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Developmental, sensory and behavioral outcomes among infants and toddlers with prenatal alcohol exposure

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ABSTRACT

Background: Prenatal alcohol exposure (PAE) can disrupt children's neurodevelopment and exert lasting influences on overall child well-being and family functioning. A comprehensive exploration of developmental outcomes in infants/toddlers with PAE seen for a diagnosis on the fetal alcohol spectrum can inform early identification and intervention.

Aims: To describe the prevalence and patterns of neurodevelopment, sensory processing, and emotional and behavioral functioning in a clinical sample of infants/toddlers with PAE.

Methods: In this retrospective analysis, clinical data from 125 infants/toddlers with PAE, aged 2–42 months, assessed at the University of Washington Fetal Alcohol Syndrome Diagnostic and Prevention Network clinic were analyzed.

Results: Seventy-four to 87% of infants/toddlers demonstrated delayed development in one or more domains of the Bayley Scales of Infant and Toddler Development ($n = 125$). Adverse developmental outcomes were significantly correlated with PAE and/or postnatal risk factors. All 93 infants/toddlers with a complete Infant/Toddler Sensory Profile obtained definite difference scores in at least one quadrant/section. Over half of infant/toddlers with a completed Child Behavior Checklist/1½–5 had total problem scores in the borderline or clinical range.

Conclusions: Findings suggest that several domains of child functioning may be vulnerable to the teratogenic impact of PAE, and that these delays are evident in the first years of life. Early screening, ongoing monitoring and comprehensive assessment is needed to facilitate earlier identification and guide clinical intervention.

What this paper adds?

Prenatal alcohol exposure (PAE) appears to be under-recognized by early childhood practitioners. Challenges related to effective

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screening processes, as well as difficulties detecting early delays or problems in the absence of physical features, may be inhibiting the early identification and intervention of infants/toddlers with PAE. Intervening early can buffer against the developmental vulnerability and postnatal adversity associated with PAE and optimize child and family outcomes in the long term. A comprehensive exploration of developmental, sensory processing and behavioral outcomes in young children seen in a Fetal Alcohol Spectrum Disorder (FASD) diagnostic clinic for individuals with PAE can inform early identification and intervention practices.

Clinically significant developmental delays were highly prevalent in this sample of infants/toddlers with PAE. These adverse developmental outcomes were significantly correlated with PAE and/or postnatal risk factors. Atypical sensory processing behaviors in at least one area were reported for every infant/toddler in the clinical sample. Over half of infants/toddlers presented with emotional and behavior problems outside of the normal range. Present findings, considered with similar studies reported in the literature, suggest that most domains of child functioning are vulnerable to the teratogenic impact of PAE and that these delays are evident in the first years of life. Findings reinforce the value of early screening, ongoing monitoring, and comprehensive assessment to facilitate earlier identification and to provide opportunities for infants/toddlers with PAE and their caregivers to benefit more fully from early supports and intervention.

1. Introduction

A wide range of adverse neurodevelopmental, sensory, and behavioral outcomes have been documented among children with prenatal alcohol exposure (PAE) (Astley, 2010; Astley et al., 2009; Carr et al., 2010; Subramoney et al., 2018). Fetal alcohol spectrum disorders (FASD), an umbrella term representing the full range of physical, cognitive, and behavioral impairments caused by PAE, are estimated to occur in at least 1% of children and youth in the general population (Popova et al., 2019; Roozen et al., 2016). Children with PAE and FASD are a clinically heterogeneous group, who may experience brain-based challenges across multiple domains including cognition, language, executive function, motor, self-regulation, and adaptive behavior functioning (Astley, 2010; Mattson et al., 2019). Although earlier diagnosis provides opportunities for children to benefit more fully from intervention and is predictive of more positive life outcomes in this population (Streissguth et al., 2004), PAE appears to be under-recognized by early childhood practitioners. In fact, many referrals for FASD diagnosis are not initiated until a child reaches school age (Olson et al., 2007), well beyond the time for early intervention. Challenges related to effective screening processes for maternal alcohol use history, as well as difficulties detecting early signs of delays or problems in the absence of physical features, may be inhibiting the early identification and intervention of infants/toddlers with PAE (Clarren & Astley, 1998; Olson et al., 2007; Watson et al., 2011).

Understanding the early neurodevelopmental effects among infants/toddlers with PAE is necessary to facilitate early diagnosis and intervention, a top priority in the field of FASD (SAMSHA, 2014). Decades of research has documented the developmental outcomes of infants/toddlers with PAE, frequently relying on global, standardized measures such as the Bayley Scales of Infant Development (Bayley, 1993; 2006; for a review of literature see (Garrison et al., 2019; Subramoney et al., 2018). Many of these earlier studies used the Bayley-II (Bayley, 1993), for example, which provides a general indication of functioning when it combines cognitive, expressive, and receptive language outcomes into one index (i.e., Mental Developmental Index) and gross and fine motor development into a second index (i.e., Psychomotor Developmental Index). Given that young children with PAE show considerable individual variability in development (Astley, 2010; Astley et al., 2009), more useful information may be generated from a developmental profile that examines outcomes across distinct scales. Other global measures of infant/toddler development, including newer versions of the Bayley (Bayley et al. (2019); Bayley (2006), are comprised of distinct domains and/or subdomains, which broadens the scope of the assessment. Furthermore, assessment of infant/toddlers' functioning in each of the five core developmental domains (i.e., cognitive, language, motor, social-emotional and adaptive functioning), for the purpose of identifying suspected delays and determining early intervention eligibility, is consistent with federal (Yell et al., 2006) and professional early childhood (Zeanah et al., 2016) standards.

Impairments in self-regulation have been frequently observed in young children (birth to 8 years) with PAE (Astley, 2010; Reid & Petrenko, 2018) and as such, have been recognized as a core symptom in the FASD 4-Digit Code diagnostic criteria for "Neuro-behavioral Disorder/Alcohol-Exposed and the proposed DSM-5 diagnostic criteria for "neurobehavioral disorder associated with PAE (ND-PAE)" (Kable et al., 2016). Self-regulation is defined as the ability to manage internal sensory, emotional, and behavioral states (Wells et al., 2012). These are the skills that allow children to regulate and respond to sensory input, pay attention, practice self-control, and manage strong emotions in an adaptive and age-typical manner. Sensory processing refers to an individual's capacity to detect, modulate, interpret, and respond to everyday sensations (Dunn, 2007; Miller et al., 2007). As such, information about early regulatory skills, which may be reflected in sensory processing behaviors and emotional and behavioral functioning, helps to broaden the picture of overall child development.

Documenting self-regulatory difficulties is important both for understanding the variable developmental performance of infants/toddlers with PAE and for guiding intervention. Caregiver rating scales and questionnaires have frequently been used to assess sensory processing differences and emotional/behavioral problems in the early childhood period (Astley, 2004, 2010; Coles et al., 2015; Fjeldsted & Xue, 2019; Molteno et al., 2014). Findings from a recent comparative analysis demonstrate the usefulness of caregiver-reported assessments for identifying behavioral deficits in infants with PAE, including those with light/moderate PAE (Bakhireva et al., 2018). However, for children older than three, researchers suggest that both caregiver-reported observations/ratings and performance-based measures of neurodevelopment be included to establish a comprehensive profile of FASD (Astley & Clarren, 2000; Lange et al., 2017). Taken together, findings underscore the importance of collecting relevant information from multiple sources to accurately identify developmental and behavioral problems across a continuum of infants/toddlers at risk for FASD (Astley, 2013).

This study aimed to explore the prevalence of neurodevelopmental delay, atypical sensory processing, and emotional and behavioral problems in a clinical sample of infants/toddlers with confirmed PAE to assist early childhood practitioners in identifying

early delays or problems that may arise from PAE. Given that alcohol is a neurobehavioral teratogen and children with PAE often experience high levels of co-occurring postnatal risks, outcomes were also explored in relation to PAE and selected demographic and environmental factors (Hemingway et al., 2020). The following research questions were asked:

1. What is the prevalence of neurodevelopmental delay in five core developmental domains (i.e., cognitive, language, motor, social-emotional and adaptive behavior)?
2. What is the prevalence of atypical sensory processing behaviors among infants/toddlers with PAE?
3. What is the prevalence of emotional and behavioral problems among infants/toddlers with PAE?
4. Are these adverse outcomes positively correlated with PAE and/or other postnatal risk factors? Does the prevalence of adverse outcomes differ by gender and age?

