain how missing data were addressed	n/a
		(d) If applicable, describe analytical methods taking account of sampling strategy	n/a
		(e) Describe any sensitivity analyses	n/a
Results			
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	8
		(b) Give reasons for non-participation at each stage	8
		(c) Consider use of a flow diagram	n/a
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	9,10
		(b) Indicate number of participants with missing data for each variable of interest	n/a
Outcome data	15*	Report numbers of outcome events or summary measures	12-13

Main results	16	(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	10-13
		(b) Report category boundaries when continuous variables were categorized	10-13
		(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	n/a
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	n/a
Discussion			
Key results	18	Summarise key results with reference to study objectives	14-15
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	17
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	14-16
Generalisability	21	Discuss the generalisability (external validity) of the study results	15-16
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	17

*Give information separately for exposed and unexposed groups.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at <http://www.plosmedicine.org/>, Annals of Internal Medicine at <http://www.annals.org/>, and Epidemiology at <http://www.epidem.com/>). Information on the STROBE Initiative is available at www.strobe-statement.org.

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Title: Canadian clinical capacity for Fetal Alcohol Spectrum Disorder assessment, diagnosis, disclosure and support to children and adolescents: a cross-sectional study

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3 **ABBREVIATIONS**

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- 5 CanDiD - Canadian Interdisciplinary Clinical Capacity to Diagnose FASD study
- 6
- 7
- 8 CanFASD – Canada FASD Research Network
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- 10 FASD – Fetal Alcohol Spectrum Disorder
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- 12 FAS - Fetal Alcohol Syndrome
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- 14 NGO - non-governmental organization
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- 17 PAE – prenatal alcohol exposure
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- 19 SD – standard deviation
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ABSTRACT

Objective: Canadian Fetal Alcohol Spectrum Disorder (FASD) guidelines encourage an age-specific interdisciplinary diagnostic approach. However, there is currently no standard-of-care regarding FASD diagnosis disclosure and few studies document Canadian FASD clinical capacity. Our objectives were to describe clinical capacity (defined as skills and resources) for FASD assessment, diagnosis, disclosure, and support in Canada.

Design, setting and participants: Data were drawn from the CanDiD study, a cross-sectional investigation of Canadian FASD clinical capacity. Forty-one clinics participated in the study. Data were collected in 2021 on the number and types of health professionals included in the assessment and diagnostic teams, the presence (or absence) of a minor patient when the FASD diagnosis is disclosed to parents/guardians, who is responsible for the diagnosis disclosure, the use of explanatory tools, and the types of support/counseling services available. The proportion of clinics that follow the Canadian interdisciplinary diagnostic guidelines by age group is described among participating clinics.

Results: Overall, 21, 13 and 7 specialized FASD clinics were in Western/Northern, Central and Atlantic Canada, respectively. The number of referrals per year surpassed the number of diagnostic assessments completed in all regions. Approximately 60% of clinics who diagnosed FASD in infants and preschool children (n=4/7 and 15/25, respectively) followed the interdisciplinary guidelines compared to 80% (n=32/40) in clinics who diagnosed school-aged children/adolescents. Diagnostic reporting practices were heterogeneous, but most used an explanatory tool with children/adolescents (67%), offered support/counseling (90-95%), and used case-by-case approach (80%) when deciding who would disclose the diagnosis to the child/adolescent and when.

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Conclusions: Limited diagnostic capacity and lack of FASD resources across Canada highlights a critical need for continued FASD support. This study identifies gaps in assessment, diagnosis, and reporting practices for FASD in children/adolescents across Canada.

Keywords: FASD; clinical capacity; diagnosis disclosure; children; interdisciplinary team

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Strengths and Limitations of this study:

- This is the first study that aims to describe Canadian clinical capacity for assessment, diagnosis, disclosure, and support of FASD in children and adolescents.
- This study used an extensive search to identify eligible clinics including membership in the Canadian FASD Research Network (CanFASD) and through an exhaustive Internet search using purposive sampling (i.e., snowball or network sampling).
- Quantitative data of Canadian specialty clinics who engage in FASD diagnostic assessment of children and adolescent <18 years were collected cross-sectionally via a telephone/videoconference survey.
- Surveys included questions pertaining to: i) general clinic information (e.g., location, source(s) of funding, services offered); ii) number of referrals and assessments done per year; iii) interdisciplinary team composition for FASD diagnosis by age group; iv) current diagnosis reporting practices for children/adolescents; and v) support and counseling following the diagnosis disclosure.
- Clinical capacity data could not be collected from non-participating clinics, therefore, results from this study may not be generalizable to all Canadian clinics.

