Toward Earlier Identification and Strengths-Based Intervention for Infants and Toddlers with Prenatal Alcohol Exposure: Evidence from the Washington State Fetal Alcohol Syndrome Diagnostic & Prevention Network Clinical Database

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Abstract

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Chair of the Supervisory Committee:
Professor Tracy Jirikowic
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The purpose of this two-article dissertation was to describe the early developmental outcomes of infants/toddlers with prenatal alcohol exposure (PAE) based on results from 10 years of retrospective clinical data, toward the goal of earlier identification. Although there is extensive research on school-age children and youth, there is a limited body of research describing the developmental delays, sensory processing differences and challenging behaviors among infants/toddlers with PAE. In addition, little research has focused on the positive attributes and strengths of young children with PAE, which can provide direction for a strengths-based approach to assessment and intervention. The first article in the dissertation, "Developmental, Sensory and Behavioral Outcomes Among Infants and Toddlers with Prenatal Alcohol Exposure" was a retrospective analysis of diagnostic clinical data from the University of

Washington Fetal Alcohol Syndrome Diagnostic and Prevention Network (FASDPN). Descriptive statistics were used to document the proportion of infants/toddlers classified with typical, at-risk, or delayed development on three measures used in the FASD diagnostic evaluation. Empirical analyses were conducted to examine relationships between outcomes, PAE and/or other postnatal risk factors. Results showed that a majority of infants/toddlers presented with clinically significant delays in development, sensory processing and/or behavioral functioning. Adverse developmental outcomes were significantly correlated with PAE and/or postnatal risk factors. Present findings, considered with similar studies reported in the literature, 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. The second article in the dissertation, “Concerns and Strengths: Caregiver Perceptions of Their Infant/Toddler with Prenatal Alcohol Exposure”, described caregiver-reported concerns and strengths in the same clinical sample of infants/toddlers. Using a directed approach to content analysis, caregivers’ written responses to open-ended questions on two parent-report questionnaires were coded and analyzed. Results indicate the caregivers’ most frequently reported concerns were related to aggressive behavior, language/communication, and sensory processing. Frequently reported strengths included happiness, sociability, and love. These findings demonstrate the value of eliciting and understanding caregiver perspectives as an aid to identifying the needs of infants/toddlers with PAE within the context of their families, and for informing a strengths-based approach to assessment and intervention.

This research was conducted with the approval of the Human Subjects Division

of the

University of Washington

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Plain Language Summary

Why is early identification important for young children with prenatal alcohol exposure?

The first three years of life are the most important for a child's brain development. For children whose mothers drank alcohol during pregnancy, development and learning can be interrupted by damage to the brain. For some young children with prenatal alcohol exposure (PAE), paying attention or behaving in ways their caregivers expect them to can be hard. Some young children may also have problems forming close relationships with their caregivers. When children with PAE are identified early, they can receive special intervention at a time when the greatest gains in the brain are possible. Therapists who provide intervention can help children learn new skills and give support to caregivers.

What was the focus of this research study?

This study focused on describing the overall development of children who were 3.5 years or younger, with PAE, at the time of their diagnosis. To help with diagnosis and to learn more about how a child was developing, three tests were given: one test that measured overall development (cognitive, language, motor, social-emotional, and everyday life skills), one test that measured how well a child manages everyday sensory experiences, and one test that measured challenging behaviors. Caregivers were also asked the following questions in two of the tests: "What are your concerns?" and "What are your child's strengths?". The researchers wanted to understand areas of difficulty and areas of strength for these young children.

What did the researchers find out about child development?

Nearly all the children in our study had some degree of delay in development, sensory processing, and/or behavior. Results showed that children who scored worse on the tests tended to be exposed to more alcohol prenatally or were exposed to other risks such as neglect, abuse, or lived in more than one home.

What did the researchers find out about child concerns and strengths?

Parents described a range of concerns about their child. Concerns that came up most often were about aggressive behavior, language and communication problems, and difficulties with managing sensory information. Parents also believed their child had a lot of positive traits and strengths. Child strengths that came up most often were about happiness, being friendly and outgoing, and being loving and lovable.

What are the key takeaways from these research results?

Early childhood providers (doctors, therapists, childcare workers) can help identify young children with PAE who are at risk for developing problems. They can help with developmental monitoring: watching how a child grows and changes over time and noticing whether they are meeting typical milestones in playing, learning, speaking, behaving, and moving. We also learned that parents have a good sense of their child's challenges and strengths. Providers can make the most of child strengths in their work with children and families.

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I would like to express my gratitude to all the people who have supported me on this journey. In particular, I would like to thank Dr. Tracy Jirikowic, my Chair and my mentor. I have felt her support every step of the way for the past five years. Thank you also to Dr. Susan Astley Hemingway and Dr. Carolyn Baylor for your invaluable work and support towards this research. I have learned so much. Thank you also to my committee members Drs. Sue Spieker, Chun Wang and Holly Schindler, who provided timely and helpful feedback. I would also like to acknowledge Dr. Deborah Kartin for always believing in me. Thank you also to my PhD cohort, an amazing group of people who I am lucky to have gone through the program with. I continue to learn from and be inspired by all of you.

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Chapter 1. General Introduction

1.1 Background

Prenatal alcohol exposure (PAE) can disrupt the architecture of the developing brain and interfere with a child's developmental progress. Child development challenges can have an impact on child and family well-being. Children with fetal alcohol spectrum disorders (FASD), an umbrella term representing the full range of physical, cognitive, and behavioral impairments associated with PAE, may experience challenges that begin early and persist across the lifespan (Chudley et al., 2005). FASD is a world-wide public health problem, impacting individuals and families across all demographic groups (Popova et al., 2019). FASDs are estimated to occur in at least 1% of children and youth in the general population (Lange et al., 2017), with disproportionately higher rates among children in foster care (Popova et al., 2019).

The Benefits of Early Intervention

Developmental and intervention science have documented the short and long-term benefits of early intervention on child development and family well-being for children who are experiencing or are at risk for delays in the early childhood period (Guralnick, 2011; 2019). The capacity of the brain to learn from experience is greatest during these first three years of life (Shonkoff & Garner, 2012), thus creating a prime opportunity for children to benefit from an optimally supportive environment. As mandated by Part C of the Individual with Disabilities Education Improvement Act (IDEA, 2004), early intervention services optimize child development by promoting stable and responsive relationships, enriching learning environments and strengthening family adaptation (Adams & Tapia, 2013). Given that children with PAE are at risk for a wide range of developmental (Subramoney et al., 2018), regulatory (Bakhireva et al, 2018; Chen et al., 2012; Molteno et al., 2014), sensory (Fjelstad & Xue, 2019; Wengel et al., 2011), and relational (O’Connor et al., 2002; Paley et al., 2006) challenges, early intervention’s comprehensive and family-centered approach is particularly important in this population (Olson et al., 2007; Olson & Montague, 2011). For children at significant risk, intervening early can buffer against the developmental vulnerability and adverse environmental influences associated with PAE and optimize child and family outcomes in the long term. Understanding early developmental and behavioral profiles, as well as caregiver-reported concerns and strengths, can aid in the earlier identification and strengths-based intervention for infants/toddlers with PAE.

1.2 Overview of the Literature

Children with PAE and FASD have a wide range of neurodevelopmental risks and challenges, including impairments in cognitive, communication, motor, social-emotional and adaptive functioning (Astley & Clarren, 2000; Astley, 2019; Reid et al., 2015). Delays in any one of these domains may impact on children’s ability to participate successfully in everyday life and their caregivers’ ability to provide sensitive and responsive care (Mattson et al., 2019; Pluess & Belsky, 2010; Olson et al., 2007; Olson & Montague, 2011)). Young children with PAE also experience sensory processing differences (Fjeldsted & Xue, 2019; Jirikowic et al., 2008, 2020), described as a decreased capacity to notice, interpret, and respond to sensory input in one or more sensory domains. Sensory abnormalities compromise a child’s ability to respond to sensory information in an adaptive, age-typical manner and interfere with healthy development of emotional regulation (Dunn, 2007). Emotional and behavioral problems related to altered development and other environmental risks such as early adversity and trauma, are also common among young children with PAE (Astley 2010; Astley et al., 2009; Astley, 2019b; Subramoney et al., 2018). An overview of relevant research findings on neurodevelopmental, sensory, and behavioral outcomes among infants and toddlers with PAE are summarized in the following section.

Cognitive Development

Early cognitive development comprises several skills including attention, memory, imitation, information processing, problem-solving, and executive functioning, and lays the foundation for success in learning, school achievement and relationships (McClelland & Tominey, 2014; Weiss et al., 2010). Evidence regarding the cognitive development of infants/toddlers with PAE show that deficits are not uniform. Findings show that some infants/toddlers with PAE exhibit global delays in intellect and learning ability (Astley, 2019a; Coles et al., 2015; Garrison et al., 2019; O'Connor et al., 1993), while other studies document more specific delays including problems with attention regulation, information processing and efficiency (Jacobson, 1998; Kable & Coles, 2004; Kable et al., 2016). Meanwhile, deficits in higher-order cognitive processes such as intelligence quotient (IQ), memory and executive functioning have been documented among older children with PAE and FASD (Astley, 2010, 2019; Astley et al., 2009; Janzen et al., 1995; Kodituwakku, 2009; Mattson et al., 2019; Olson & Montague, 2011).

Language Development

Early language development reflects a young child's ability and desire to communicate with others, both verbally and non-verbally (Zero to Three, 2016). In a review of the literature, studies investigating language outcomes found evidence of lower language scores in infants 6-12 months (Coles et al., 2000) and toddlers 24-30 months of age (Autti-Rämö et al., 1992; O'Leary et al., 2009) with moderate to heavy PAE. In one study of toddlers with lower levels of PAE, delays in language development were not reported (O'Leary et al., 2009). Hanlon-Dearman &

colleagues (2020) found expressive and receptive language impairments in preschoolers with PAE (ages 3-6 years), with poorer language abilities noted in children diagnosed with FASD compared with PAE only. Research conducted with school-aged children with FASD (ages 6-12 years), demonstrate language and communication deficits, especially difficulties with social functioning (Astley, 2019; Astley et al., 2009; McGee et al., 2009; Coggins et al., 2003; Mattson et al., 2019).