2. Methods

2.1. Research design

This study is a retrospective exploratory analysis of clinical data obtained from 125 infant/toddlers, ages 2–42 months, who received an interdisciplinary FASD diagnostic evaluation at the University of Washington Fetal Alcohol Syndrome Diagnostic and Prevention Network (FASDPN) clinic between 2009 and 2019. Children were referred to the clinic if they had a confirmed prenatal alcohol exposure history, at any level. Referral to the FASDPN does not require evidence of developmental delay. Data used for this study were collected with University of Washington Human Subjects Division oversight and approval and caregiver consent at the time of diagnosis.

The clinic has provided FASD diagnostic evaluations for individuals of all ages with PAE since 1993 and is one of the few clinics nationally that diagnose children under the age of three. The FASDPN database currently contains over 2000 fields of data (exposures and outcomes) on approximately 3000 patients (newborn to adult) with PAE. All patients in the FASDPN database received an FASD diagnostic evaluation by an interdisciplinary team (medical doctor, occupational therapist, psychologist, and speech language pathologist) using the FASD 4-Digit Diagnostic Code (Astley, 2004, 2013). The 4 digits of the code reflect the magnitude of expression of the 4 key diagnostic features of FASD in the following order: (1) growth deficiency, (2) FAS facial features, (3) central nervous system (CNS) structural, neurological and/or functional abnormality, and (4) prenatal alcohol exposure. The magnitude of expression of each feature is ranked independently on a 4-point Likert scale with 1 reflecting complete absence of the FASD feature and 4 reflecting strong

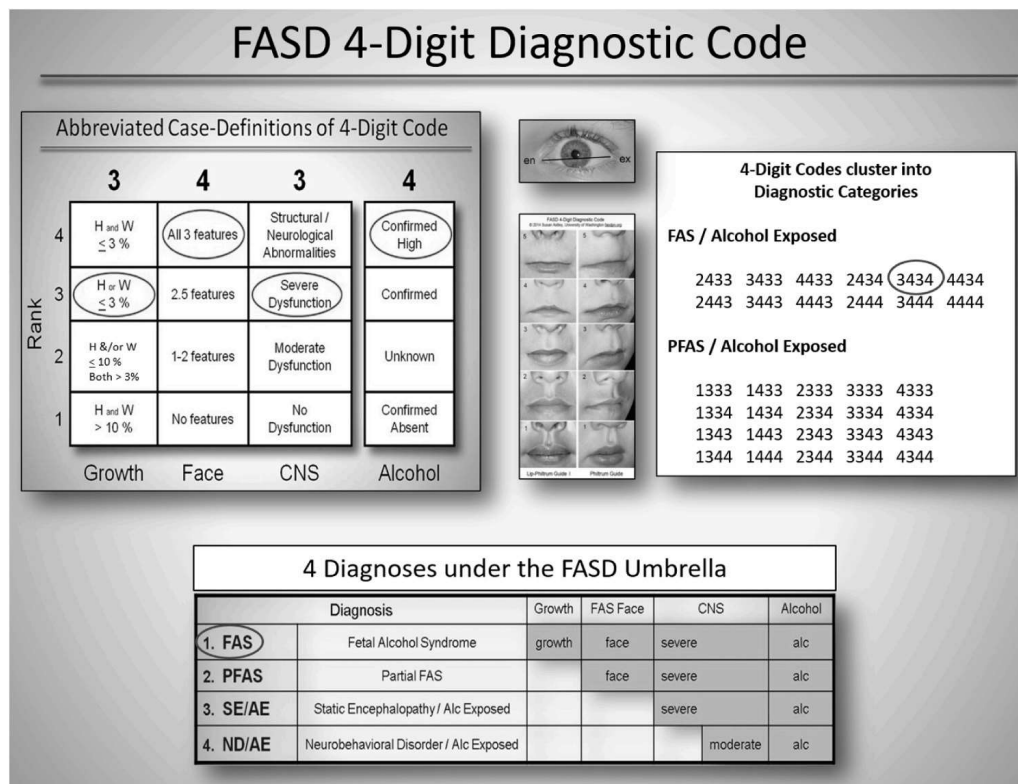


Fig. 1. Abbreviated case-definitions of the FASD 4-Digit Code (Astley, 2004, 2013). The 4-Digit Code 3434 is one of 12 Codes that fall under the diagnostic category FAS. The 4-Digit Code produces four diagnostic subgroups under the umbrella of FASD: FAS, PFAS, SE/AE and ND/AE. Abbreviations: Alc alcohol; CNS central nervous system; h height; w weight; % percentile.

“classic” presence of the FASD feature. Each Likert rank is specifically case defined (Fig. 1). There are 102 4-Digit codes that fall broadly under the umbrella of FASD. These codes cluster into four clinically meaningful FASD diagnostic subcategories (Astley, 2004): Fetal Alcohol Syndrome (FAS) (diagnostic categories A, B); Partial FAS (PFAS) (diagnostic category C); Static Encephalopathy/Alcohol Exposed (SE/AE) (diagnostic categories E, F) and Neurobehavioral Disorder/Alcohol Exposed (ND/AE) (diagnostic categories G, H). Not all individuals with PAE present with adverse outcomes that meet criteria for FASD. The FASD 4-Digit Code classifies these individuals as follows: “Sentinel Physical Findings/Alcohol-Exposed” (SPF/AE) (diagnostic category I; individuals with PAE who present with growth and/or facial abnormalities, but normal CNS outcomes) and “No Physical Findings or CNS Abnormalities/Alcohol-Exposed” (Normal/AE) (diagnostic category J). See Fig. 1. The FASD 4-Digit Code also ranks the magnitude other prenatal risks (e.g., other teratogens, illicit drugs, tobacco, poor prenatal care, family genetics) and postnatal risks (e.g., neglect, trauma, multiple home placements).

2.2. Participants

Data from children who met the following inclusion criteria were used in this study: 1) age 1 month to 3.5 years at the time of their FASD diagnostic evaluation; 2) received one of the following diagnoses FAS, PFAS, SE/AE and ND/AE, SPF/AE, Normal/AE reflecting the full continuum of outcomes observed among individuals with PAE using the FASD 4 Digit Diagnostic code; 3) had complete data on a minimum of two domains from the Bayley Scales of Infant and Toddler Development, third edition (Bayley-III; Bayley, 2006); and D) were of any race, ethnicity, or gender. The upper age range of the inclusion criteria was based on the age range of the Bayley Scales of Infant Development.

2.3. Measures and methods of data collection

The data used for this study were collected by an interdisciplinary diagnostic team using the FASD 4-Digit Code (Astley, 2004). While a core set of assessments are used during the diagnostic clinic visit, the final battery of assessments administered to each infant/toddler are based on clinician judgement. Therefore, data may be limited by clinically warranted situations and decisions where testing was not appropriate or not completed (e.g., child had a recent comprehensive assessment from another provider, or child was unable to attend to items, or child became irritable, upset or tired). Thus, complete data for outcomes on the Bayley-III from this clinical sample vary based on factors such as age, presenting developmental concern(s), and child’s ability to complete the assessment. Standardized parent questionnaires were completed by the primary caregiver prior to the scheduled diagnostic clinic date. Time, effort, or other demands placed on a caregiver may have resulted in some caregiver-report measures not being fully completed.

2.3.1. Child and family demographics

Information about child demographics, birth and medical history, growth, prenatal and postnatal experiences was collected during the intake process and caregiver interview at the time of diagnosis.

2.3.2. Assessment of infant/toddler development across five domains

Infants/toddlers were clinically assessed by the occupational therapist using the Bayley-III, a widely used, standardized developmental assessment for infants/toddlers, 1–42 months of age. The Bayley-III has five domains (i.e., Cognitive, Language, Motor, Social-Emotional, and Adaptive Behavior) that are presented as standard scores (mean = 100, $SD = 15$). Each of the five domains has 1–10 subdomains that are presented as scaled scores (mean = 10, $SD = 3$). Three of the domain scales are performance-based measures (i.e., Cognitive, Language, and Motor) and two scales are caregiver-report measures (i.e., Social-Emotional and Adaptive Behavior). For this study, domain and subdomain scores were collapsed into the following categories: domain scores: typical development (standard scores ≥ 86 , scores $\geq -0.9 SD$), at-risk development (78–85, scores between -1.0 and $-1.4 SD$), and delayed development (≤ 77 , scores $\leq -1.5 SD$); subdomain scores: typical development (scaled scores ≥ 8 , scores $\geq -0.9 SD$), at-risk development (6–7, scores between -1.0 and $-1.4 SD$), and delayed development (≤ 5 , scores $\leq -1.5 SD$). These categories were created to reflect current eligibility criteria for early intervention services in the state of WA. The Bayley-III is reported to have high internal consistency demonstrated by Cronbach’s alphas, ranging from .91 to .93 for domain scores and .86 to .91 for scaled scores (Albers & Grieve, 2007).