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3 **INTRODUCTION**

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5 Fetal alcohol spectrum disorder (FASD), which results from prenatal alcohol exposure (PAE)¹, is

6 one of the most frequent neurodevelopmental disorders in North America.² A recent meta-analysis

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8 reported that 8% of children exposed to PAE are diagnosed with FASD and that North America

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10 has the second highest FASD prevalence after Europe.³ Children exposed to PAE are at high risk

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12 of developing FASD regardless of the frequency or amount of alcohol consumed.⁴ Globally,

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14 current FASD prevalence estimates in the general population is approximately 1% with most

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16 countries in North America, Europe and the Western Pacific regions ranging between 1-3%.³

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18 However, experts believe that FASD is widely underdiagnosed worldwide because it is often

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20 overshadowed by other diagnoses (due to the high level of comorbidity) and because of limited

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22 expertise and/or resources available.⁵⁻⁷

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31 Canadian FASD evidence-based diagnostic guidelines (first published by the Public Health

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33 Agency of Canada in 2005,⁷ and most recently updated by the Canadian FASD Research Network

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35 (CanFASD) in 2016^{1,8}) include recommendations on key components for FASD assessment

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37 including screening, referral and support; medical assessment; sentinel facial features;

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39 neurodevelopmental assessments; nomenclature and diagnostic criteria; and management and

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41 follow-up.⁸ In fact, Canada is the only country that has developed and adopted a uniform diagnostic

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43 capacity for FASD through the harmonization of two American FASD guidelines, namely, the

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45 1996 Institute of Medicine (for the diagnosis of Fetal Alcohol Syndrome (FAS), partial FAS,

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47 alcohol-related birth defects, and alcohol-related neurodevelopmental disorder), and the 4-Digit

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49 Code approach (which measures growth, facial features, central nervous system impairments and

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51 PAE).^{7,9-11} Importantly, the Canadian guidelines require an age-specific interdisciplinary

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diagnostic team approach.⁸ Specifically, they recommend that the diagnostic team for infants (<18 months) should include a pediatrician/physician and a child development specialist able to conduct physical and functional assessments (i.e., speech-language pathologist, physiotherapist, occupational therapist or clinical psychologist) and that the diagnostic team for pre-schoolers (18mo–5yr) and school-aged children (6-18 yrs) should include a physician, psychologist, speech-language pathologist and an occupational therapist.⁸ The Canadian guidelines have demonstrated significant promise for setting the bar for FASD recognition and service development nationally and internationally. When used in population studies in Europe, Africa and North America, the Canadian guidelines were shown to be a key source for health professionals.⁹ For example, the Scottish Intercollegiate Guidelines Network (SIGN) and Healthcare Improvement Scotland have adapted their FASD clinical guidelines from Canada^{12,13} and Australia recently reviewed their guidelines and adopted several concepts from the Canadian FASD guidelines.¹⁴

Because FASD guidelines require an age-specific interdisciplinary diagnostic team approach, FASD clinical capacity, defined herein as skills and resources to assess, diagnose, disclose and support FASD, remains challenging and complex in many jurisdictions and is further exacerbated due to the current healthcare workforce shortage in several jurisdictions.¹⁵ A previous study reviewed Canadian FASD clinical practices in seven provinces and reported that only 46% of Canadian clinics had a complete multidisciplinary team on-site, but 90% used a team approach for diagnosis and treatment plan.¹⁶ However, the authors did not specify which health professional participated in the diagnostic assessments by age group.¹⁶ There are no empirical data on the Canadian clinical capacity for interdisciplinary diagnostic assessment since the guidelines were updated in 2016.

Skillful diagnosis disclosure and psychological support following a medical diagnosis to children and adolescents is extremely important to limit psychological trauma,¹⁷ increase adherence to treatment plans,¹⁸ and generally improve quality of life. While Canadian FASD guidelines recommend that individuals with FASD and their caregivers have access to resources to improve diagnostic outcomes,⁸ there are currently no specific recommendations regarding the presence of a minor patient (≤ 18 years) when the FASD diagnosis is disclosed to parents/guardians and, to our knowledge, no known studies describing clinical practices for FASD diagnosis disclosure to children - who should make the diagnosis disclosure, under which circumstances (e.g., favorable environment) and how (e.g., use of explanatory tools).

These knowledge gaps compel a research agenda that aims to describe FASD clinical capacity to inform clinical and diagnostic protocols to children and adolescents.¹²⁻¹⁴ Specific objectives of this study were to describe: (i) diagnostic interdisciplinary team composition (< 18 months; ≥ 18 months- ≤ 5 years; $\geq 6-18$ years) and the number of clinics who follow the Canadian interdisciplinary diagnostic team guidelines by age group; (ii) FASD assessment and diagnosis reporting practices for children and adolescents including if the minor patient is present during the diagnosis disclosure, who discloses the diagnosis to the child/adolescent patient, and use of explanatory tools to facilitate the FASD diagnosis disclosure; and (iii) FASD support and counseling provided to patients and families during the diagnosis disclosure and in the 3-month period following the diagnosis. Because Canada is one of the key leaders in FASD research and policy, exposing gaps in Canadian clinical capacity may have great relevance internationally when developing clinical guidelines.