Motor Development

Gross and fine motor development in the first few years of life includes skills that allow a child to move and explore their environment, participate in play and social activities, and work towards independence in self-care tasks (Doney et al., 2014; Weiss et al., 2010). A few studies examining motor outcomes among infants/toddlers with PAE found a dose-response relationship, where higher levels of PAE were associated with an increased likelihood of motor delays (Autti-Ramo & Granstrom, 1991; Davies et al., 2011; Jacobson et al., 1993). Delays in gross motor (i.e., sitting, standing, walking) and fine motor (i.e., visual-motor, steadiness) development were observed in infants, ages 6-18 months (Autti-Ramo & Granstrom, 1991; Davies et al., 2011; Fried & Watkinson, 1990), toddlers ages 2-3 years (Kalberg et al., 2006; Kaplan-Estrin et al., 1999) ,preschoolers (Hanlon-Dearman et al., 2020) and individuals with PAE and FASD across the age span (Astley, 2010). Research involving school-aged children with PAE or FASD (ages 4-12 years) found that fine motor impaiments were more likely to be associated with moderate to high levels of PAE (Doney et al., 2014). Similarly, gross motor deficits (i.e., balance, coordination and ball skills) were significantly associated with moderate to heavy PAE in children (birth to 18 years) (Lucas et al., 2014).

Social-Emotional Development

Social-emotional development refers to a child's ability to experience, express and manage a full range of emotions, and to form satisfying and trusting relationships with others (Parlakian & Seibel, 2002; Weiss et al., 2010). To the best of our knowledge, no studies could be found using the Bayley-III Social-Emotional scale or other skill-based developmental standardized test to assess the attainment of these important age-related milestones in this clinical population. However, problems related to emotional and behavioral functioning and early regulatory skills, which are inextricably linked to social-emotional development, have been documented in the birth to three population, including fewer social monitoring behaviors (Jirikowic et al., 2016), emotional withdrawal (Molteno et al., 2014), decreased arousal and activity level (Bakhireva et al., 2018; Oberlander et al., 2010), difficult temperament (Alvik et al., 2011), negative affect (Bakhireva et al., 2018) and increased stress reactivity (Haley et al., 2006; Jirikowic et al., 2016). Further, an extensive number of studies with preschoolers and school age children with PAE and FASD report problems related to attention (Astley, 2010, 2019b; Astley et al., 2009; Mattson et al., 2011), negative affect and depression (O'Conner & Paley, 2006) and a variety of externalizing behaviors (Franklin et al., 2008; O'Connor & Paley, 2009; Fryer et al., 2007).

Adaptive Behavior

Adaptive behavior involves the ability to manage the demands of the environment and meet every day needs (Harrison & Oakland, 2003). Problems in this domain have been documented among infants/toddlers with PAE, and include deficits in communication, daily living skills and socialization behavior (Garrison et al., 2019; Whaley et al., 2001). Findings from studies with preschool and school-age children with FASD clearly demonstrate that adaptive behavior delays persist and increase through childhood (Astley, 2010, 2019a; Astley et al., 2009; Carr et al., 2010; Hanlon-Dearman et al., 2020; Jirikowic et al., 2008; Whaley et al., 2001).

Sensory Processing

Atypical sensory processing can be characterized as a decreased capacity of the central nervous system (CNS) to detect, modulate, interpret, or respond to sensory input from the environment and the body (Miller et al., 2007; Dunn, 2007). Among infants/toddlers with PAE and FASD, differences in auditory processing (reflecting an inconsistent ability to modulate sounds with over- or under-responsiveness) and low registration (reflecting a high threshold for sensory input and use of passive strategies to respond) have been reported (Fjeldsted & Hanlon-Dearman, 2009; Fjeldsted & Xue, 2019). Similar sensory processing differences have been observed among preschool and school age children with PAE and FASD (Abele-Webster et al., 2012; Jirikowic et al., 2020; McLaughlin et al., 2019). In addition, studies with school-age children with FASD have documented sensory processing deficits correlated with problem behaviors (Franklin et al., 2008), sleep disturbances (Wengel et al., 2011), decreased adaptive function (Jirikowic et al., 2008; Carr et al., 2010) and decreased academic performance (Jirikowic et al., 2008).

Risk and Protective Factors

Risk and protective factors play a vital role in shaping a child’s development and family functioning (Guralnick, 2011). The likelihood of positive social, emotional, and physical well-being will be increased when protective factors are promoted, and risk factors are decreased. Responsive caregiving and a stable, nurturing home have been identified as environmental influences that buffer children from the disruptive effects of PAE (Olson et al., 2009). Supportive relationships promote whole child development and help strengthen resilience, especially for children growing up in adverse circumstances (Shonkoff, 2017). A positive home environment may also reduce the risk of children and adolescents with FASD experiencing challenges later in life, such as difficulty at school or work, being able to live independently, or becoming involved with the criminal justice system (Streissguth et al., 2004). Yet, many children with PAE or FASD are at increased risk for adverse childhood experiences, including abuse, neglect, multiple home placements and caregiver substance use or mental illness (Astley, 2010; Astley et al., 2009; Price et al., 2017; Coggins et al., 2007). Understanding that co-occurring environmental risks and traumatic experiences can also affect child outcomes and family functioning in this population is also important to inform early intervention practices to help families adapt and thrive in the face of adversity.

1.3 Assessment of Infants/Toddlers with PAE

Developmental competencies in the context of early intervention are conventionally organized into five domains: cognitive, language, motor, social-emotional and adaptive behavior (Part C of IDEA, 2004). These domains of brain function are also considered when assessing the teratogenic impact of PAE when rendering an FASD diagnosis (Astley, 2013; Astley et al., 2017). These core domains are often assessed in infants and toddlers using a global test of neurodevelopment such as the Bayley Scales of Infant and Toddler Development (Bayley, 2006; Subramoney et al., 2018), although a range of standardized measures may be used. Self-regulation, an especially vulnerable domain in young children with PAE (Garrison et al., 2019), provides an important foundation for development, learning and adaptation (Carlson et al., 2013). Self-regulatory skills in early childhood can be evaluated with measures of sensory processing and emotional and behavior functioning such as the Infant Toddler Sensory Profile (ITSP; Dunn, 2002) and the Child Behavior Checklist for Ages 1½−5 years (CBCL; Achenbach and Rescorla 2000). Figure 1 presents an illustration of these core domains and common assessment tools used to evaluate infants/toddlers with PAE. These core domains and the data collected for the purpose of a FASD diagnostic evaluation provide the context for the two studies that were conducted for this dissertation.

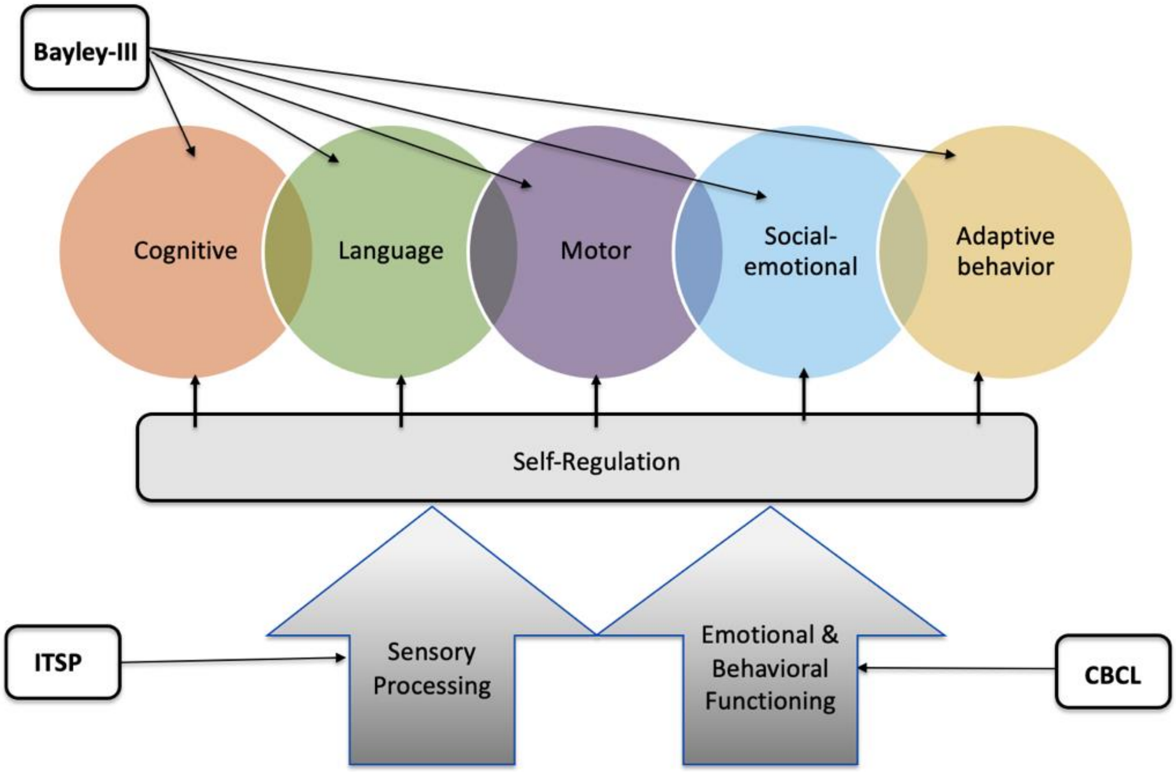


Figure 1.1 Core domains assessed during early childhood and common assessment tools used to evaluate infants/toddlers with PAE at the FASDPN (Reid & Petrenko, 2018; Astley, 2010). Abbreviations: Bayley-III Bayley Scales of Infant Development – Third Edition; ITSP Infant Toddler Sensory Profile; CBCL Child Behavior Checklist 1.5-5 years.

1.4 Summary of Studies

The purpose of this two-article dissertation is to describe the early developmental outcomes of infants/toddlers with PAE based on information from two sources, standardized assessment data and caregiver perspectives. Although there is extensive research on school-age children and youth, there is a limited body of research describing the developmental delays, sensory processing differences and challenging behaviors among infants/toddlers with PAE. In addition, little research has focused on the positive attributes and strengths of young children with PAE, which can buffer against the developmental vulnerability and adverse environmental influences associated with PAE (Shonkoff, 2017).

The first article in the dissertation, “Developmental, Sensory and Behavioral Outcomes Among Infants and Toddlers with Prenatal Alcohol Exposure” was a retrospective analysis of diagnostic clinical data from the University of Washington Fetal Alcohol Syndrome Diagnostic and Prevention Network (FASDPN). The purpose of this study was to describe the prevalence and patterns of neurodevelopment, sensory processing, and emotional and behavioral functioning in a clinical sample of infants/toddlers with PAE. Descriptive statistics were used to document the proportion of infants/toddlers classified with typical, at-risk, or delayed development on three measures used in the FASD diagnostic evaluation. Chi-square tests, *t*-tests and a one-way analysis of variance (ANOVA) were used to examine whether outcomes were correlated with PAE and/or other postnatal risk factors. This study was conducted to better understand the neurodevelopmental, sensory processing, and behavioral presentation in infants/toddlers with PAE to assist early childhood practitioners and other relevant allied service providers in identifying early delays or problems that may arise from PAE.