2.3.3. Assessment of sensory processing

The Infant Toddler Sensory Profile (ITSP; 7–36 months; (Dunn, 2002) is a 48-item caregiver-report questionnaire designed to measure sensory processing abilities in children ages 7–36 months. Caregivers rate the frequency of their child’s daily behavior on a scale from “almost always” (score of 1) to “almost never” (score of 5). Sensory processing was evaluated across five sections (i.e., auditory, visual, tactile, vestibular, and oral sensory processing). Infants/toddlers also received a score on their behavioral responses to sensation within four quadrants: low registration (i.e., fails to notice and respond to sensory input), sensation seeking (i.e., derives pleasure from and seeks out sensory experiences), sensory sensitivity (i.e., notice sensory input easily, tends to be reactive) and sensation avoiding (i.e., notices sensory input easily, tends to withdraw quickly). The ITSP categorizes the raw scores as typical performance (scores at or between plus 1.0 SD and minus 1.0 SD), probable differences (scores within -1.1 to $-1.9 SD$ s or $+1.0$ to $+1.9 SD$ s) and definite difference (scores at or below $-2 SD$ s or at or above $+2 SD$ s). The ITSP is reported to have excellent test-retest reliability ($\alpha = .86$) for domain/section scores and adequate ($\alpha = .74$) for quadrant scores (Eeles et al., 2013). Validity was established in several studies (Dunn, 2002; Dunn & Daniels, 2002; Eeles et al., 2013).

2.3.4. Assessment of emotional and behavioral problems

The Child Behavior Checklist for ages 1½–5 years (CBCL; (Achenbach & Rescorla, 2000) is a widely used instrument used to identify a range of behavioral and emotional problems in young children. Completed by the primary caregiver, the CBCL contains 100 items, rated as 0 = not true, 1 = sometimes true, and 2 = very true or often true, based on the preceding 2 months. The CBCL yields scores on three summary scales (Internalizing, Externalizing, and Total Problems), seven syndromes (Emotionally Reactive, Anxious/Depressed, Somatic Complaints, Withdrawn, Sleep Problems, Attention Problems, and Aggressive Behavior), and five DSM-oriented scales (Affective, Anxiety, Pervasive Developmental, Attention Deficit/ Hyperactive and Oppositional Defiant Problems). Summary scale *t*-scores are categorized as normal range (*t*-scores < 60), borderline clinical range (*t*-scores 60 to 63), and clinical range (*t*-scores ≥ 64). Syndrome and DSM-oriented *t*-scores are categorized as normal range (*t*-scores ≤ 64), borderline clinical range (*t*-scores 65 to 69), and clinical range (*t*-scores ≥ 70). This measure is reported to have high levels of internal consistency ($\alpha = 0.97$) and good test–retest reliability (mean *r* of .85 across all scales) (Rescorla, 2005).

2.3.5. Diagnostic outcome data

FASD 4-Digit Code Diagnoses. FAS; PFAS; SE/AE; ND/AE; Sentinel Physical Findings/AE; No Physical or CNS Abnormalities/AE. See full description above (Astley, 2004) and case definitions for the following diagnostic features (Fig. 1).

Growth Deficiency. ‘Growth Rank’: 1 = none; 2 = mild; 3 = moderate; 4 = severe. This variable yields the first digit in the FASD 4-Digit Diagnostic Code and documents the magnitude of prenatal and/or postnatal growth deficiency (Astley, 2004).

FAS Facial Phenotype. ‘Face Rank’: 1 = absent; 2 = mild; 3 = moderate; 4 = severe. This variable represents the second digit in the FASD 4-Digit Diagnostic Code and documents the magnitude of expression of FAS facial phenotype defined by short palpebral fissure lengths, a smooth philtrum, and a thin upper lip using the FAS Facial Photographic Analysis Software (Astley, 2004; Astley, 2016).

CNS Likelihood of Structural Abnormality. ‘CNS Rank’: 1 =unlikely; 2 =possible; 3 =probable;4 =definite. This variable yields the third digit in the FASD 4-Digit Diagnostic Code. These four ranks document the increasing likelihood of CNS structural abnormality. Alcohol is a teratogen that interferes with the structural development of the fetal brain. This, in turn, can lead to abnormal function. The greater the dysfunction, the higher the probability of CNS structural abnormality (Astley et al., 2009; Astley et al., 2009; Astley, 2013). The first three CNS Ranks document the severity of CNS dysfunction (Rank 1-no dysfunction; Rank 2- mild-to-moderate dysfunction; Rank 3-severe dysfunction). CNS Ranks 1–3 are based on brain function (executive function, memory, cognition, social/adaptive skills, academic achievement, language, motor, attention, and activity level) assessed by an interdisciplinary team using standardized psychometric tools. CNS Rank 4 documents the presence of direct evidence of CNS structural and/or neurological abnormalities (e.g., microcephaly, structural brain abnormalities, a seizure disorder of prenatal origin, or other hard neurological signs).

Prenatal Alcohol Exposure. ‘Alcohol Rank’: 1 = confirmed absence of exposure; 2 = unknown exposure; 3 = confirmed exposure; level unknown or low to moderate; 4 = confirmed exposure; level high). Alcohol exposure is the fourth digit in the FASD 4-Digit Diagnostic Code, which is ranked according to the quantity, timing, frequency, and certainty of exposure during pregnancy. A confirmed prenatal alcohol exposure at any level is required for a diagnostic evaluation in the FASDPN clinic. The clinic intake form (New Patient Information Form (Astley, 2004)) requests the following exposure information before and during pregnancy: average and maximum number of drinks per drinking occasion, average number of drinking days per week, trimesters of exposure and source of information. The primary sources of information are past and present medical and social service records. Roughly half of patients with confirmed prenatal alcohol exposure report quantity, frequency and duration of exposure. (see appendix 2 from Astley, 2004, 2013).

2.4. Other risk factors

Other Prenatal Risks. Rank: 1 = no risk; 2 = unknown risk; 3 = some risk; 4 = high risk (Astley, 2004). Other prenatal risk factors documented in the FASDPN clinical database include, but are not limited to poor prenatal care, pregnancy complications, prematurity, presence of other syndromes/genetic abnormalities, and prenatal exposure to other substances (e.g., medications, tobacco, illicit drugs, and/or other teratogens). The 4-Digit Code ranks the magnitude of these other prenatal risks in a single composite measure labeled “Other Prenatal Risks Rank.” Rank 4 is assigned when there is exposure to another teratogen (e.g., Dilantin) or when another syndrome or genetic condition is present (e.g., Down syndrome, Fragile X, etc.). Rank 3 is assigned to all other prenatal risks. The clinic intake form requests the patient to report these other prenatal risk factors when known. The ranking is determined by available records and caregiver or other report on intake forms and/or clinical interview (Astley, 2004).

Other Postnatal Risks. Rank: 1 = no risk; 2 = unknown risk; 3 = some risk; and 4 = high risk (Astley, 2004). Postnatal risk factors documented in the FASDPN database include, but are not limited to perinatal complications, number of home placements, physical and/or sexual abuse, neglect, and trauma. The 4-Digit Code ranks the magnitude of these other postnatal risks in a single composite measure labeled “Other Postnatal Risks Rank”. Rank 4 is used to note severe postnatal circumstances that have been shown to have a significant adverse effect on development in most instances. Examples include physical or sexual abuse, multiple home placements, and severe neglect. Rank 3 is used to note conditions akin to those in Rank 4, but the circumstances are less severe. The clinic intake form requests the patient to report these other postnatal risk factors when known.

3. Data analysis

Data analyses were completed using SPSS 27.0 (IBM Corp., New York). Descriptive statistics (e.g., means, standard deviations, proportions) were used to summarize the sociodemographic and clinical profiles of the study sample and to describe children’s scores

Table 1
Demographic and clinical profiles of infants/toddlers with Bayley-III, ITSP, and CBCL assessments.