METHODS

Data were drawn from the Canadian Interdisciplinary Clinical Capacity to Diagnose FASD (CanDiD) study – a cross-sectional survey-based study developed by clinicians and researchers at Vitalité Health Network in 2021. CanDiD is the first investigation that aims to describe Canadian clinical capacity for assessment, diagnosis, disclosure, and support of FASD in children and adolescents. Quantitative data were collected via a telephone/videoconference survey (available in French and English) of Canadian specialty clinics who engage in FASD diagnostic assessment of children and adolescent <18 years. Clinics were identified through membership in the Canadian FASD Research Network (CanFASD), and through an exhaustive Internet search using purposive sampling (i.e., snowball or network sampling). Introductory recruitment emails were sent to all identified clinics in spring 2021, followed by a phone call one week later to schedule an interview. To increase response proportion, up to 3 follow-up emails (or phone calls) were sent. Semi-structured 20-minutes surveys were completed with a key respondent in each clinic identified by the clinic manager or lead pediatrician who is most knowledgeable with clinical capacity. A copy of the questionnaire was sent to respondents prior to survey completion to allow preparation and consultation with clinic staff. The study was approved by the ethics committee of Vitalité Health Network (ethics #101241; approved on 2021-04-12). Participants provided verbal informed consent.

Study variables

Questionnaire items were developed *de novo* or adapted from the literature.⁸ All items were translated in French by two Francophone health professionals and back translated (by Anglophone

health professionals) to ensure accurateness of the translation. Questionnaires were then extensively pilot-tested in both French and English with healthcare professionals who work closely with children and families affected by FASD for readability and ease of comprehension. Variables measured included questions pertaining to: i) general information on the clinic (e.g., location, source(s) of funding, services offered); ii) number of referrals and assessments done per year (used to calculate diagnostic capacity, defined as the proportion of assessment completed in a year amongst the number of referrals received); iii) interdisciplinary team composition for FASD diagnosis by age group; iv) current diagnosis reporting practices for children/adolescents and use of explanatory tools; and v) immediate and post-diagnosis support and counseling following the diagnosis disclosure. Supplementary Table 1 describes each variable investigated including the item used in the survey, response options, and re-coding of response options for analysis.

Data analysis

Of 78 clinics identified, 18 were excluded (i.e., did not diagnose FASD in children ≤ 18 years, or had permanently closed), 6 refused to participate, and 13 did not return our recruitment efforts. Forty-one clinics completed the study questionnaire (68% of eligible clinics (41/60)).

To protect the clinics confidentiality, select characteristics were compared in three provincial groupings: *Western and Northern Canada* included clinics located in British Columbia, Alberta, Saskatchewan, Manitoba and Yukon; *Central Canada* included clinics located in Ontario and Quebec; and *Atlantic Canada* included clinics located in New Brunswick, Nova Scotia, Prince Edward Island and Newfoundland. Because of the small sample size, FASD clinical capacity characteristics were reported by age group for all clinics. Means for continuous variables and

frequency distributions for categorical variables were compared. Analyses were performed using SPSS, Version 26.0 (IBM Corp. Released 2012. IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp.).

PATIENT AND PUBLIC INVOLVEMENT

Patients or the public were not involved in this study.

RESULTS

In this study, more than half of respondents were clinic administrators (56%, n=23), 15% (n=6) were physicians/pediatricians, and 29% (n=12) were other health professionals.

Socio-demographic and clinic characteristics

Half of the FASD clinics were in Western/Northern Canada (n=21), and 13 and 7 clinics were in Central and Atlantic Canada, respectively (compared to n=11, 7 and 1 of non-participating clinics (n=19), respectively). Western/Northern Canada had the highest proportion of clinics that were operational before 2006 (including the oldest clinic in the sample (i.e., operational since 1999)) and Atlantic Canada had the highest proportion of clinics that were operational in the last decade (Table 1). No clinic located in Central or Atlantic Canada was operational before 2005.

Table 1. Clinical characteristics of Canadian clinics offering FASD diagnosis to children ≤18. CanDiD study, 2021 (n=41).