The second article in the dissertation, “Concerns and Strengths: Caregiver Perceptions of Their Infant/Toddler with Prenatal Alcohol Exposure”, is a qualitative study. The purpose of this study was to describe caregiver-reported concerns and strengths in the same clinical sample of infants/toddlers with PAE used in the first article. Using a directed approach to content analysis, caregivers’ written responses to open-ended questions on two parent-report questionnaires were coded and analyzed. The findings, which include descriptions of caregivers' most frequently reported concerns and strengths, demonstrate the value of eliciting and understanding caregiver perspectives as an aid to identifying the needs of infants/toddlers with PAE within the context of their families, and for informing a strengths-based approach to assessment and intervention.

1.5 Flow of Dissertation

This is a two-article dissertation that incorporates the linked-papers format. Following the brief introduction and overview in chapter one, chapters 2 and 3 are comprised of two complete articles in various stages of preparation and submission to peer-reviewed journals. Each article contains an abstract, introduction, methods, results, discussion, conclusion, and reference list. Chapter 4 summarizes overall findings and contributions of this dissertation research.

Although there is extensive research on school-age children and youth, there is a limited body of research describing the developmental delays, sensory processing differences and challenging behaviors among infants/toddlers with PAE. In addition, little research has focused on the positive attributes and strengths of young children with PAE, which can buffer against the developmental vulnerability and adverse environmental influences associated with PAE (Shonkoff, 2017).

Chapter 2. Developmental, Sensory and Behavioral Outcomes Among Infants and Toddlers with Prenatal Alcohol Exposure

2.1 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 examination of developmental, sensory processing and behavioral outcomes in infants/toddlers with PAE is needed to inform early identification and intervention practices.

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, aged 2-42 months, who received an FASD diagnostic evaluation at the University of Washington Fetal Alcohol Syndrome Diagnostic and Prevention Network clinic were analyzed. Measures included the Bayley Scales of Infant and Toddler Development, 3rd edition (Bayley-III) (n = 125), the Infant/Toddler Sensory Profile (ITSP) (n = 93), and the Child Behavior Checklist/1½ -5 (CBCL) (n = 67).

Results: The majority of infants/toddlers in this sample (74% to 87%) were classified with at-risk (≤ -1.0 SD) or delayed (≤ -1.5 SD) development in one or more of the five domains on the Bayley-III, with the highest prevalence of delayed development in the Adaptive Behavior (33%) and Language (31%) domains. All 93 infants/toddlers with a complete ITSP obtained definite difference scores in at least one quadrant/section, with the highest prevalence of definite differences in Low Registration (48%) and Auditory Processing (37%). Of the 67 infant/toddlers administered the CBCL, 43% scored in the clinical range on the Internalizing, Externalizing and/or Total Problem scales, with the highest prevalence of scores in the clinical range in

Attention Problems (29%) and Pervasive Developmental Problems (28%). These adverse developmental outcomes were significantly correlated with PAE and/or postnatal risk factors. Conclusions: Developmental concerns that varied by domain and severity were prevalent for infants/toddlers in this clinical sample. Early screening, ongoing monitoring and comprehensive assessment using standardized measures for neurodevelopment, sensory processing and behavior is needed to facilitate the earlier identification of infants/toddlers at risk for developmental delay due to their PAE and inform clinical intervention.

2.2 Introduction

A wide range of adverse neurodevelopmental, sensory, and behavioral outcomes have been documented among children with prenatal alcohol exposure (PAE) (Astley, 2010, 2019a; Astley et al., 2009; Subramoney et al., 2018; Carr et al., 2010). 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 heterogenous 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; Subramoney et al., 2018; Testa et al., 2003).

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 (AAP, 1993; 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 (for a review of literature see Garrison et al., 2019 and Subramoney et al., 2018). Many of these earlier studies used the Bayley-II, 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., Pyschomotor 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 describes outcomes across distinct scales. Other global measures of infant/toddler development, including newer versions of the Bayley (Bayley, 2006; 2019), 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 (IDEA, 2004) 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, 210; Astley et al., 2010; Reid & Petrenko, 2018) and as such, have been recognized as a core symptom in the proposed 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. 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 and emotional/behavioral problems in the early childhood period (Astley, 2013; Astley et al., 2010; Coles et al., 2015; Fjeldsted & Xue, 2019; Molteno et al., 2014; Subramoney et al., 2018). Findings from a recent comparative analysis demonstrate the usefulness of caregiver-reported assessments for identifying behavioral deficits in infants with PAE, especially for those with light/moderate PAE (Bakhireva et al., 2018. However, for children older than three, Astley & Clarren (2000) and Lange & colleagues (2017) suggest that both caregiver-reported observations/ratings and performance-based measures of neurodevelopment be included to establish a comprehensive profile of FASD. 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, 2103).

This study aimed to comprehensively document 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 (Astley et al., 2020). The following research questions were asked:

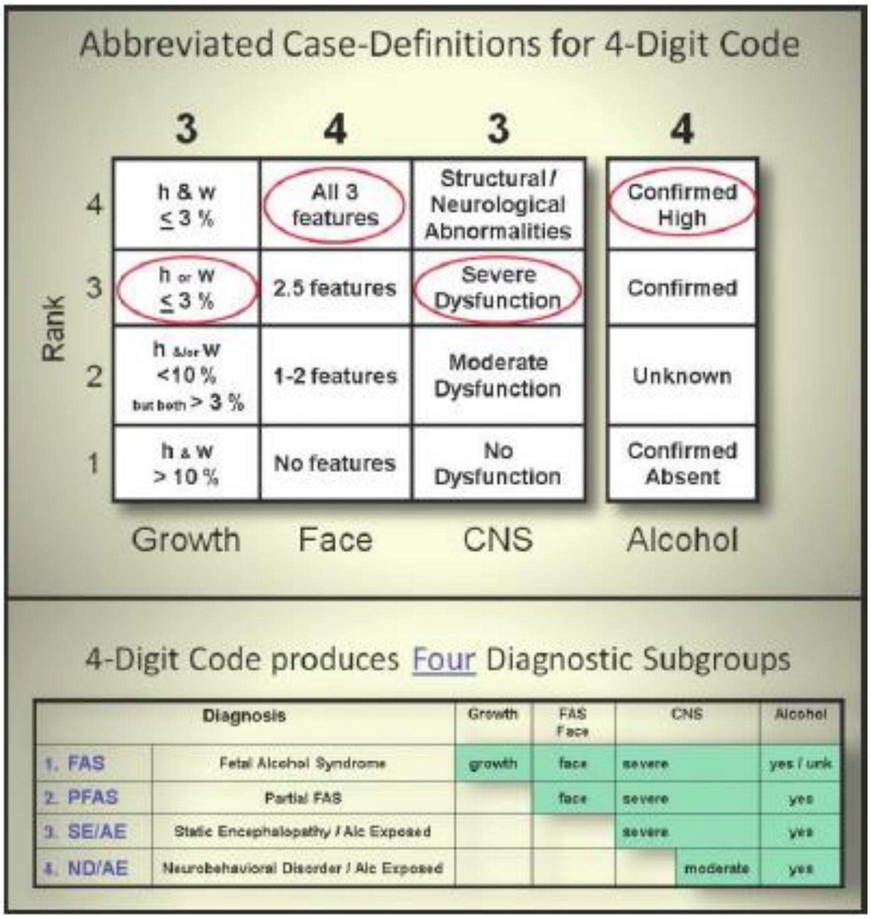
1. What is the prevalence of neurodevelopmental delay in five core developmental domains (i.e., cognitive, language, motor, social-emotional and adaptive behavior)?
2. What is the prevalence of atypical sensory processing behaviors among infants/toddlers with PAE?
3. What is the prevalence of emotional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

2.3 Methods

2.3.1 Research Design

This study is a retrospective 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. 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 2,000 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, Astley, 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) damage–dysfunction, 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 FAS feature and 4 reflecting strong "classic" presence of the FASD feature. Each Likert rank is specifically case defined (Figure 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 4-Digit Code classifies these individuals as follows: "Sentinel Physical Findings/Alcohol-Exposed" exposed (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" (diagnostic category J). See Figure 2.1.



Abbreviated Case-Definitions for 4-Digit Code

Rank	3	4	3	4
4	h & w ≤ 3 %	All 3 features	Structural / Neurological Abnormalities	Confirmed High
3	h or w ≤ 3 %	2.5 features	Severe Dysfunction	Confirmed
2	h &/or w <10 % but both > 3 %	1-2 features	Moderate Dysfunction	Unknown
1	h & w > 10 %	No features	No Dysfunction	Confirmed Absent
	Growth	Face	CNS	Alcohol

4-Digit Code produces Four Diagnostic Subgroups

Diagnosis		Growth	FAS Face	CNS		Alcohol
1. FAS	Fetal Alcohol Syndrome	growth	face	severe		yes / unk
2. PFAS	Partial FAS		face	severe		yes
3. SE/AE	Static Encephalopathy / Alc Exposed			severe		yes
4. ND/AE	Neurobehavioral Disorder / Alc Exposed				moderate	yes

Figure 2.1 Abbreviated case-definitions of the FASD 4-Digit Code (Astley, 2004; Astley, 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

2.3.2 Participants

Data from children who met the following inclusion criteria were used in this study: A) age 1 month to 3.5 years at the time of their FASD diagnostic evaluation; B) received one of the following diagnostic classifications reflecting the full continuum of outcomes observed among individuals with PAE using the FASD 4 Digit Diagnostic code: Diagnostic Categories A, B, C, E, F, G, H, I and J (as defined above); C) had complete data on a minimum of two domains from the Bayley-III; and D) were of any race, ethnicity, or gender.

2.3.3 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 clinical judgement. Thus, complete data for outcomes on the Bayley-III vary based on factors such as age, presenting developmental concern(s), and child factors such as attention. 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 (i.e., Bayley-III Social-Emotional and Adaptive Behavior domains, ITSP, and CBCL). Data and measures used for the study are described below.

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.

Assessment of Infant/Toddler Development Across Five Domains. Infants/toddlers were clinically assessed by the occupational therapist using the Bayley Scales of Infant and Toddler Development–Third Edition (Bayley-III; Bayley, 2006), 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 have 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 ≥ -0.9 *SD*), at-risk development (78-85, scores between -1.0 and -1.4 *SD*), and delayed development (≤ 77, scores ≤ -1.5 *SD*); subdomain scores: typical development (scaled scores ≥ 8, scores ≥ -0.9 *SD*), at-risk development (6-7, scores between -1.0 and -1.4 SD), and delayed development (≤ 5, scores ≤ -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 & Grieves, 2007).