Characteristic	Bayley-III		ITSP		CBCL	
	<i>n</i>	(valid %)	<i>n</i>	(valid %)	<i>n</i>	(valid %)
Total <i>n</i>	125		93		67	
Sex						
Female	64	(51.2)	50	(53.8)	39	(58.2)
Male	61	(48.8)	43	(46.2)	28	(41.8)
Age at FASD Diagnostic Evaluation (months)						
Birth to 11	22	(17.6)	9	(9.7)	0	0.0
12 - 23	45	(36.0)	42	(45.2)	18	(26.9)
24 - 35	44	(35.2)	39	(41.9)	36	(53.7)
36 - 42	14	(11.2)	3	(3.2)	13	(19.4)
Mean (SD)	22	(0.84)	22	(0.69)	28	(0.57)
Race/Ethnicity						
White	58	(46.4)	47	(50.5)	33	(49.3)
Black	5	(4.0)	3	(3.2)	2	(3.0)
Native American/Canadian	9	(7.2)	7	(7.5)	5	(7.5)
Hispanic	3	(2.4)	2	(2.2)	0	0.0
Asian	1	(0.8)	0	0.0	0	0.0
Mixed race	50	(40.0)	34	(36.6)	27	(40.3)
4-Digit Code FASD Diagnosis (Diagnostic category)						
FAS (AB)	5	(4.0)	0	0.0	0	0.0
PFAS (C)	5	(4.0)	4	(4.3)	1	(1.5)
SE/AE (EF)	13	(10.4)	8	(8.6)	8	(11.9)
ND/AE (GH)	75	(60.0)	62	(66.7)	41	(61.2)
Sentinel physical findings/AE (I)	5	(4.0)	4	(4.3)	5	(7.5)
No evidence of CNS abnormalities/AE (J)	22	(17.6)	15	(16.1)	12	(17.9)
Growth Rank						
Rank 1	71	(56.8)	54	(58.1)	41	(61.2)
Rank 2	21	(16.8)	15	(16.1)	10	(14.9)
Rank 3	22	(17.6)	20	(21.5)	12	(17.9)
Rank 4	11	(8.8)	4	(4.3)	4	(6.0)
Face Rank						
Rank 1	68	(54.4)	53	(57.0)	38	(56.7)
Rank 2	41	(32.8)	31	(33.3)	24	(35.8)
Rank 3	9	(7.2)	8	(8.6)	4	(6.0)
Rank 4	7	(5.6)	1	(1.1)	1	(1.5)
CNS Rank: Structural Damage Risk						
Rank 1, unlikely	27	(21.6)	19	(20.4)	17	(25.4)
Rank 2, possible	75	(60.0)	62	(66.7)	41	(61.2)
Rank 3, probable	2	(1.6)	8	(8.6)	0	0.0
Rank 4, definite	21	16.8)	1	(1.1)	9	(13.4)
CNS Functional Rank						
Rank 1, no dysfunction	29	(23.2)	20	(21.5)	18	(26.9)
Rank 2, moderate dysfunction	81	(72.8)	72	(77.4)	48	(71.6)
Rank 3, severe dysfunction	5	(4.0)	1	(1.1)	1	(1.5)
Prenatal Alcohol Exposure: Alcohol Rank						
3. Confirmed exposure: Amount moderate or unknown	55	(44.0)	43	(46.2)	31	(46.3)
4. Confirmed exposure: Amount high	70	(56.0)	50	(53.8)	36	(53.7)
Other Prenatal Risks: Rank						
1. No risk	1	(0.8)	0	0.0	0	0.0
2. Unknown risk	1	(0.8)	1	(1.1)	1	(1.5)
3. Some risk	121	(97.6)	91	(98.9)	66	(98.5)
4. High risk	1	(0.8)	0	0.0	0	0.0
Postnatal Risk: Rank						
1. No risk	11	(8.8)	9	(9.7)	4	(6.0)
2. Unknown risk	1	(0.8)	1	(1.1)	1	(1.5)
3. Some risk	75	(60.0)	55	(59.1)	36	(53.7)
4. High risk	38	(30.4)	28	(30.1)	26	(38.8)

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Table 1 (continued)

	Bayley-III		ITSP		CBCL	
Number of Home Placements						
One	39	(31.2)	29	(31.2)	23	(34.3)
Two	44	(35.2)	35	(37.6)	26	(38.8)
Three to ten	42	(33.6)	29	(31.2)	18	(26.9)
Caregiver at Diagnosis						
Biological mother	39	(31.2)	28	(30.1)	15	(22.4)
Biological father	3	(2.4)	0	0.0	2	(3.0)
Other biological family member	28	(22.4)	23	(24.7)	16	(23.9)
Foster parent	45	(36.0)	35	(37.6)	26	(38.8)
Adoptive parent	7	(5.6)	4	(4.3)	6	(9.0)
Other/caseworker	3	(2.4)	2	(2.2)	2	(3.0)

Notes: fetal alcohol spectrum disorder (FASD); fetal alcohol syndrome (FAS); partial FAS (PFAS); static encephalopathy/alcohol exposed (SE/AE); neurobehavioral disorder/alcohol exposed (ND/AE); occipital-frontal circumference (OFC).

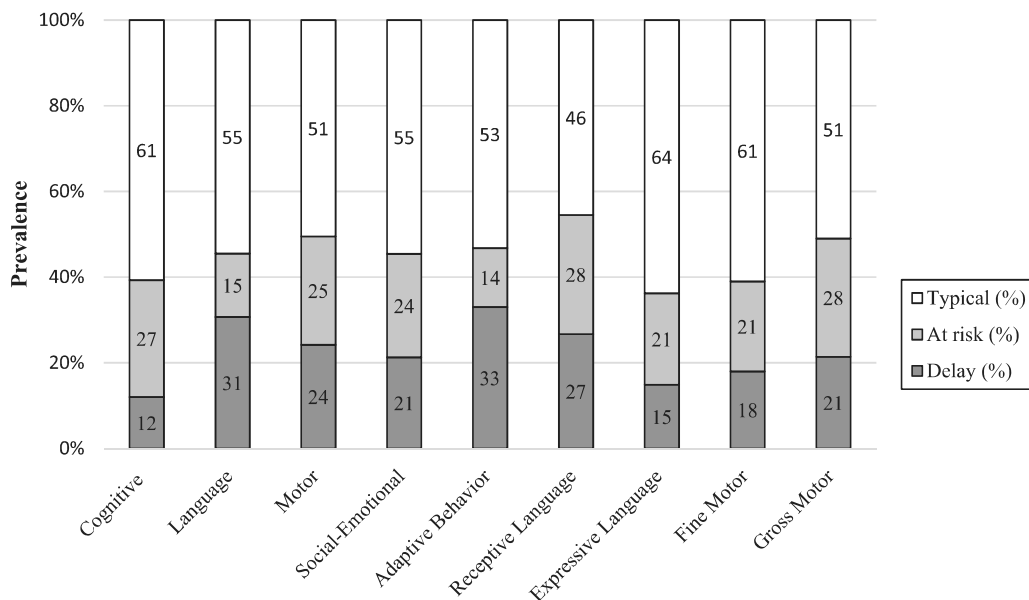
on measures of neurodevelopment, sensory processing, and emotional and behavioral problems (Research Questions 1–3). Associations between Bayley-III, ITSP and CBCL outcomes and selected child demographics and exposures (age, gender, PAE and postnatal risk factors) were examined using chi-squared tests (χ^2), *t*-tests or one-way analysis of variance (ANOVA), as appropriate (Research Question 4). Results having an alpha level of $p \leq .05$ (two-tailed) were considered statistically significant. No adjustments were made for multiple comparisons due to the exploratory nature of this study; thus, significant findings should be interpreted accordingly.

With regard to missing data, any subject with complete data on two or more Bayley-III domains and the entire CBCL were included in the analyses. For the ITSP, any subjects with missing data on more than one-third of items in any one quadrant/section were excluded. On the rare occurrences ($n = 8$) when less than a third of items were missing, the average of the child’s remaining scores was calculated and rounded to the nearest whole number. This value replaced the missing score(s) in that quadrant/section.