	FASD Clinics in Canada			
	Western and Northern Canada	Central Canada	Atlantic Canada	Total (all clinics)
Number of clinics, n	21	13	7	41
Year clinic became operational, n				
≤2005	8	3	1	12
2006-2011	8	5	2	15
≥2012	4	4	3	11
Don't know	1	1	1	3
Number of referrals per year				
Total	1503	780	254	2537
mean (SD)	84 (111)	65 (74)	36 (39)	69 (90)
Number of assessments per year				
Total	1257	430	110	1797
mean (SD)	63 (64)	36 (33)	16 (13)	46 (52)
Source of funding for clinic services, % (n)				
Federal funding	5 (1)	15 (2)	43 (3)	15 (6)
Provincial funding	95 (20)	85 (11)	86 (6)	90 (37)
Research grant	14 (3)	0	14 (1)	10 (4)
Non-governmental organization (NGO)	33 (7)	15 (2)	0	22 (9)
Other (donations, private funding)	14 (3)	46 (6)	57 (4)	32 (13)
Services offered, % (n)				
Prevention	38 (8)	31 (4)	29 (2)	34 (14)
Screening	43 (9)	69 (9)	57 (4)	54 (22)
Diagnosis	100 (21)*	100 (13)*	100 (7)*	100 (41)*
Support to families	86 (18)	100 (13)	71 (5)	88 (36)
Specialized FASD training for health professionals	71 (15)	54 (7)	57 (4)	63 (26)

*Participating clinics were selected because they engaged in FASD diagnostic assessment of children/adolescent <18 years (i.e., inclusion criteria). SD=Standard deviation.

Most regions received provincial funding (85-95%). Proportion of in-kind donations and private funding were highest in Central (46%) and Atlantic Canada (57%). Atlantic clinics reported the highest proportion of federal funding (43%), while Central and Western/Northern clinics reported receiving non-governmental organization (NGO) funding (15% and 33%, respectively; 0 Atlantic clinics reported NGO funding) (Table 1).

Among participating clinics, approximately 2537 referrals are received every year and only 1797 assessments are completed (diagnostic capacity: 71%). Western/Northern clinics received the highest number of FASD referrals per year (mean(sd): 84(111); total of 1503/2537 (59%)) and their diagnostic capacity was highest (83%) (Table 1). Diagnostic capacity in Central and Atlantic clinics was approximately 50% (55% in Central and 43% in Atlantic Canada).

All participating clinics offered diagnosis services (i.e., inclusion criteria for the CanDiD study). Approximately one third of Canadian clinics were involved in prevention efforts (34%), 43-69% of regions were involved in FASD screening, almost two thirds of clinics offered FASD training for health professionals (63%), and most offered support to families (88%) (Table 1).

Assessment and diagnosis by age group

Requirement of the confirmation of PAE or the presence of 3 sentinel facial features for FASD diagnosis assessment was variable among regions: 67% (n=14), 77% (n=10) and 43% (n=3) of clinics located in Western/Northern, Central and Atlantic regions required this confirmation, respectively.

Only 17% (n=7/41) of clinics diagnosed FASD in infants <18 months; among these 7 clinics, 4 followed the Canadian interdisciplinary team guidelines (Figure 1). Similarly, 60% (n=15/25) of clinics who diagnosed FASD in preschool children aged 18 months-5 years followed the interdisciplinary team guidelines. Most clinics (n=40/41) diagnosed FASD in school-aged children (i.e., ≥6-18 years), of which 80% (n=32/40) followed the Canadian guidelines (Figure 1).

Diagnosis disclosure

Designated individuals in charge of the FASD diagnosis disclosure to children/adolescents varied considerably across clinics. In 41% of clinics (n=17/41), pediatricians/physicians were responsible for the disclosure. Other health professionals and clinic administrators were responsible for diagnosis disclosure in 15% (n=6/41) and 5% of clinics (n=2/41), respectively. One quarter of clinics used a multidisciplinary team for the disclosure (20% (n=8/41) used more than one professional including a physician and 5% (n=2/41) used more than one professional excluding a physician). Finally, in 15% of clinics (n=6/41) the person responsible for the disclosure varied and often included parents in the diagnosis delivery.

Only 5% of clinics (n=2/41) always disclose the diagnosis to children/adolescents at the same time as the parents/guardian disclosure, 15% (n=6/41) never included children when first disclosing the diagnosis to parents/guardian, and 80% (n=33/41) included the child sometimes, depending on certain factors. Most frequently cited determining factors included parents/guardian request, child developmental age, child chronological age, clinical judgement and clinic’s policy.

Most clinics reported using at least one type of explanatory tools when announcing the diagnosis with parents/guardian (85%) or children/adolescents (67%) (Table 2). In both parents and children, visual tools (pictures, graphs) or communication tools (analogies) were used most often (Table 2).

Table 2. Description of explanatory tools used when announcing the FASD diagnosis to parents/guardian and children/adolescents. CanDiD study, 2021 (n=41).

	Parent/ guardian	Child*
Use of tools when announcing the FASD diagnosis, % (n)		
Visual tools (pictures, graphs)	73 (30)	53 (20)
Electronic tools (ipads, computers)	17 (7)	13 (5)
Communication tools (analogies)	63 (26)	53 (20)
Use at least one tool	85 (35)	67 (27)

*Among 38 clinics who disclose the FASD diagnostic to child

Immediate and post-diagnosis support/counseling following diagnosis disclosure

Almost all clinics offered support and counseling to parents/guardian at the time of disclosure (95%) and in the 3-month period following the disclosure (90%). Seventy-one percent and 76% of clinics offered support and counseling to children when announcing the diagnosis and in the 3-month following the disclosure, respectively. While only one third of clinics offered support/counseling to other family members (i.e., siblings, other close family members) at the time of FASD diagnosis disclosure, 65% offered them support/counseling in the 3-month period following the disclosure.