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). Lower scores indicate a higher frequency of response. The ITSP categorizes the raw scores as typical performance (scores at or between ± 1.0 *SD*), probable differences (scores within the 1 to 2 *SD* range) and definite difference (scores outside ± 2.0 *SD*). The ITSP is reported to have excellent test-retest reliability (α = .86) for domain/section scores and adequate (α = .74) for quadrant scores (Eeles et al., 2013). Validity was established in several studies (Dunn, 2002; Dunn & Daniels, 2002; Eeles et al., 2013).

Assessment of Emotional and Behavioral Problems. The Child Behavior Checklist for ages 1½−5 years (CBCL; Achenbach and Rescorla 2000) is a widely used instrument used to identify a range of behavioral and emotional problems in children 1½−5 years. 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 (α = 0.97) and good test–retest reliability (mean *r of .85* across all scales) (Rescorla, 2005).

Diagnostic Outcome Data.

4-Digit Diagnosis. 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 (Figure 2.1).

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

FAS Facial Phenotype. 'Face Rank': 1 = none; 2 = mild; 3 = moderate; 4 = severe. This variable represents the second digit in the 4-Digit FASD 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 (Astley, 2004).

CNS Likelihood of Structural Abnormality. 'CNS Rank': 1=unlikely; 2=possible; 3=probable;4=definite) This variable yields the third digit in the 4-Digit FASD 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, sensory, 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 moderate; 4 = confirmed exposure; level high). Alcohol exposure is the fourth digit in the 4-Digit FASD Diagnostic

Code, which is ranked according to the quantity, timing, frequency, and certainty of exposure during pregnancy. The ranking is determined by available records, maternal report or report from others who observed exposure.

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, 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 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 ranking is determined by available records and caregiver or other report on intake forms and/or clinical interview.

2.4 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 on measures of neurodevelopment, sensory processing, and emotional and behavioral problems. Relationships between Bayley-III domain and subdomain score categories (typical, at-risk, and delayed development) and selected child demographics (age, gender, and postnatal risk factors) were examined using chi-squared tests (χ^2), *t*-tests and a one-way analysis of variance (ANOVA). 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. The number of items in each quadrant/section ranged between 6 and 15 items. 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.

2.5 Results

2.5.1 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 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 2.1). 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%) presented with at least some level of other 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 was 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.

Table 2.1 Demographic and clinical profiles of infants/toddlers with Bayley-III, ITSP, and CBCL assessments

	Bayley-III		**ITSP**		**CBCL**	
Characteristic	*n*	(valid %)	*n*	(valid %)	*n*	(valid %)
Total *n*	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 (years)						
Birth to 0.99	22	(17.6)	9	(9.7)	0	0.0
1 - 1.99	45	(36.0)	42	(45.2)	18	(26.9)
2 - 2.99	44	(35.2)	39	(41.9)	36	(53.7)
3 - 3.5	14	(11.2)	3	(3.2)	13	(19.4)
Mean (SD)	1.9	(0.84)	1.9	(0.69)	2.4	(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						
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)
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).

2.5.2 Neurodevelopmental Performance on the Bayley-III

Bayley-III outcomes are presented in Figures 2.2- 2.4 (and Table A1 in Appendix). 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%). The prevalence of delay was significantly lower in the cognitive domain relative to the Adaptive Behavior and Language domains ($X^2 = 14.5$, $p = .000$; $X^2 = 11.0$, $p = .001$ respectively). Within the Language and Adaptive Behavior domains, the most prevalent delays (≤ 1.5 *SDs*) 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 (≤ -1.5 *SDs*). 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 (≤ -1.5 *SDs*). Since not all domains were assessed, 74% serves as a minimal estimate.

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 (≤ -1.5 *SDs*) 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).

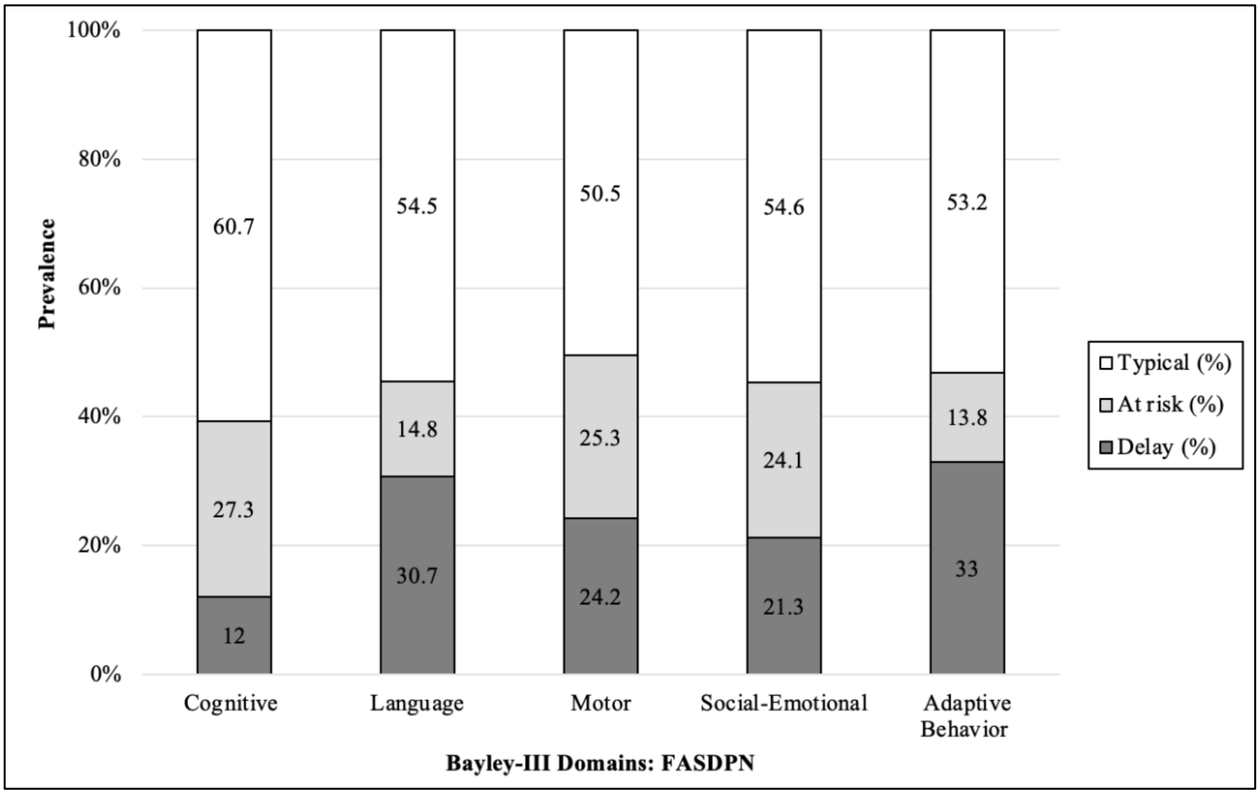


Figure 2.2. Proportion of 125 infants/toddlers with typical, at-risk, and delayed development across the five Bayley-III domains.

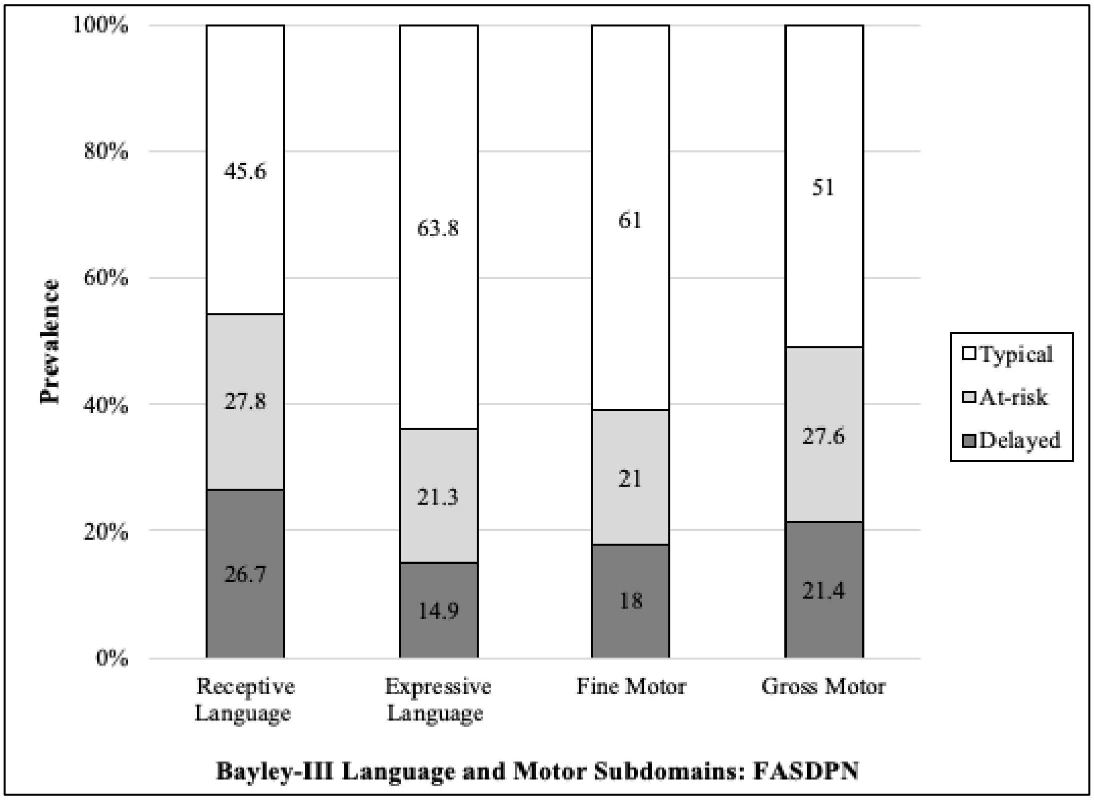


Figure 2.3. Proportion of infants/toddlers with typical, at-risk, or delayed development in the Bayley-III Language and Motor subdomains. Receptive language: n = 90, expressive language: n = 94, fine motor: n = 100, and gross motor: n = 98.