4. Results

4.1. Demographic and clinical outcomes

Records from 125 infants/toddlers with PAE met the inclusion/exclusion criteria for this study. All 125 had a Bayley-III administered. Sixty-one (49%) infants/toddlers had all five domains on the Bayley-III completed; 27 (22%) had 4 domains; 31 (25%) had 3



Bayley-III Domains and Subdomains (Language & Motor): FASDPN

Fig. 2. Proportion of 125 infants/toddlers with typical, at-risk, and delayed development across the five Bayley-III domains: typical (standard scores ≥ 86 , scores $\geq -0.9 SD$); at-risk (78–85, scores between -1.0 and $-1.4 SD$); delayed (≤ 77 , scores $\leq -1.5 SD$) and proportion of infants/toddlers with typical, at-risk, or delayed development in the Bayley-III Language and Motor subdomains. Receptive language: $n = 90$, expressive language: $n = 94$, fine motor: $n = 100$, and gross motor: $n = 98$. Typical (scores $\geq -0.9 SD$). At-risk (scores between -1.0 and $-1.4 SD$). Delayed (scores $\leq -1.5 SD$).

domains; and 6 (5%) had 2 domains. ITSP outcomes were available for 93 of the 125 infant/toddlers and CBCL outcomes were available on 67 infant/toddlers. The full sample of 125 ranged in age from 0.28 to 3.5 years (mean = 1.9 years), was 51% female and 46% white (Table 1). Diagnostic outcomes spanned the full continuum of FASD, with a majority of these young children receiving a diagnosis of ND/AE (60%). An overwhelming majority of our sample (90–98%) had documented exposure to prenatal and/or postnatal risks, in addition to their PAE. At the time of the assessment, 34% percent of infants/toddlers were living with their birth mother or father. The demographic and clinical profiles of the current study sample were largely representative of the entire birth to 3.5-year population evaluated in the FASDPN clinic ($n = 468$) from which they were drawn. The demographic and clinical profiles of infants/toddlers with Bayley-III, ITSP, and CBCL assessments were comparable to one another.

4.2. Research Question 1: Developmental performance on the Bayley-III

Bayley-III outcomes are presented in Figs. 2 and 3. The proportion of infants/toddlers that presented with delayed development (≤ -1.5 SDs) within each of the five domains ranged from Cognitive (12%), Social-Emotional (21%), Motor (24%), Language (31%), and Adaptive Behavior (33%). Within the Language and Adaptive Behavior domains, the most prevalent delays were observed in Receptive Language (27%) and Adaptive Behavior’s Self Care (44%).

Of the 61 infants/toddlers with complete data across all five domains, 53 (87%) had one or more domains or subdomains with a developmental delay. Of the 125 infants/toddlers with two or more domains assessed using the Bayley-III, 93 (74%) had one or more domains or subdomains with a developmental delay. Since not all domains were assessed, 74% serves as a minimal estimate.

4.3. Research question 2: Sensory processing performance on the ITSP

Ninety-three infants/toddlers ranging in age from 7–36 months had a completed ITSP. Compared to the larger group of 125 children with Bayley-III assessments, this subgroup had no infants/toddlers diagnosed with FAS. The distribution of Bayley-III domain scores by classification category (typical, at-risk, delayed) in this subgroup was, however, very similar to the distribution of Bayley-III domain scores in the full group of 125 infants/toddlers. This would suggest the subgroup of 93 was likely reasonably representative of the full study group of 125.

Within each quadrant and section of the ITSP, roughly half of the infant/toddlers presented with outcomes in the probable or definite difference range (Fig. 4), with two exceptions (i.e., sensory seeking and visual processing). The most prevalent atypical patterns observed were Low Registration (reflecting a high threshold for sensory input and use of passive strategies to respond) and Auditory Processing (reflecting an inadequate ability to modulate sounds representing over or under responsiveness). Of the 93 infants/toddlers who completed the ITSP, all were rated with a definite difference in at least one quadrant/section of the ITSP.

4.4. Research question 3: Emotional and behavioral functioning on the CBCL

Sixty-seven infants/toddlers ranging in age from 18–42 months had a completed CBCL assessment. Compared to the larger group of

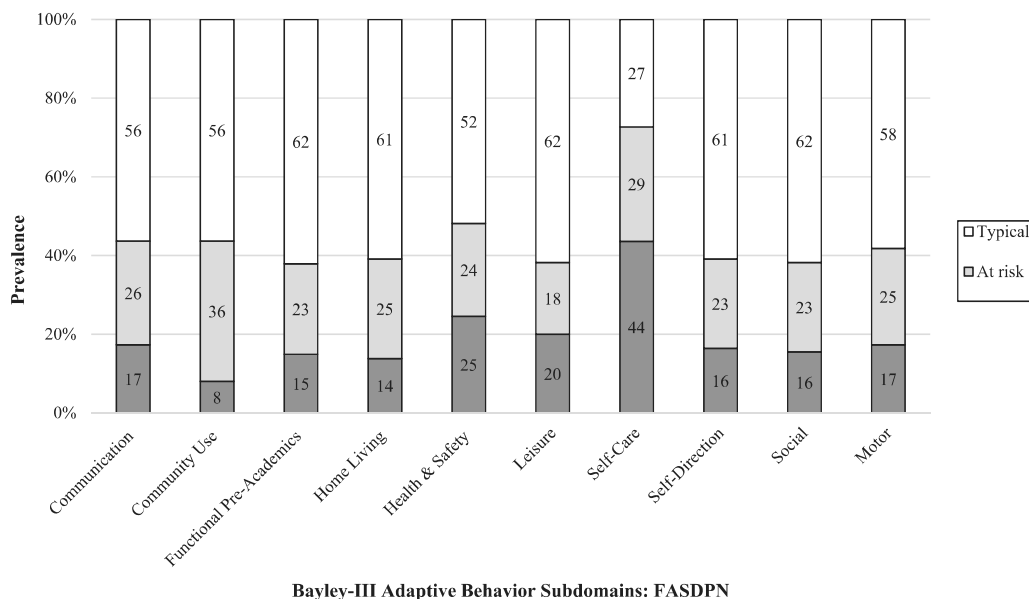
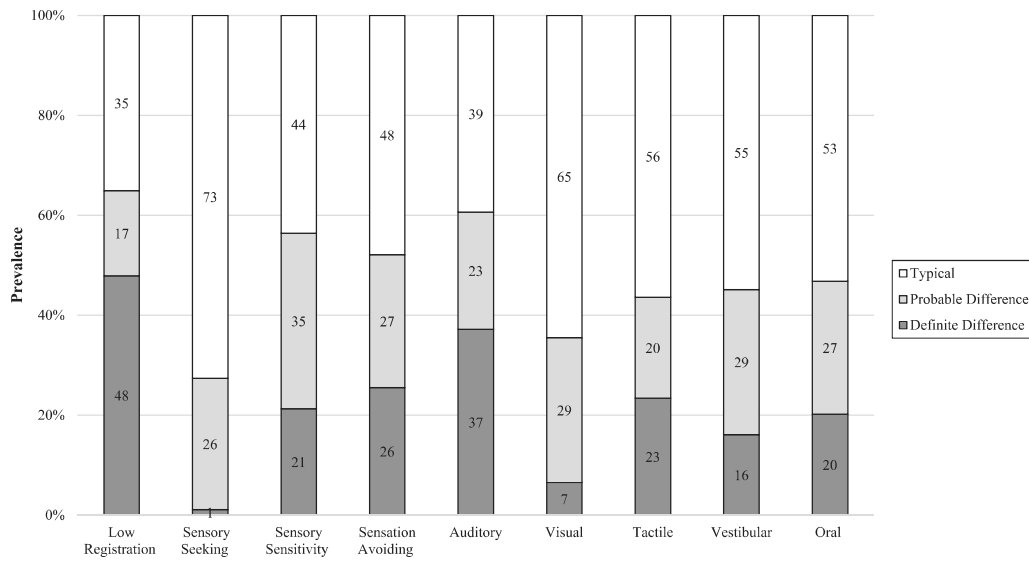


Fig. 3. Proportion of infants/toddlers with typical, at-risk, or delayed development in the Bayley-III Adaptive Behavior subdomains. Communication: $n = 110$, community use: $n = 87$, functional pre-academics: $n = 87$, home living: $n = 87$, health & safety: $n = 110$, leisure: $n = 110$, self-care: $n = 110$, self-direction: $n = 110$, social: $n = 110$, and motor: $n = 110$. Typical (scores ≥ -0.9 SD). At-risk (scores between -1.0 and -1.4 SD). Delayed (scores ≤ -1.5 SD).