All clinics provided information to families in need post-diagnosis. The information requested most often included information on financial aid programs, mental health programs outside the clinic, support group programs and information pertaining to Jordan's principal Implementation Act (i.e., ensures equality in health, social and educational services to all First Nations children).

DISCUSSION

Principal findings

Although the majority (53%) of specialized FASD clinics were located in Western Canada, this region deserved the largest population of youth <19 years (i.e., approximately 4.9 million compared to 2.7 million in Central Canada and 467K in Atlantic Canada).¹⁹ Consequently, 59% of total requests for FASD consultations were received in Western Canadian clinics. While it is important to note that their diagnostic capacity was higher than eastern and central clinics, clinics in all jurisdictions were not able to meet FASD referral demands highlighting a critical lack of resources.

Our results show that few clinics assessed and diagnosed FASD in infants and preschool-aged children. Although FASD can be diagnosed at various ages, it is commonly diagnosed when children enter the school-system (>5 years) and fail to attain behavioural and developmental milestones (e.g., motor skills, social skills, language development).^{20,21} Most clinics followed the Canadian interdisciplinary guidelines for infants, preschool and school age children and adolescents (57% (n=4/7), 60% (n=15/25) and 80% (n=32/40), respectively).

Diagnostic reporting practices were heterogeneous across Canada with the exception that most used some type of explanatory tool when announcing the diagnosis to both parents/caregivers and patients. While physicians were involved in diagnosis disclosure to minor patients in most clinics (60%), approximately one-fifth of clinics entrusted other health professionals with the disclosure, and remaining clinics used administrators, parents, or a variable approach depending on the clinical judgment of the healthcare team. Most clinics (80%) did not systematically include or exclude

children/adolescents when announcing the diagnosis to parents/caregivers and used a case-by-case approach when delivering the diagnosis.

Finally, our results indicate that most Canadian clinics supported families during and after the diagnosis disclosure. This is of utmost importance as receiving a life-altering diagnosis can be traumatizing for patients and families.¹⁸ Prompt and easy access to counseling following a medical diagnosis can impact adherence to treatment plans¹⁷ and, in turn, impact FASD-related outcomes.

Strengths and limitations

Limitations of this study include that self-report data are subject to misclassification. Although key respondents were appointed by clinic managers or lead pediatricians and represented the person “most knowledgeable about the clinic’s clinical capacities”, data were provided by a single person and may not adequately reflect clinical capacities of each clinic. To protect each clinic’s confidentiality, it was impossible to describe inter-provincial differences. Finally, because we had no clinical capacity data for non-participating clinics, results may not be generalizable to all clinics in Canada.

Despite these limitations, CanDiD is the first study that aims to better understand national clinical capacity for FASD assessment, diagnosis, disclosure and support in children and adolescents providing important evidence of available FASD resources - or lack thereof – for policy makers, clinicians, and researchers. Only one previous research team aimed to determine FASD clinical capacity in Canada. However, their studies precedes the 2016 updated FASD guidelines and only report FASD clinical programs in select Canadian territories and provinces.^{16,22}

Clinical and policy implications, and future directions

Our work highlights the imminent need for a National FASD Strategy to ensure that all individuals with FASD and their families have access to services they need. Results from this study demonstrate that although progress has been made in FASD advocacy, monitoring and training, diagnostic capacity remains an important public health issue.¹⁶ Our study mirrors findings from Claren et al., (2011) highlighting a critical need for increased diagnostic capacity.¹⁶ If FASD prevalence rates are underestimated in Canada as well as in other jurisdictions worldwide, and current resources do not meet demand, policy makers and clinicians need to mobilize efforts to find solutions to offer healthcare services to all individuals with FASD. Equality and equity in health services access is essential to improve health condition, support youth and families and improve quality of life of individuals with FASD. Future studies should investigate FASD prevention and potential FASD health inequality in Canada.

Early diagnosis, diagnostic treatment planning, skillful treatment delivery and support of patients and families experiencing FASD is of utmost importance to improve child development²³ and mitigate negative factors associated with its neurodevelopmental impairments including academic failure, substance abuse, poor mental health, problems with law enforcement and maintaining employment.²³⁻²⁶ In a recent viewpoint article, Nunn²⁷ stated that medical news delivery should be age-appropriate, and in most cases, interaction and delivery is more important than the information itself. Because a child’s chronological age does not necessarily correspond to their developmental age, when and how a diagnosis should be disclosed is also unclear.¹⁸ Other factors such as cognitive impairments or maturity levels may represent additional barriers when delivering a neurodevelopmental diagnosis.¹⁸ While existing medical news delivery protocols such as

BREAKS²⁸ or SPIKES²⁹ can offer some guidance, they are not specific to neurodevelopment disorders and do not offer insight for medical news delivery to minors. Although recommendations on reporting practices of neurodevelopmental disorders cannot take a one-size-fits all approach, high heterogeneity in Canadian reporting practices highlight the need for more research to better understand which tools should be used and which health professionals should make the disclosure to improve clinical and diagnostic delivery protocols for children/adolescents.