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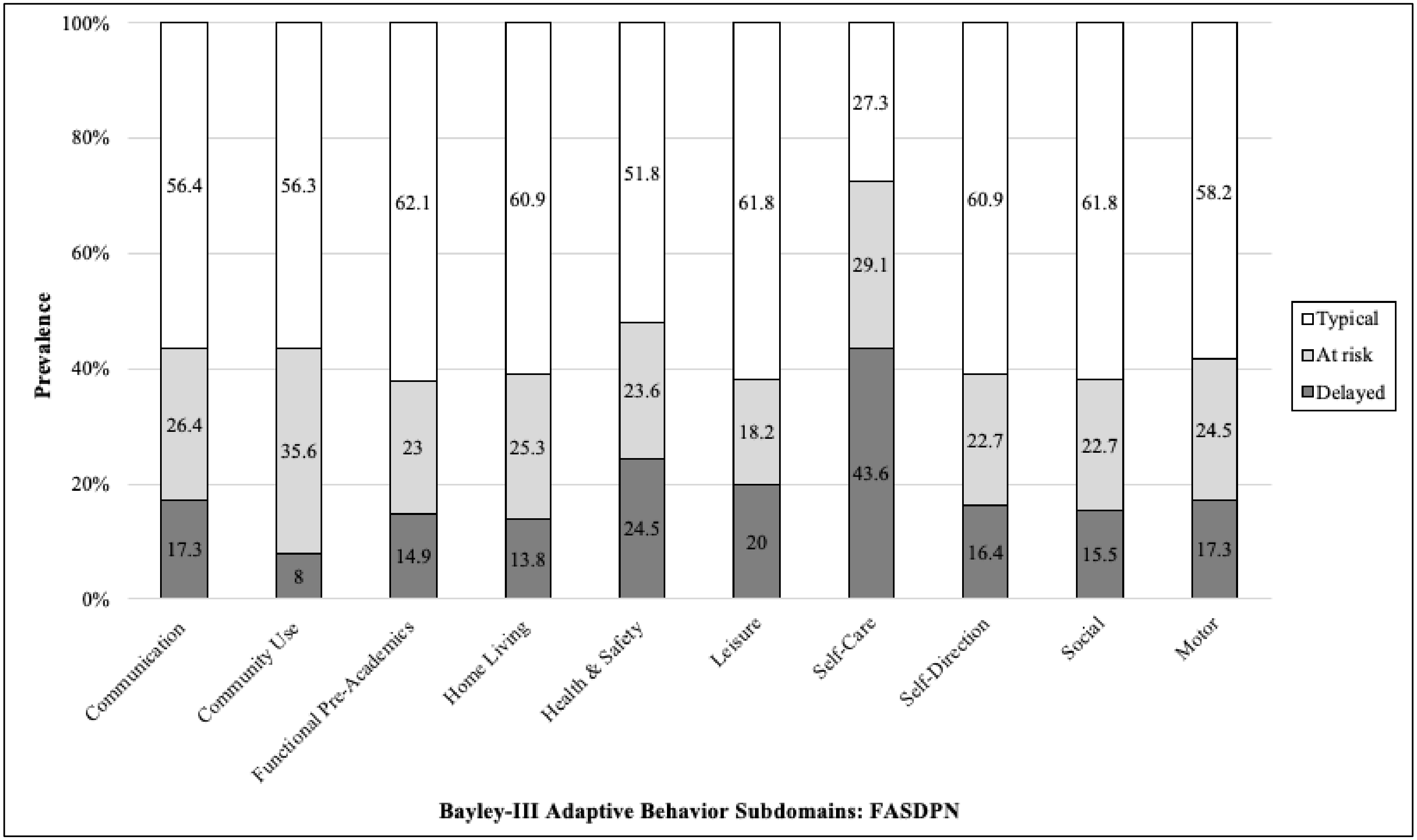

Figure 2.4. Proportion of infants/toddlers with typical, at-risk, or delayed development in the Bayley-III Adaptive Behavior subdomains. Communication: n = 110, community use: n = 87, functional pre-academics: n = 87, home living: n = 87, health & safety: n = 110, leisure: n = 110, self-care: n = 110, self-direction: n = 110, social: n = 110, and motor: n =110.

2.5.3 Sensory Processing Performance on the ITSP

Ninety-three infants/toddlers ranging in age from 7-36 months had a completed ITSP assessment. Compared to the larger group of 125 children with Bayley-III assessments, this subgroup had no infants/toddlers diagnosed with FAS (Table A2 in Appendix). The distribution of Bayley-III domain scores by classification category (typical, at-risk, delayed) in this subgroup was very similar to the full group of 125 infants/toddlers.

Within each quadrant and section of the ITSP, roughly half of the infant/toddlers presented with outcomes in the probable or definite difference range (Figure 2.5 and Table A2). 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.

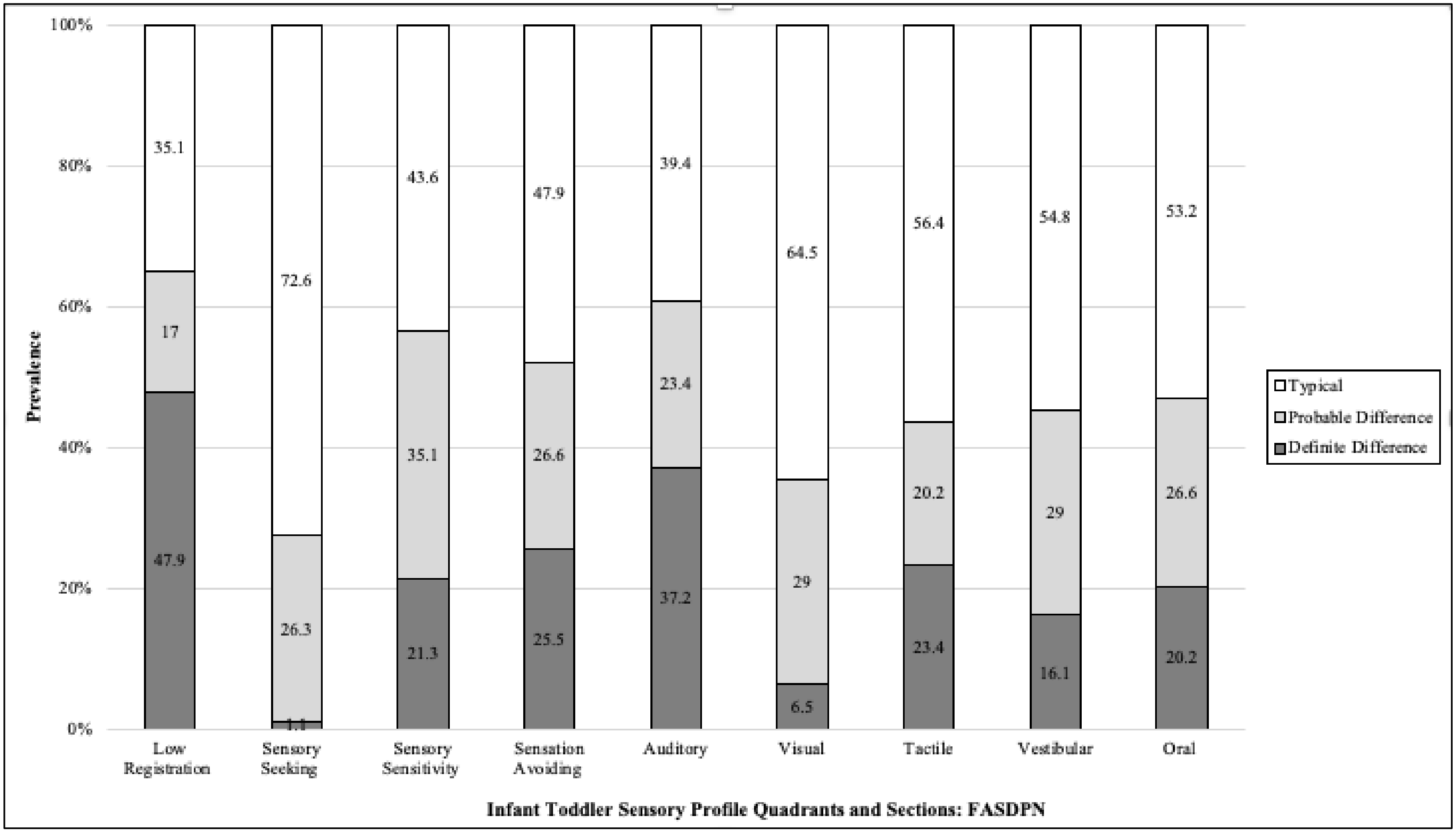

Figure 2.5. Proportion of 93 infants/toddlers with typical performance, probable difference, or definite difference across the four ITSP quadrants (low registration, sensory seeking, sensory sensitivity, sensory avoiding) and five section (auditory, visual, tactile, vestibular, and oral sensory processing).

2.5.4 Emotional and Behavioral Functioning on the CBCL

Sixty-seven infants/toddlers ranging in age from 1.5 - 3.5 years had a completed CBCL assessment. Compared to the larger group of 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 (Figure 2.6 and Table A3 in Appendix). Attention Problems had the highest prevalence of elevated scores on the Syndrome scales (36%) and Pervasive Developmental Problems had the highest prevalence of elevated scores on the DSM-Oriented Scales (Figure 2.7). Of the 67 infant/toddlers with CBCL data, 29 (43%) scored in the clinical range on the Internalizing, Externalizing and/or Total Problem scales.