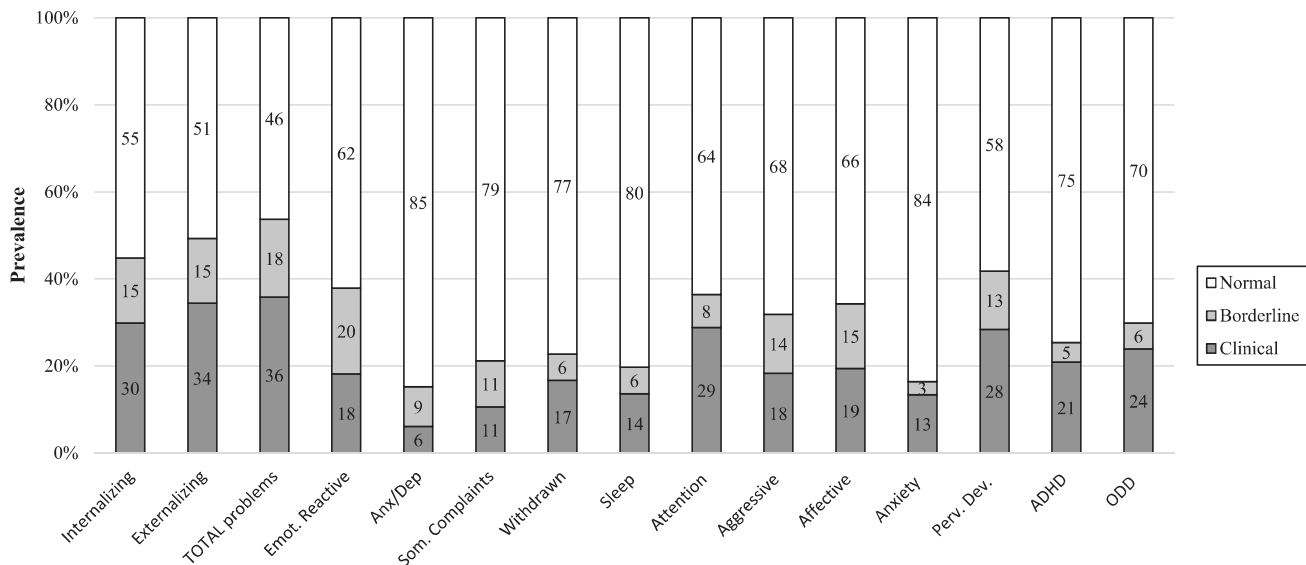


Infant Toddler Sensory Profile Quadrants and Sections: FASDPN

Fig. 4. Proportion of 93 infants/toddlers with typical performance, probable difference, or definite difference across the four ITSP quadrants (low registration, sensory seeking, sensory sensitivity, sensory avoiding) and five section (auditory, visual, tactile, vestibular, and oral sensory processing). Typical performance (scores at or between plus 1.0 SD and minus 1.0 SD), probable difference (scores within -1.1 to -1.9 SDs or within +1.0 to +1.9 SDs), and definite difference (scores at or below -2 SDs or at or above +2 SDs).

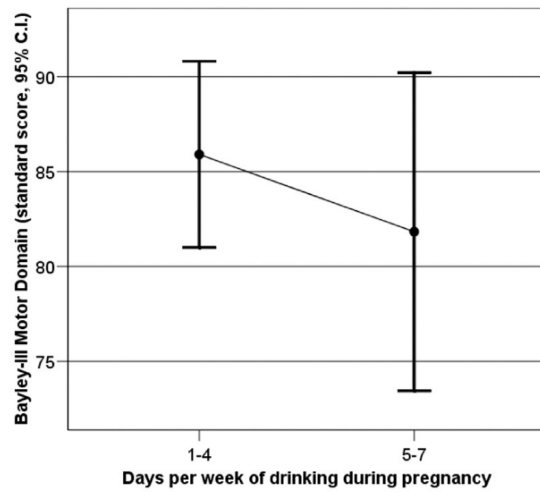
125 infants/toddlers, this subgroup had no children diagnosed with FAS and only one child with PFAS. The distribution of Bayley-III domain scores by classification category (typical, at-risk, delayed) in this subgroup was comparable to the full group of 125 infants/toddlers.

Approximately half of the infants/toddlers presented in the borderline or clinical range on the Internalizing, Externalizing and/or Total problem scales (Fig. 5). Attention Problems had the highest prevalence of elevated scores on the Syndrome scales (37%) and Pervasive Developmental Problems had the highest prevalence of elevated scores on the DSM-Oriented Scales (42%; Fig. 5). Of the 67

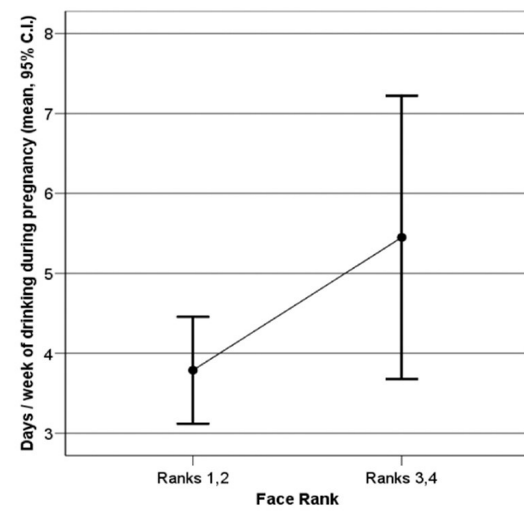


CBCL/1.5-5 years Summary, Syndrome and DMS-Oriented Scales: FASDPN

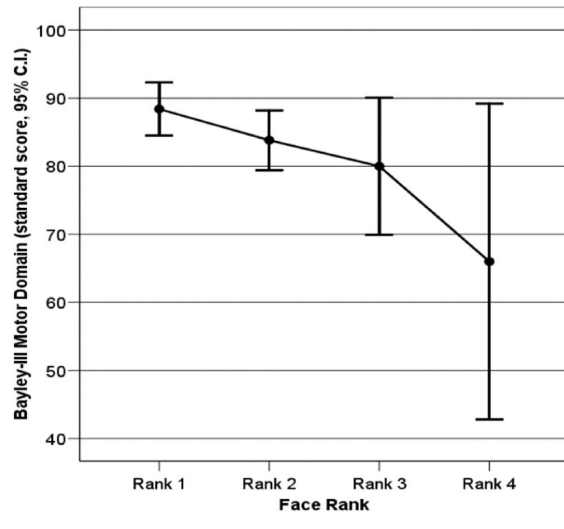
Fig. 5. Proportion of 67 infants/toddlers with scores in the normal, borderline clinical and clinical range across three Summary Scales (Internalizing, Externalizing and Total problems), seven Syndrome Scales (Emotionally Reactive, Anxious/Depressed, Somatic Complaints, Withdrawn, Sleep, Attention, and Aggressive problems) and five DSM-Oriented Scales (Affective, Anxiety, Pervasive Developmental, Attention Deficit Hyperactivity, and Oppositional Defiant problems) of the CBCL. Summary *t*-scores are categorized as Normal (*t*-scores < 60), Borderline (*t*-scores 60 to 63) and Clinical (*t*-scores ≥ 64). Syndrome and DSM-Oriented Scale *t*-scores are categorized as Normal (*t*-scores ≤ 64), Borderline (*t*-scores 65 to 69), and Clinical (*t*-scores ≥ 70).



A.



B.



C.

(caption on next page)

Fig. 6. Mean scores across all Bayley-III domains and subdomains (with the exception of Adaptive Behavior) decreased with increasing PAE. Using the Motor domain as an example: A. The mean Bayley-III Motor score decreased with increasing number of days/week of drinking during pregnancy ($t = 0.9, p = 0.36$). Only 60 infants/toddlers had days/week of exposure reported, limiting the statistical power to identify significant outcomes. B. Face Rank is a proxy measure for PAE and was available on all 125 infants/toddlers. Infants/toddlers with Face Ranks 3 and 4 had significantly higher days/week of PAE than infants/toddlers with Face Ranks 1 and 2 ($t = -2.0, p = .05$). C. Using Face Rank as a proxy measure for PAE, mean Bayley-III Motor scores decreased significantly with increasing Face Rank (ANOVA F linear term 14.3, $p = .000$). Mean scores across all Bayley-II domains decreased with increasing PAE.

infant/toddlers with CBCL data, 29 (43%) scored in the clinical range on the Internalizing, Externalizing and/or Total Problem scales.