It is noteworthy that following the survey completion, most clinics who did not have a complete interdisciplinary diagnostic team acknowledged that they did not meet the Canadian guidelines because of limited human and/or financial resources – they were simply doing the “best they could”. Specialized multidisciplinary FASD clinics have the potential to: (i) develop and implement prevention programs to reduce alcohol consumption in pregnant women and in women of child-bearing age; (ii) offer timely resources to individuals and families afflicted by FASD; (iii) provide continued training to health care professionals to ensure proper screening and diagnoses; and (iv) improve health care utilization (and indirectly improve justice and education services) by providing early accurate diagnoses. Consequently, continued national support for FASD clinical capacity including funding for development, training and maintenance of interdisciplinary teams is imperative to improve health services for children living with this life-long difficult condition.⁷

Because FASD is a lifelong multifaceted medical diagnosis associated with a heavy individual, clinical, public health and economic burden, experts highlight the urgent need for increased capacity to recognize, diagnose and monitor the full range of FASD.^{5,6} Lange et al., (2017) recommended the development of a universal screening protocol as well as other strategies such

as education and support to decrease FASD.³ We hope that findings from this study may increase understanding that FASD is a critical public health problem and that continued monitoring, healthcare services increase, training for health professionals, as well as the development of clinical tools and guidelines are prioritized nationally and internationally.

CONCLUSION

The CanDiD study provides evidence-base data that identifies outstanding knowledge gaps in FASD clinical capacity. Specifically, results from this study indicate that a lack of resources appears to be a serious impediment to healthcare for youth with FASD highlighting a need for continued FASD support.

ACKNOWLEDGEMENTS

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COMPETING INTERESTS STATEMENT

The authors declare no conflict.

ETHICS APPROVAL

The study was approved by the ethics committee of Vitalité Health Network (ethics #101241; approved on 2021-04-12). Participants provided verbal informed consent.

AUTHORS' CONTRIBUTIONS

ED, DB and NL developed the survey instruments and contributed to conceptualization of the study. DB conducted the interviews. ED and NL supervised the data collection. DB and LL entered the data. ED conducted data analysis. ED, MP and NB coordinated drafting the article. All authors reviewed the literature, contributed to the design of the analysis and interpretation of data, drafted sections of the article, reviewed the article critically, approved the final version, and are responsible for the reported research.

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DATA SHARING STATEMENT

Data are available upon reasonable request

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FIGURE LEGEND

Figure 1: Canadian FASD diagnostic guidelines recommend that the diagnostic team for infants (<18 months) be composed of a pediatrician/physician and a child development specialist able to conduct physical and functional assessments (i.e., speech-language pathologist, physiotherapist, occupational therapist, or clinical psychologist). Recommendations for the diagnostic core team for pre-schoolers (18mo–5yr) and school-aged children (6-18 yrs) include a physician, psychologist, a speech-language pathologist, and an occupational therapist.

Figure 1. Proportion of Canadian clinics that follow the Canadian interdisciplinary team composition guidelines by age group. CanDiD study, 2021 (n=41).