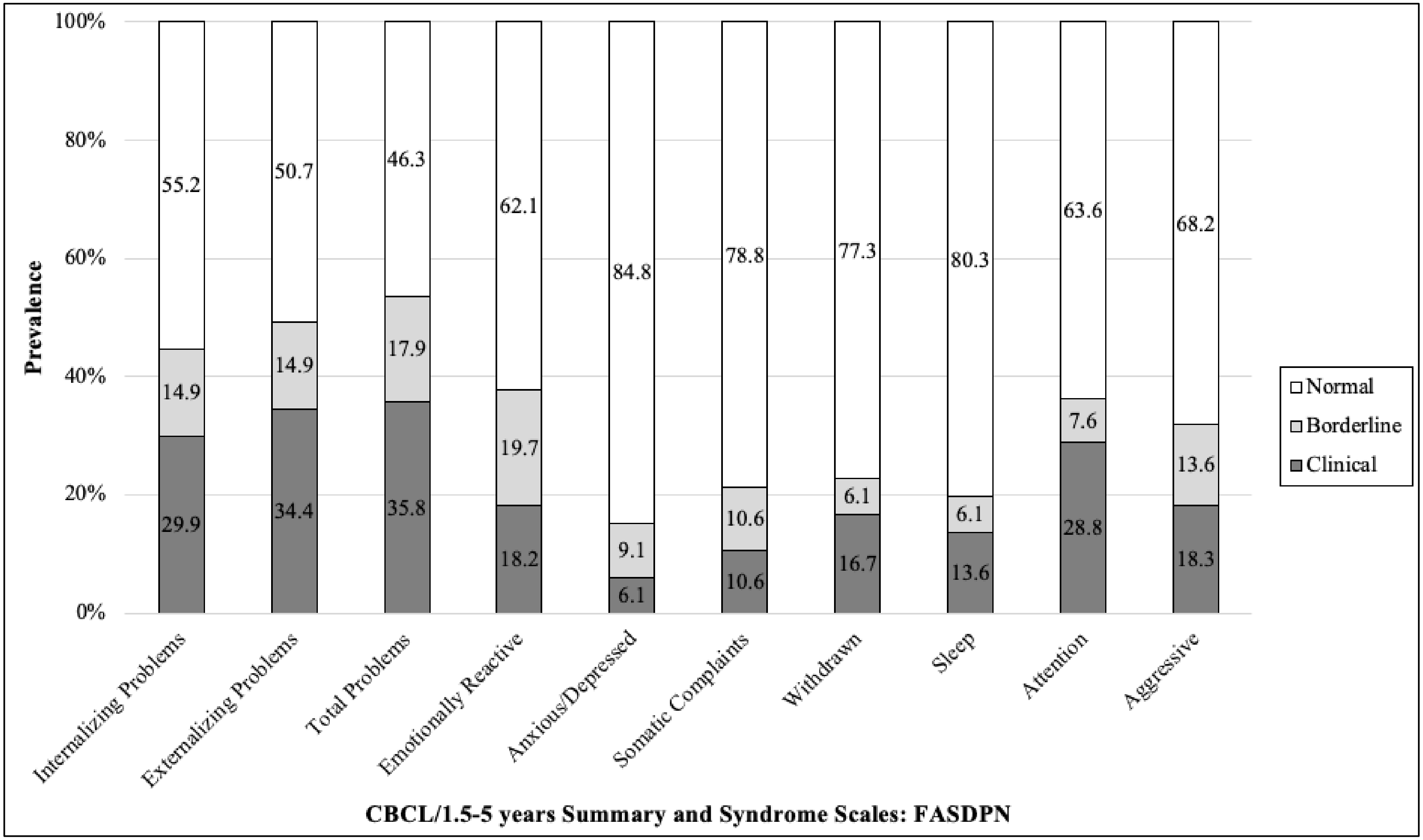

Figure 2.6. Proportion of 67 infants/toddlers with scores in the normal, borderline clinical and clinical range across three Summary Scales (internalizing, externalizing and total problems)and seven Syndrome Scales (emotionally reactive, anxious/depressed, somatic complaints, withdrawn, sleep, attention, and aggressive problems) of the CBCL

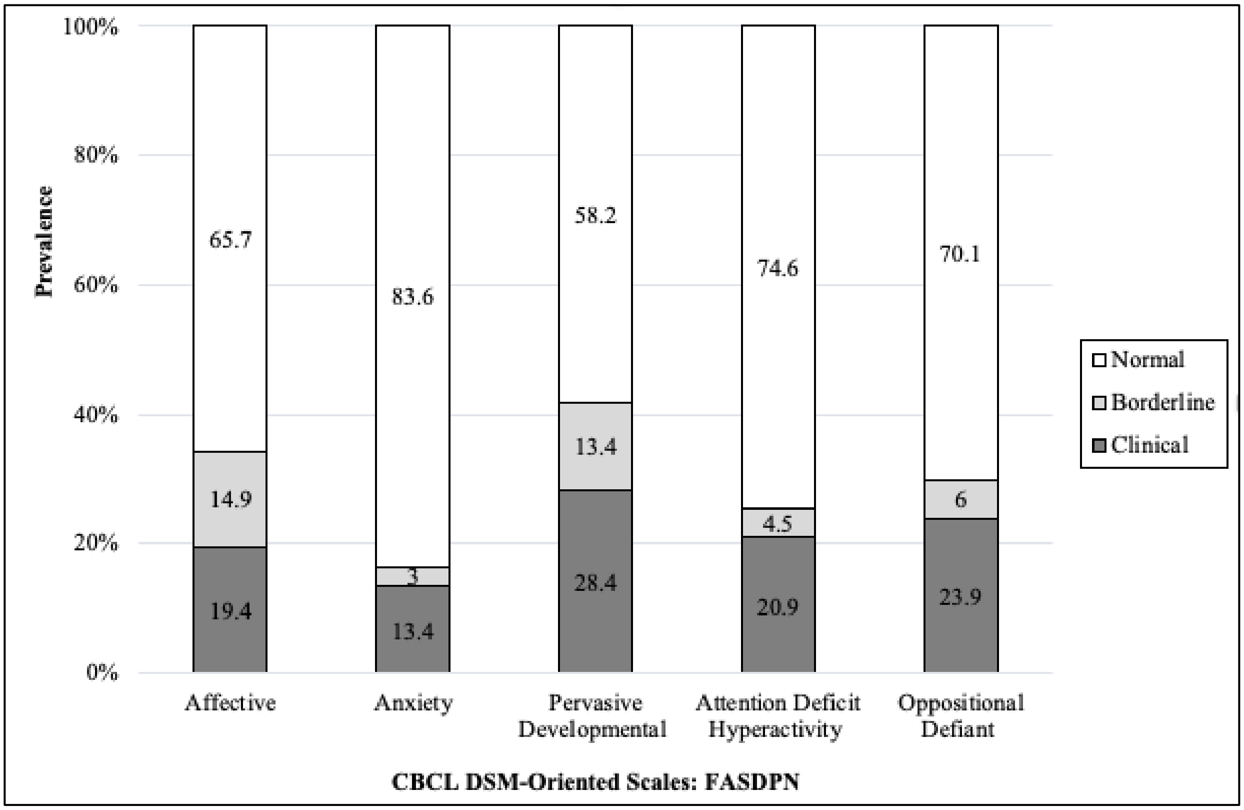

Figure 2.7. Proportion of 67 infants/toddlers with scores in the normal, borderline, and clinical range across five DSM-Oriented Scales of the CBCL

2.5.5 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 (≤ -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.

2.5.6 Correlations Between PAE, Other Postnatal Risk Factors and Three Measures

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 (Figure 2.8A). 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 1,400 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 (Figure 2.8B), 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 Figure 2.8C for the Motor domain). Domain standard scores decreased roughly 10-20 points from Face Rank 1 to Face Rank 4. Subdomain scaled scores decreased roughly 2-4 points from Face Rank 1 to Face Rank 4.

Interestingly, the Adaptive Behavior domain appeared more highly correlated with other postnatal risks than PAE (Figure 2.9). There was a near significant decrease in mean Adaptive Behavior scores with increasing levels of postnatal risk (ANOVA: F = 3.4, *p* = .067). Lastly, 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).

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).

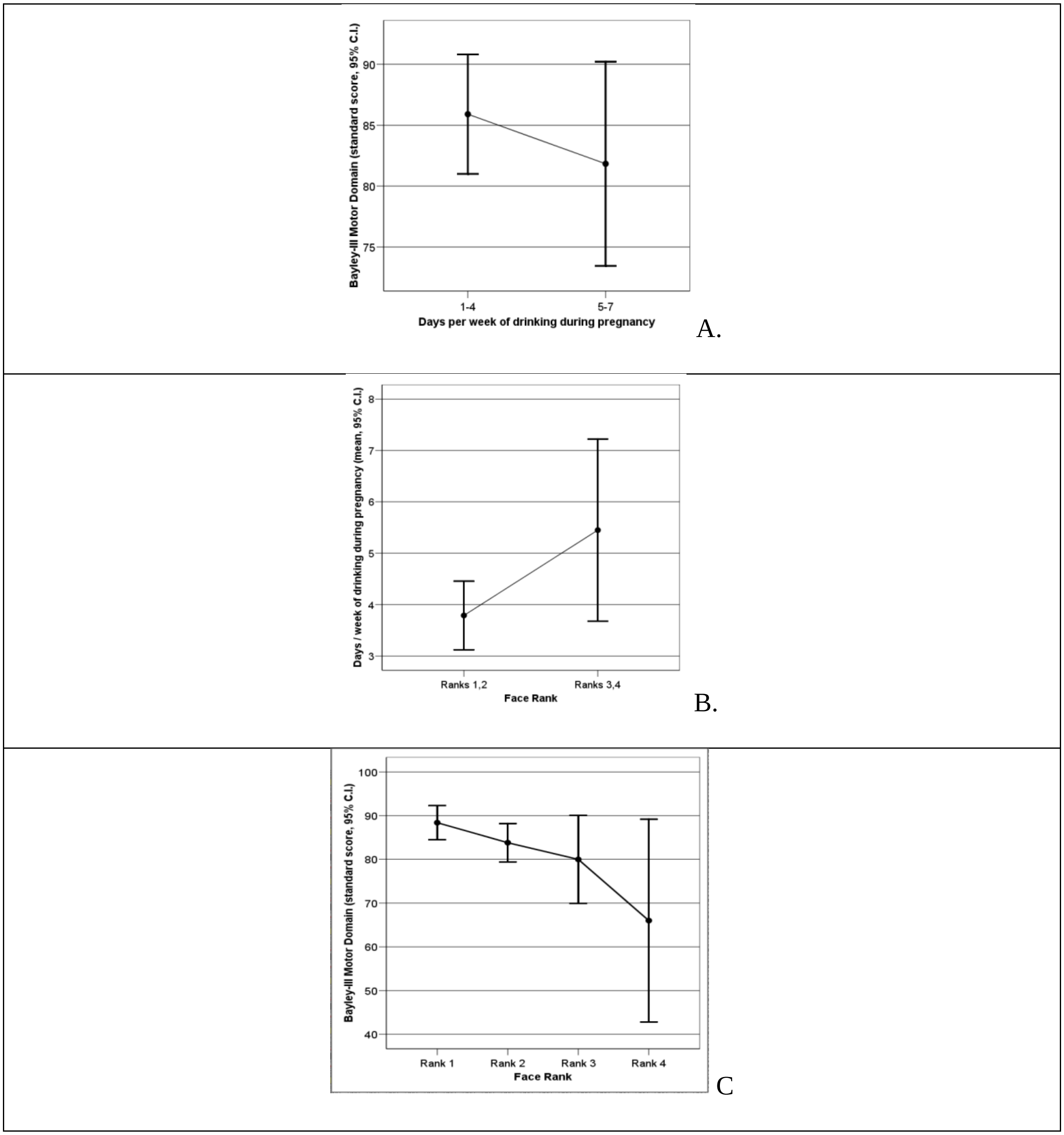

Figure 2.8. Mean scores across all Bayley-III domains and subdomains (with the exception of Adaptive Behavior) decreased with increasing PAE. Using the Motor domain as an example: A. The mean Bayley-III Motor score decreased with increasing number of days/week of drinking during pregnancy (t = 0.9, p = 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