4.5. Outcomes spanning all three measures

Two additional exploratory analyses were conducted to document the proportion of infants/toddlers who presented with a clinically significant delay ($\leq -1.5 SDs$) in one or more areas of the Bayley-III, ITSP and CBCL. Of the 31 infants/toddlers with full data across all three assessments, 30 (97%) presented with a clinically significant delay in at least one area of the Bayley-III, ITSP and/or CBCL, while 17 (55%) presented with a clinically significant delay across all three assessments. For the entire sample of 125 infants/toddlers (including those with complete and incomplete data), 124 (99%) presented with a clinically significant delay in at least one area of the Bayley-III, ITSP and/or CBCL.

4.6. Research Question 4: Associations between independent variables (PAE, other postnatal risk factors, gender and age) and dependent variables (Bayley-III, ITSP, CBCL)

Mean scores across all Bayley-III domains and subdomains (with the exception of the Adaptive Behavior domain) were lower (although not significantly lower) among those infants with 5–7 days/week of PAE compared to those with 1–4 days/week of PAE (Fig. 6A). Decades of analyses conducted with FASDPN data has shown that the greater the number of days/week of drinking during pregnancy (i.e., 5–7 days/week versus 1–4 days/week), the more severe the FAS Facial Rank (Astley, 2010, 2013). Only 62 infants had days/week PAE reported, limiting the statistical power to identify significant associations. Previous research has confirmed the FAS Facial Rank serves as an accurate proxy measure of PAE. Data from the first 1400 patients diagnosed at the WA FASDPN document the more severe the 4-Digit FAS facial phenotype (Facial Ranks 1–4), the greater the number of days/week of drinking during pregnancy (significant linear trend, $F=10.7, p = 0.001$) (see Figure 10 from Astley, 2013). Since this same association was observed in the current study (Fig. 6B), the FAS facial rank was used as a proxy for PAE in the current study. Unlike the limited number of infants/toddlers with days/week of PAE reported, all 125 infants/toddlers had a Facial Rank. All Bayley-III domain and subdomain scores (with the exception of Adaptive Behavior) decreased significantly with increasing severity of the FAS facial phenotype (as demonstrated in Fig. 6C for the Motor domain). Domain standard scores decreased roughly 10–20 points from Face Rank 1 to Face Rank 4. Subdomain

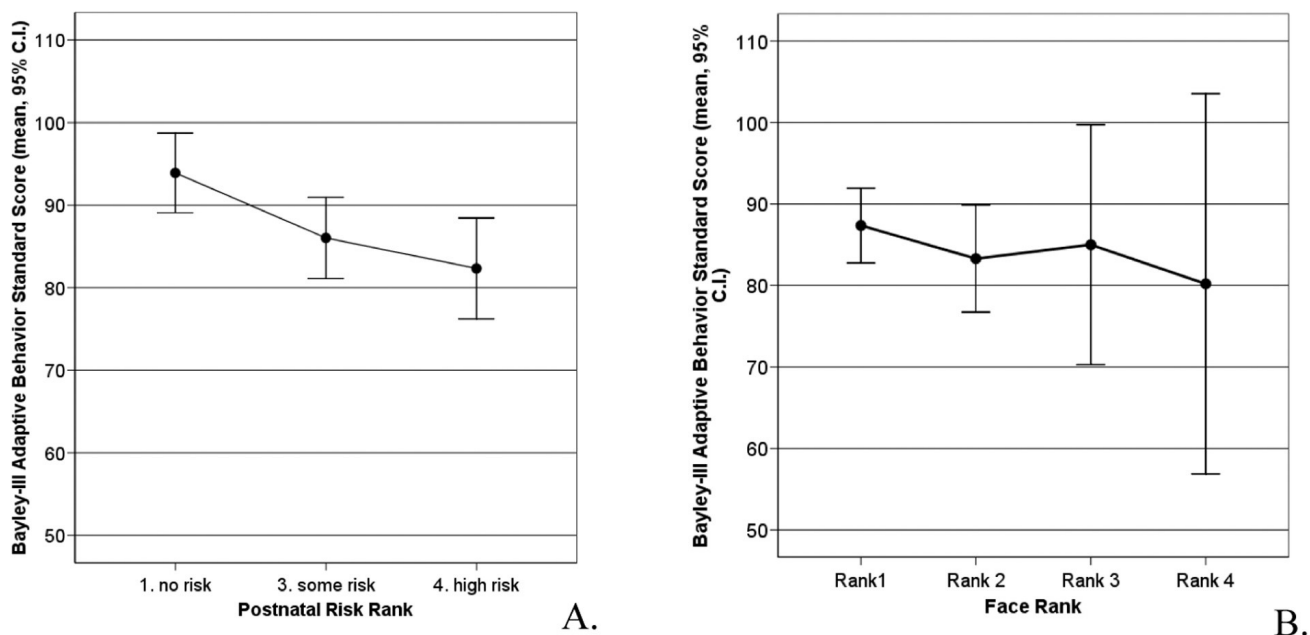


Fig. 7. Impairment in Adaptive Behavior appeared to be more strongly associated with postnatal risk factors than PAE. A. The mean Bayley-III Adaptive Behavior standard score decreased approaching statistical significance with increasing severity of the postnatal risk rank (Linear Term: $F = 1.7, p = 0.067$). B. The mean Bayley-III Adaptive Behavior standard score did not decrease linearly with Face Rank (a proxy measure for PAE) (Linear Term: $F = 0.6, p = 0.46$).

scaled scores decreased roughly 2–4 points from Face Rank 1 to Face Rank 4. Fig. 7.

The magnitude of postnatal risk was also explored with outcomes across the Bayley-III domains. Interestingly, the mean scores in the Adaptive Behavior domain decreased with increasing levels of postnatal risk, and approached significance (ANOVA: $F = 3.4$, $p = .067$). This is in contrast to mean scores in the other developmental domains whereby scores decreased relative to PAE, but not for the postnatal risk rank. The number of home placements, as a proxy measure of postnatal risk, were also explored relative to Bayley-III outcomes. Mean Cognitive and Expressive Language scores were significantly inversely correlated with the number of home placements ($r = -.18$, $p = .049$ and $r = -.21$, $p = .041$, respectively).

To explore developmental competencies across the early intervention years, two age categories were created (2–23 months and 24–42 months). Limited sample sizes precluded the use of four age categories (i.e., one for each year of life). Delays in the Language and Motor domains were significantly more prevalent (45% vs 10%, $X^2 = 12.8$, $p = .002$ and 35% vs 11%, $X^2 = 8.3$, $p = .016$ respectively) among the younger age group (2–23 months) than the older age group (24–42 months). Conversely, Social-Emotional delays were significantly more prevalent among the older age group relative to the younger group (28% vs 15%, $X^2 = 6.0$, $p = .049$). Mean Language domain scores (89, $SD 13.9$) were significantly higher among females than males (83.2, $SD 13.3$) ($t = -2.0$, $p = .048$).

Limited sample sizes precluded in-depth analyses of three ITSP quadrants/sections because they were divided into multiple age categories (i.e., Sensation Seeking, Tactile and Oral Sensory processing). However, for those remaining quadrants/sections without multiple age categories (i.e., Low Registration, Sensory Sensitivity, Sensation Avoiding, Auditory, Visual and Vestibular processing), no significant associations were found for age, gender, PAE, postnatal risk, and number of home placements.

The prevalence of emotional and behavioral problems did not vary significantly by age, gender, PAE, or number of home placements. However, scores on the Withdrawn and Oppositional Defiant Problems scales were positively correlated with increasing level of postnatal risk rank ($r = .25$, $p = .048$ and $r = .28$, $p = .024$ respectively).

5. Discussion

This research is notable as it is the first paper to describe a wide range of developmental, sensory, and behavioral outcomes in a very young sample of children with confirmed PAE from a specialty clinic for individuals with PAE. Of the 31 infants/toddlers with full data across all three assessments, 30 (97%) presented with a clinically significant delay in one or more areas of the Bayley-III, ITSP and/or CBCL, while 17 (55%) presented with one or more clinically significant delays in each of the three assessments. For the entire sample of 125 infants/toddlers (including those with complete and incomplete data), 124 (99%) presented with a clinically significant delay in one or more areas of the Bayley-III, ITSP and/or CBCL. These findings highlight the substantial diversity and prevalence of challenges experienced by infants/toddlers with PAE during the early intervention period.