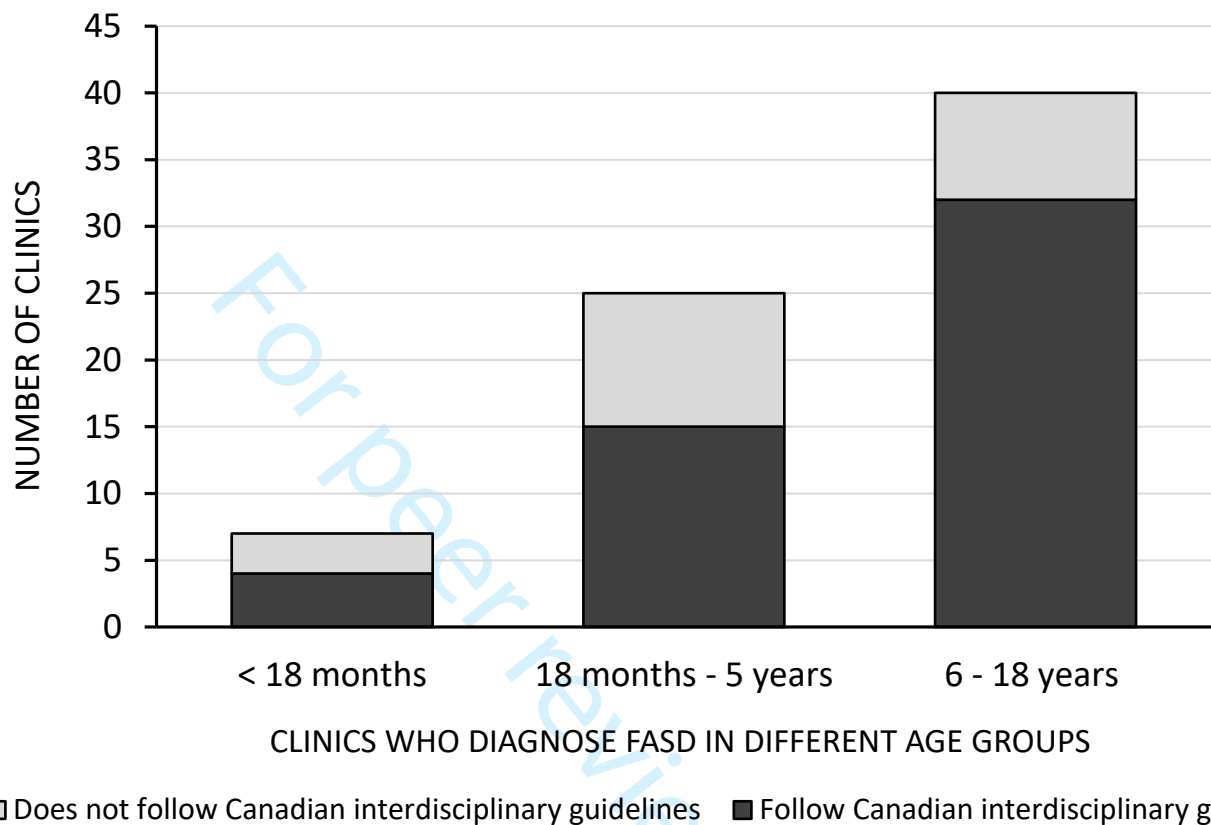


Figure legend: Canadian FASD diagnostic guidelines recommend that the diagnostic team for infants (<18 months) be composed of a pediatrician/physician and a child development specialist able to conduct physical and functional assessments (i.e., speech-language pathologist, physiotherapist, occupational therapist or clinical psychologist). Recommendations for the diagnostic core team for pre-schoolers (18mo–5yr) and school-aged children (6-18 yrs) include a physician, psychologist, a speech-language pathologist, and an occupational therapist (Cook et al., 2016).

Supplementary Table 1. Description of CanDiD study variables.

Study variable	Response choices	Recoded for analysis
General information on the clinic		
What best describes your role at the clinic?	Pediatrician; Family physician; Occupational therapist; Speech-language pathologist; Psychologist; Clinic manager; Community coordinator; Other (specify)	-
Where is your clinic located (province)	As is	Atlantic provinces (NB, NS, NFLD, PEI); Central Canada (ON, QC); Western and Northern Canada (MB, SK, AB, BC, YK)
What year did your clinic become operational?	As is	-
What are your sources of funding (check all that apply)?	Federal funding; Provincial funding; Research grant; Non-governmental organization (NGO); Other	-
What services does your clinic offer (check all that apply)?	Prevention; Screening; Diagnosis; Support to families; Training for health professionals; Other (specify)	Prevention; Screening; Diagnosis; Support to families; Training for health professionals
In a typical year (<i>please think about a normal year prior to COVID-19 pandemic</i>), how many new FASD referrals does your clinic receive?	As is	-
In a typical year (<i>please think about a normal year prior to the COVID-19 pandemic</i>), how many FASD assessments are completed?	As is	-
Does your clinic require a confirmation of prenatal alcohol exposure OR the presence of the 3 sentinel facial features for a FASD diagnostic assessment?	Yes; No	-
Interdisciplinary team composition		
Does your clinic diagnose FASD in infants <18 months?	Yes; No	-

Is the FASD diagnosis of infants <18 months done by an interdisciplinary team?	Yes; No	-
Which healthcare professionals are included in the interdisciplinary team for the diagnosis of infants <18 months (check all that apply)?	Pediatrician; Family physician; Occupational Therapist; Speech-language pathologist; Psychologist; Other (specify)	-
Does your clinic diagnose FASD in children 18 months – 5 years?	Yes; No	-
Is the FASD diagnosis of children 18 months – 5 years done by an interdisciplinary team?	Yes; No	-
Which healthcare professionals are included in the interdisciplinary team for the diagnosis of children 18 months – 5 years (check all that apply)?	Pediatrician; Family physician; Occupational Therapist; Speech-language pathologist; Psychologist; Other (specify)	-
Does your clinic diagnose FASD in children/adolescents aged 6 – 18 years?	Yes; No	-
Is the FASD diagnosis of children/adolescents aged 6 – 18 years done by an interdisciplinary team?	Yes; No	-
Which healthcare professionals are included in the interdisciplinary team for the diagnosis of children/adolescents aged 6 – 18 years (check all that apply)?	Pediatrician; Family physician; Occupational Therapist; Speech-language pathologist; Psychologist; Other (specify)	-
Current diagnosis reporting practices		
Who is responsible for announcing the FASD diagnosis to parent(s)/legal guardian(s)?	Pediatrician; Family physician; Occupational Therapist; Speech-language pathologist; Psychologist; Other (specify)	-
When announcing the FASD diagnosis to parent(s)/legal guardian(s), is the diagnosis reported to the child/adolescent at the same time?	No; Yes, always; Yes, sometimes	-
Who is responsible for announcing the FASD diagnosis to the child/adolescent assessed?	Pediatrician / Family physician at the clinic; Another member of the interdisciplinary team at the clinic; Parent(s)/legal guardian(s); Family doctor (outside of the clinic); Other (specify)	-