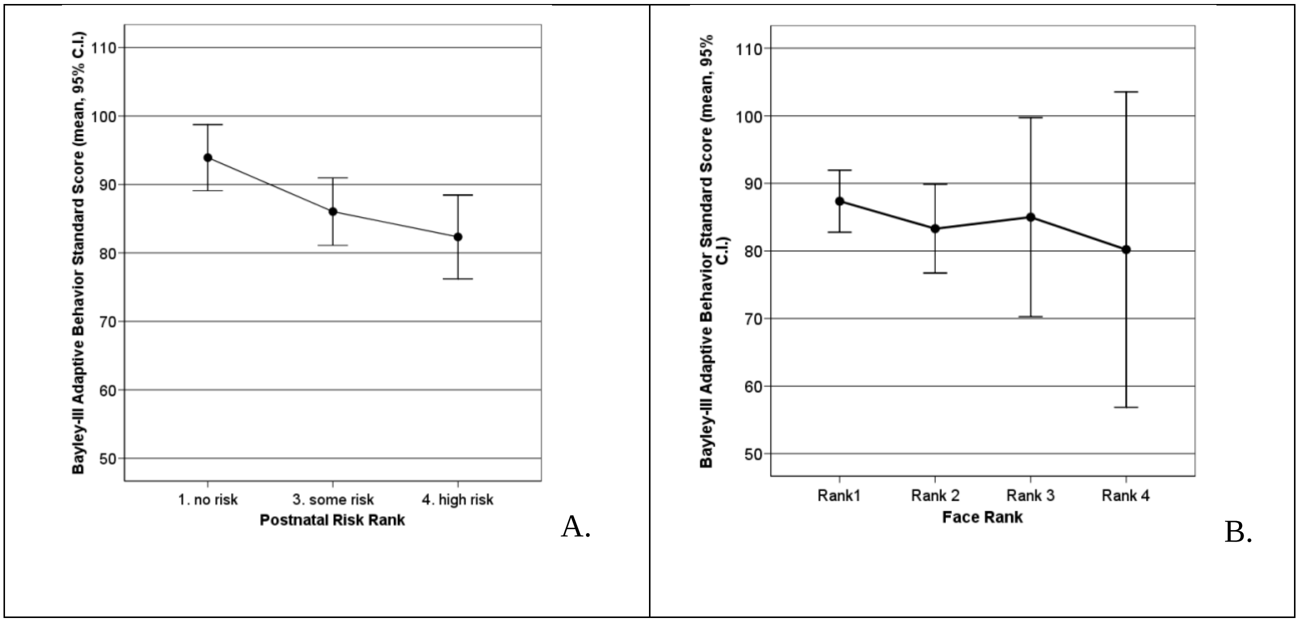

Figure 2.9. Impairment in Adaptive Behavior appeared to be more strongly associated with postnatal risk factors than PAE. A. The mean Bayley-III Adaptive Behavior standard score decreased near significantly with increasing severity of the postnatal risk rank (Linear Term: F = 1.7, p = 0.67). B. The mean 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).

2.6 Discussion

This study examined the developmental, sensory processing and behavioral outcomes in a clinical sample of 125 infants/toddlers with PAE using the Bayley-III (n = 125), ITSP (n =93) and CBCL (n =67) respectively. 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.

2.6.1 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 (infant to adult) evaluated in the FASDPN clinic (Astley, 2010) and among the 1,131 patients (7.0 years old to adult) presented on the FASDPN Tableau Dashboard (FASDPN, n.d.). While the pattern of developmental delay (from least prevalent to most prevalent) is comparable between this infant/toddler study sample and our older adolescent/adult FASD clinical population, the prevalence of impairment in each domain is roughly two-fold greater in the older adolescent/adult population (cognition 28%, motor 18%, language 48%, adaptive behavior 67%)

(FASDPN, n.d.). 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 neurodevelopmental 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 how well 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. Astley 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 positive 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 (Shonkoff et al., 2012, van der Kolk, 2003), and among individuals with PAE (Coggins et al., 2007; Astley

Hemingway et al., 2020; Price et al., 2017; Streissguth et al., 2004), our analyses revealed a significant positive correlation between other postnatal risks and adaptive behavior delays.

2.6.2 Sensory Processing Outcomes and Implications

Atypical sensory processing behaviors were observed in a large proportion of infants/toddlers. As postulated 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.

2.6.3 Emotional and Behavioral Functioning Outcomes and Implications

Atypical emotional and behavioral problems were also 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 (Franklin et al., 2008; Jirikowic et al., 2008; Astley, 2010; Astley et al., 2009; Astley 2019b). Notably, the prevalence of Total Problem scores in the clinical range for the older children (86%; Franklin et al., 2008) were much higher in comparison to the current sample of infants/toddlers with PAE (36%). 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.

2.6.4 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 sample size 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 support 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. 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 and CBCL 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 (Bakhireva et al., 2018; Astley, 2010), 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.

2.7 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.

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APPENDIX A

Table A1. Bayley-III Distributions of Domain and Subdomain Scores and Classification Categories for 125 Infants/Toddlers with Prenatal Alcohol Exposure

				Categories		
Bayley-III Domains & Subdomains	Total *n*	Mean Score (SD)	Min-Max	Typical *n* (%)	At-risk *n* (%)	Delay *n* (%)
Cognitive	117	89.3 (11.5)	55-120	71 (60.7)	32 (27.3)	14 (12.0)
Language	88	85.9 (13.8)	58-118	48 (54.5)	13 (14.8)	27 (30.7)
Receptive communication	90	7.1 (2.8)	1-12	41 (45.6)	25 (27.8)	24 (26.7)
Expressive Communication	94	8.1 (2.6)	2-14	60 (63.8)	20 (21.3)	14 (14.9)
Motor	95	84.9 (14.6)	46-121	48 (50.5)	24 (25.3)	23 (24.2)
Fine motor	100	7.9 (2.8)	1-14	61 (61.0)	21 (21.0)	18 (18.0)
Gross motor	98	7.2 (2.6)	1-13	50 (51.0)	27 (27.6)	21 (21.4)
Social-Emotional	108	92.1 (17.6)	55-140	59 (54.6)	26 (24.1)	23 (21.3)
Adaptive Behavior	110	85.6 (18.2)	44-133	58 (53.2)	15 (13.8)	36 (33.0)
Communication	110	8.3 (3.2)	1-16	62 (56.4)	29 (26.4)	19 (17.3)
Community Use	87	8.2 (2.3)	3-14	49 (56.3)	31 (35.6)	7 (8.0)
Functional Pre-Academics	87	8.5 (2.9)	3-16	54 (62.1)	20 (23.0)	13 (14.9)
Home Living	87	8.4 (2.7)	3-15	53 (60.9)	22 (25.3)	12 (13.8)
Health and Safety	110	8.0 (3.0)	3-16	57 (51.8)	26 (23.6)	27 (24.5)
Leisure	110	8.6 (3.1)	2-16	68 (61.8)	20 (18.2)	22 (20.0)
Self-Care	110	5.9 (2.5)	1-12	30 (27.3)	32 (29.1)	48 (43.6)
Self-Direction	110	8.5 (3.2)	1-15	67 (60.9)	25 (22.7)	18 (16.4)
Social	110	8.5 (3.0)	2-17	68 (61.8)	25 (22.7)	17 (15.5)
Motor	110	8.4 (2.8)	2-16	64 (57.7)	27 (24.3)	20 (18.0)

Note. Standardized scores for the cognitive, language, motor, and adaptive behavior domains have a normative mean of 100 and a standard deviation (SD) of 15. Standardized scores are categorized as typical development (≥86, ≥ -0.9 *SD*), at-risk development (78-85, scores between -1.0 and -1.4 *SD*), and delayed development (≤ 77, scores ≤ -1.5 *SD*). Scaled scores for the subdomains have a normative mean of 10 and a SD of 3. Scaled scores are categorized as typical development (≥8, ≥ -0.9 *SD*), at-risk development (6-7, scores between -1.0 and -1.4 SD), and delayed development (≤ 5, scores ≤ -1.5 *SD*). To be eligible for early intervention services in Washington state, a child must demonstrate a -1.5 *SD* delay in one or more of the above domains.

APPENDIX B

Table A2. ITSP Distributions of Quadrant and Section Scores by Classification Categories for 93 Infants/Toddlers with PAE

	Classifications		
ITSP Quadrants	Typical *n* (valid %)	Probable Difference *n* (valid %)	Definite Difference *n* (valid %)
Low Registration	33 (35.1)	16 (17.0)	45 (47.9)
Sensation Seeking	69 (72.6)	25 (26.3)	1 (1.1)
Sensation Sensitivity	41 (43.6)	33 (35.1)	20 (21.3)
Sensation Avoiding	45 (47.9)	25 (26.6)	24 (25.5)
ITSP Sections			
Auditory Processing	37 (39.4)	22 (23.4)	35 (37.2)
Visual Processing	60 (64.5)	27 (29.0)	6 (6.5)
Tactile Processing	53 (56.4)	19 (20.2)	22 (23.4)
Vestibular Processing	51 (54.8)	27 (29.0)	15 (16.1)
Oral Sensory Processing	50 (53.2)	25 (26.6)	19 (20.2)

Notes. Quadrant and section scores fall along a continuum with scores categorized as typical performance (scores at or between ± 1.0 *SD*), probable differences (scores within the 1 to 2 *SD* range) and definite difference (scores outside ± 2.0 *SD*). Lower scores indicate a higher frequency of problems in a given area of sensory processing.