5.1. Developmental outcomes and implications

Within each of the five Bayley-III domains, roughly half the infant/toddler study population had at-risk or delayed development. The prevalence of delayed development was lowest in the Cognitive (12%) and Motor (24%) domains, higher in the Language domain (31%) and highest in the Adaptive Behavior domain (33%). This pattern of delayed development was remarkably comparable to the pattern observed among the first 1400 patients with PAE (infant to adult) evaluated in the FASDPN clinic through 2005 (Astley, 2010) and among the 2283 patients with PAE evaluated at the FASDPN through 2020 (infant to adult) presented on the FASDPN Tableau Dashboard (Hemingway, 2022a). While the pattern of developmental delay (from least prevalent to most prevalent) is comparable between this infant/toddler study sample and our predominantly older childhood/adult FASD clinical population, the prevalence of severe impairment (2 or more SDs below the mean) in each domain is roughly two-fold greater in the older childhood/adult population (cognition 24%, motor 25%, language 39%, adaptive behavior 59%) (Hemingway, 2022a). Given that older children and adults have more mature neuropsychological function and can be assessed with more sophisticated neuropsychological instruments, one might expect to detect a higher prevalence of impairment when using these more sensitive instruments (Clarren et al., 2000). These findings provide evidence that a global developmental measure, such as the Bayley-III, may be useful for identifying early indicators of delay among infants/toddlers with PAE. Early identification leads to early intervention. A future longitudinal study is planned to assess whether the Bayley-III predicts which individual infant/toddlers go on to present with cognitive, language, motor, social emotional and/or adaptive behavior impairments later in childhood.

In this clinical sample of infants/toddlers, significant correlations were observed between PAE and developmental delay. Findings demonstrated that Bayley-III domain and subdomain scores (with the exception of Adaptive Behavior) decreased significantly with increasing levels of PAE, which further supports known trends on the gradient effect of alcohol on severity of outcomes (Astley, 2013; Carr et al., 2010; Subramoney et al., 2018). Individuals with PAE typically present with a multitude of other prenatal and postnatal risk factors that likely contribute, at least in part, to their adverse outcomes. Hemingway et al. (2020) reported other prenatal and postnatal risk factors were 3 to 7-fold more prevalent in the FASDPN clinical population than in the general population. Ninety percent of this infant/toddler population presented with adverse prenatal and postnatal risks. Significant correlations were observed between postnatal risks including multiple home placements and adaptive behavior, cognition, and expressive language delays. Similar to literature showing associations between early adverse experiences and poorer functioning in the general population (Garner & Shonkoff, 2012; van der Kolk, 2003), and among individuals with PAE (Coggins et al., 2007; Hemingway et al., 2020; Price et al., 2017; Streissguth et al., 2004), our analyses revealed a significant correlation between other postnatal risks and adaptive behavior delays. This suggests that the postnatal environment may have an impact on these developing behaviors, warranting further research since

adaptive behaviors are a lifelong disability and because there continue to be ongoing questions about the impact of biological vs environmental risks in this complex population of children.

5.2. Sensory processing outcomes and implications

Atypical sensory processing behaviors were observed in a large proportion of infants/toddlers. Based on our work with older children with PAE (Jirikowic et al., 2020; McLaughlin et al., 2019), the sensory processing patterns most impacted were low registration (65%) and auditory processing (61%). In general, infants/toddlers in this study had a high threshold for sensory input (e.g., does not notice stimuli easily) and used passive strategies to regulate (e.g., remains in situations that are uncomfortable rather than controlling for the amount and type of input). Infants/toddlers also showed a decreased capacity to modulate sound, as evidenced by ratings of over-responsiveness or under-responsiveness to auditory input. Definite differences in low registration and auditory processing were similarly reported in a clinic-referred sample of infants/toddlers with PAE, with even more severe impacts in those diagnosed with FASD (Fjeldsted & Xue, 2019). Current findings were consistent with those using FASDPN clinical data (Jirikowic et al., 2020; McLaughlin et al., 2019), showing that preschool and school-age children with FASD had the highest proportions of definite differences in Auditory Filtering and Under-responsive/Sensation Seeking domains using the Short Sensory Profile (SSP; McIntosh et al., 1999). Although an exact comparison cannot be made between ITSP and SSP domain categories, findings suggest that a caregiver-reported measure of sensory processing, such as the ITSP, may be useful for identifying sensory processing differences among infants/toddlers with PAE. Importantly, a child's ability to engage and participate successfully in everyday life, including forming healthy attachment relationships, is closely tied to their sensory processing abilities (Dunn, 2007). When early intervention providers and families have a working knowledge of sensory processing, they can reframe their understanding of their child's behavior and develop appropriate intervention strategies.

5.3. Emotional and behavioral functioning outcomes and implications

Atypical emotional and behavioral problems on the CBCL were observed in a large proportion of infants/toddlers. Results are consistent with prior research demonstrating that problem behaviors co-occur in older children with FASD (Astley, 2010; Astley et al., 2009; Franklin et al., 2008; Jirikowic et al., 2008). Notably, the prevalence of Total Problem scores in the clinical range for the older children (5–10 years of age) (86%; Franklin et al., 2008) and 79% among 997 children 6–18 years of age from the FASDPN clinic (Hemingway, 2022b) were much higher in comparison to the current sample of infants/toddlers (birth to 3.5 years of age) with PAE (36%). The prevalence of Attention Problems in the clinical range (61% among 730 children 6–18 years of age from the FASDPN clinic (Hemingway, 2022b) was 3-fold greater than the prevalence (21%) among the infant/toddlers in the current study. A future longitudinal study, with larger numbers and a comparison group, would add significantly to the literature on the trajectory of emotional and behavioral outcomes in young children with PAE and the protective and risk factors associated with these outcomes.

5.4. Limitations

This study had a number of potential limitations. First, because this was a retrospective chart review using diagnostic clinical data, our results are limited by the clinical data available. Missing data due to a flexible clinical assessment protocol and some incomplete caregiver-report measures contributed to uneven datasets for each infant/toddler, thus limiting the analysis of outcomes at the individual level. Future studies, with a larger prospective sample and assessments spanning similar age ranges, could examine neurodevelopmental profiles in relation to sensory processing differences and problem behavior. Additionally, further research could examine how factors such as attachment relationships, family engagement or early intervention impact infant/toddlers' emerging, and declining competencies. Second, this was a clinic-referred sample and might not represent all infants/toddlers with PAE. It is important to note that the FASDPN clinic does not require patients to present with a concern or delay in order to receive an FASD evaluation; they only need to have a confirmed PAE at any level. For this reason, our study sample may more closely resemble the broader population of infants/toddlers with PAE compared with other clinic-referred samples. Third, the ITSP, CBCL and Adaptive Behavior components of the Bayley-III are standardized measures based on caregiver report, which are inherently susceptible to reporting bias. Nevertheless, researchers working towards the earlier identification of children with PAE (Astley, 2010; Bakhireva et al., 2018), and children with autism spectrum disorders (Zwaigenbaum & Maguire, 2019), advocate for the use of caregiver-reported assessment to identify early appearing problems in development and behavior. One additional limiting factor to consider is the potential for cohort effects, given that data from this sample of infants/toddlers were collected over a 10-year time span (2009–2019). No annual temporal trends were observed from 2009 to 2019 across any of the following study population characteristics: number of infant/toddlers diagnosed, gender, race, FASD diagnostic outcome or level/rank of prenatal alcohol exposure.

6. Conclusion

An overwhelming majority of infants/toddlers with PAE in this sample presented with clinically significant delays in development, sensory processing and/or emotional and behavioral functioning. Present findings, considered with similar studies reported in the literature, suggest that most domains of child functioning are vulnerable to the teratogenic impact of PAE and that these delays are evident in the first years of life. Findings reinforce the value of early screening, ongoing monitoring, and comprehensive assessment to facilitate earlier identification and to provide opportunities for infants/toddlers with PAE and their caregivers to benefit more fully

from early supports and intervention.

CRedit authorship contribution statement

Jirikowic Tracy: Conceptualization, Formal analysis, Supervision, Writing – original draft, Writing – review & editing. **Pruner Misty:** Conceptualization, Formal analysis, Methodology, Writing – original draft, Writing – review & editing. **Astley Hemingway Susan J.:** Conceptualization, Data curation, Formal analysis, Methodology, Supervision, Writing – original draft, Writing – review & editing. **Baylor Carolyn:** Conceptualization, Formal analysis, Methodology, Supervision, Writing – original draft, Writing – review & editing.

Declaration of Competing Interest

None

Data Availability

The data that has been used is confidential.

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