What factor(s) determine(s) whether the child/adolescent is present during the FASD disclosure (check all that apply)?	Parents’/legal guardians’ request; Child/adolescent developmental age; Child/adolescent chronological age; Clinical judgement of possible impact of the FASD diagnosis on the child/adolescent; The clinic’s policy; Other (specify)	-
Does your clinic use any of the following explanatory tools when announcing the FASD diagnosis to parent(s)/legal guardian(s) (check all that apply)...? Please think of tools you use to facilitate the disclosure, not tools used as a mode of delivery, such as Zoom or Skype.	(i) Visual tools (pictures, graphs); (ii) Electronic tools (ipads, computers); (iii) Communication tools (analogies); (iv) Other (specify) (Yes, No) for each item	-
Does your clinic use any of the following explanatory tools when announcing the FASD diagnosis to children/adolescents (check all that apply)...? Please think of tools you use to facilitate the announce disclosure ment, not tools used as a mode of delivery, such as Zoom or Skype.	(i) Visual tools (pictures, graphs); (ii) Electronic tools (ipads, computers); (iii) Communication tools (analogies); (iv) Other (specify) (Yes, No, Clinic does not disclose the FASD diagnosis to the child/adolescent) for each item	-
Immediate and post-diagnosis support and counseling		
Does your clinic provide any support/counseling to families during the FASD disclosure?	(i) Support/counseling to the parents/caregivers; (ii) Support/counseling to the child/adolescent; (iii) Support/counseling to sibling/other family member (Yes, No) for each item	-
Does your clinic provide any support/counseling to families in the 3-month period following the disclosure?	(i) Support/counseling to the parents/caregivers; (ii) Support/counseling to the child/adolescent; (iii) Support/counseling to sibling/other family member	-

	(Yes, No) for each item	
During the FASD disclosure or in the following 3-month period, does your clinic provide information on any of the following to families in need (check all that apply)?	FASD (pamphlets, brochures); FASD support groups for families; Financial aid programs; Respite care programs; Support worker programs; Housing programs; Mental health programs (other than those offered at the clinic); Jordan's principle; Other (specify)	-

STROBE Statement—Checklist of items that should be included in reports of *cross-sectional studies*

	Item No	Recommendation	Page No
Title and abstract	1	(a) Indicate the study's design with a commonly used term in the title or the abstract	1,3
		(b) Provide in the abstract an informative and balanced summary of what was done and what was found	3
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	5,6
Objectives	3	State specific objectives, including any prespecified hypotheses	6
Methods			
Study design	4	Present key elements of study design early in the paper	7
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	7
Participants	6	(a) Give the eligibility criteria, and the sources and methods of selection of participants	7
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	7
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	8
Bias	9	Describe any efforts to address potential sources of bias	17
Study size	10	Explain how the study size was arrived at	8
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	8
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding	8
		(b) Describe any methods used to examine subgroups and interactions	8
		(c) Explain how missing data were addressed	n/a
		(d) If applicable, describe analytical methods taking account of sampling strategy	n/a
		(e) Describe any sensitivity analyses	n/a
Results			
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	8
		(b) Give reasons for non-participation at each stage	8
		(c) Consider use of a flow diagram	n/a
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	9,10
		(b) Indicate number of participants with missing data for each variable of interest	n/a
Outcome data	15*	Report numbers of outcome events or summary measures	12-13

Main results	16	(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	10-13
		(b) Report category boundaries when continuous variables were categorized	10-13
		(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	n/a
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	n/a
Discussion			
Key results	18	Summarise key results with reference to study objectives	14-15
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	17
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	14-16
Generalisability	21	Discuss the generalisability (external validity) of the study results	15-16
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	17

*Give information separately for exposed and unexposed groups.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at <http://www.plosmedicine.org/>, Annals of Internal Medicine at <http://www.annals.org/>, and Epidemiology at <http://www.epidem.com/>). Information on the STROBE Initiative is available at www.strobe-statement.org.