APPENDIX C

Table A3. CBCL Distributions of Summary, Syndrome and DSM-Oriented Scores and Category Classifications for 67 Infants/Toddlers with PAE

				Category Classifications		
				Normal range	Borderline range	Clinical range
CBCL Summary Scales	Mean *T*-Score	(SD)	Min-Max	*n* (valid %)	*n* (valid %)	*n* (valid %)
Internalizing Problems	56.8	(11.7)	29-80	37 (55.2)	10 (14.9)	20 (29.9)
Externalizing Problems	60.2	(13.3)	28-89	34 (50.7)	10 (14.9)	23 (34.3)
Total Problems	60.0	(12.9)	29-87	31 (46.3)	12 (17.9)	24 (35.8)
CBCL Syndrome Scales						
Emotionally Reactive	61.1	(9.5)	50-93	41 (62.1)	13 (19.7)	12 (18.2)
Anxious/Depressed	55.9	(7.9)	50-87	56 (84.8)	6 (9.1)	4 (6.1)
Somatic Complaints	56.2	(7.6)	50-74	52 (78.8)	7 (10.6)	7 (10.6)
Withdrawn	58.1	(8.6)	50-85	51 (77.3)	4 (6.1)	11 (16.7)
Sleep	58.1	(10.6)	50-94	53 (80.3)	4 (6.1)	9 (13.6)
Attention	60.7	(9.5)	50-80	42 (63.6)	5 (7.6)	19 (28.8)
Aggressive	61.2	(11.8)	50-95	45 (68.2)	9 (13.6)	12 (18.2)
CBCL DSM-Oriented Scales						
Affective	59.7	(9.2)	50-84	44 (65.7)	10 (14.9)	13 (19.4)
Anxiety	56.6	(9.0)	50-86	56 (83.6)	2 (3.0)	9 (13.4)
Pervasive Developmental	62.3	(10.6)	50-86	39 (58.2)	9 (13.4)	19 (28.4)
Attention Deficit Hyperactivity	60.3	(8.7)	50-76	50 (74.6)	3 (4.5)	14 (20.9)
Oppositional Defiant	60.2	(9.7)	50-80	47 (70.1)	4 (6.0)	16 (23.9)

Notes. Summary scale *t*-scores are categorized as normal range (< 60), borderline clinical range (60 to 63), and clinical range (≥ 64). Syndrome and DSM-oriented *t*-scores are categorized as normal range (< 65), borderline clinical range (65 to 69), and clinical range (≥ 70).

APPENDIX D

Diagnostic Category	**4-Digit Codes within each FASD Diagnostic Category**
FAS/AE (A) (n = 5)	444(1)3, 444(2)4, 444(2)4, 444(3)4, 444(3)4
PFAS (C) (n = 5)	234(2)3, 334(2)3, 334(2)3, 434(2)3, 434(2)4
SE/AE (EF) (n = 13)	1133, 114(3)3, 114(1)4, 114(2)4, 124(2)4, 214(2)3, 2234, 224(2)4, 324(2)4, 324(2)4, 324(2)4, 324(2)4, 424(2)4
ND/AE (GH) (n = 75)	2124 (4x), 1124 (22x), 1123 (16x), 2424, 2223 (2x), 3224 (5x), 1223 (4x), 2323, 4424, 2123 (2x), 2224 (3x), 3123 (3x), 3324, 1224, 4123, 3223 (3x)
Sentinel Physical Findings/AE (I) (n=5)	4114, 3214, 3113, 3114, 1314
No abnormal findings/AE (J) (n = 22)	1114 (6x), 1113 (5x), 1214 (2x), 1213 (5x), 2114 (2x), 2214 (2x)

Chapter 3. Concerns And Strengths: Caregiver Perceptions of Their Infant/Toddler with Prenatal Alcohol Exposure

3.1 Abstract

Background: Caregiver-reported assessments provide opportunities for caregivers to share concerns and identify the strengths of their infant/toddler with prenatal alcohol exposure (PAE). These insights may reveal under-recognized concerns and inform a strengths-based approach to early intervention.

Aims: The purpose of this study was to describe the type and frequency of caregiver-reported concerns and strengths in a sample of infants/toddlers (ages 7-42 months) at the time of their fetal alcohol spectrum disorder (FASD) diagnostic evaluation.

Methods: Caregivers' concerns and strengths were identified in the context of two parent-report questionnaires, the Infant Toddler Sensory Profile (ITSP) and Child Behavior Checklist/1½ -5 (CBCL). Using content analysis, caregivers' open-ended responses were organized, coded, and analyzed. The frequencies of all coded concerns and strengths were counted. Data were compared across two age groups and caregiver status.

Results: Caregivers (*n*=117) identified numerous concerns and strengths across multiple categories. The most frequently reported concerns were related to aggressive behavior, language/communication, and sensory processing. The most frequently reported strengths were related to happiness, sociability, and love. The type of concerns and strengths reported were relatively consistent across age and caregiver status.

Conclusions: Findings reinforce the value of caregivers' perspectives and offer a reminder to practitioners that infants/toddlers with PAE and their caregivers have many strengths that can be harnessed, in addition to a range of challenges that must be addressed.

3.2 Introduction

Prenatal alcohol exposure (PAE) can disrupt the neurodevelopmental and behavioral trajectory of infants/toddlers with lasting impacts on learning, mental health, and overall well-being (Garrison et al., 2019; Popova et al., 2016). Fetal alcohol spectrum disorder (FASD), a term used to describe the full range of physical, cognitive, and behavioral impairments caused by PAE, is estimated to occur in at least 1% children and youth in the general population (Lange et al., 2017; Roozen et al., 2016). Infants/toddlers with PAE are a heterogeneous group of children who may experience a wide range of delays in development, sensory processing and/or emotional and behavioral functioning (Pruner, 2021a). Challenges in any one of these developmental domains can limit participation in everyday routines and activities, and negatively influence the quality of parent-child interactions and early relationships (Guralnick & Bruder, 2019; Pluess & Belsky, 2010). Conversely, infants/toddlers with PAE also possess individual strengths and positive attributes (Pruner et al., 2020) that can serve as protective factors and support whole child development.

Early interventions that target risk factors and build on individual strengths can alter the course of development in a positive direction. Findings from decades of developmental and intervention science have demonstrated the substantial benefits of early intervention on child development and family well-being (Guralnick, 2011). The first three years of life have been recognized as an incredibly important time for child development given the brain’s capacity for change and sensitivity to environmental influences (Center on the Developing Child at Harvard University, 2016). With an emphasis on promoting healthy parent-child interactions and strengthening family adaptation, early intervention’s family-centered approach is well-suited to respond to the diverse needs of infants/toddlers with PAE (Guralnick & Bruder, 2019; Olson et al., 2007).

Although early identification and diagnosis may be the best way to positively influence outcomes in young children with PAE (Streissguth et al., 2004), PAE appears to be under-recognized by early childhood practitioners (Olson et al., 2007). The early identification of infants/toddlers with PAE is complicated in several ways. Challenges by multiple systems of care (i.e., health care, child welfare, early intervention, and infant mental health) to implement universal screenings or structured identification processes for maternal alcohol use history may be, in part, inhibiting earlier referrals for FASD diagnosis (Testa et al., 2003). In addition, not all infants/toddlers with PAE present with easy to recognize symptoms such as characteristic physical findings (e.g., growth problems, FAS facial features, structural brain abnormalities) or severe neurodevelopmental/ behavioral delays (Astley et al., 2016). Instead, many infants/toddlers may have more subtle developmental or behavioral indicators (Molteno et al., 2014; Kable & Coles, 2004) that are not as easily recognized by early childhood practitioners, thus translating into a missed opportunity for early identification and intervention.

Caregivers on the other hand, are often the first to raise concerns about their child's development or behavior and can therefore serve as critical step in identifying early delays or problems that may arise from PAE. Directing attention to caregivers'-reported assessments, which constitute a valuable component of early childhood assessment, is one way to understand what caregivers are concerned about. Standardized caregiver-report measures commonly used to assess infant sensory processing and behavior, provide a way for parents to examine and report child behaviors. They also permit caregivers to express their concerns through rating scales and responses to open-ended questions. Although clinicians tend to focus their attention on rating scale outcomes, Guinchat et al. (2012) endorse the use of open-ended questions to ask about parent concerns because the responses tend to be more spontaneous and personal. It is through open-ended questions that new or under-recognized concerns related to PAE may be uncovered, as caregivers provide responses in their own words and are not constrained by predetermined responses (Esses & Maio, 2002).

Primary caregivers, by nature of their role and relationship, have a unique vantage point that makes them acutely aware of the day-to-day challenges faced by their child. This increased awareness makes caregivers a vital resource to identify children whose development and behavior does not appear typical. Previous research with caregivers of children with PAE or FASD demonstrate caregivers' vigilance to variations in their child's development and behavior. One qualitative study with foster mothers of children (ages 2 - 16 years) reported a multitude of problems, including concerns related to child cognition, behavior management, and coping with the daily realities of life (Gardner, 2000). Likewise, a second study describing the lived experiences of eight birth mothers of a child/ren with FASD (8 – 30 years) reported cognitive concerns (i.e., problems with attention, comprehension, and memory) and problem behaviors (i.e., excessive crying or no crying, hyperactivity, aggressiveness), in addition to health issues and delayed developmental milestones (Salmon, 2008). A third study emphasized concerns faced by caregivers including FASD-related stigma, family stress and a lack of knowledge by professionals (Mukherjee et al., 2013). Finally, in a study by Pruner et al. (2020), caregivers were asked to reflect on challenges faced by their child with PAE during their first three years of life. Caregivers reported a diversity of concerns spanning across all domains of development and further reflected on how early interventions met (or did not meet) those needs. Collectively, these studies recognize caregivers' valuable observation skills and insights into their children's developmental needs. In accordance, the Academy of Pediatrics recommend that health professionals ask about and attend to caregiver concerns as a first step toward the developmental surveillance of infants and young children (Bright Futures, 2006; Lipkin & Macias, 2020). Furthermore, the FASDPN clinic conducts a structured caregiver interview as a standard of practice during a FASD diagnostic evaluation (Astley, 2004).

Recent literature has emphasized the need for a strengths-based approach to assessment and intervention with children with FASD (Petrenko et al., 2014; Skorka et al., 2020). Guiding caregivers to identify child strengths during a clinical encounter can extend benefits to both caregivers and practitioners (Steiner, 2011). A strengths-based approach can offer parents a sense of hope, alleviate child-related stress, and strengthen parenting capacity (Cosden et al., 2006; Grant et al., 2005; Olson et al., 2009). Twenty years of caregiver surveys of patients diagnosed with FASD at the FASDPN clinic confirm caregivers were highly satisfied with the strength-based approach to assessment and intervention used by the FASDPN (Astley, 2014). When early childhood practitioners appreciate the variety of child strengths identified by caregivers in this population, it may enable them to recognize and celebrate these assets more easily, and in partnership with caregivers. In a parallel process, a strengths-based approach can enhance the bond between the practitioner, the caregiver, and the child, thus building effective working relationships and perhaps reducing FASD-related stigma (Cosden et al., 2006; Heffron et al. 2005; Roozen et al., 2020).

Understanding the types of strengths and positive attributes of infants/toddlers with PAE from the perspective of their caregivers can inform the development of strengths-based interventions. While an extensive amount of research has documented the challenges and impairments experienced by individuals with FASD across the lifespan, less research has focused on identifying strengths at any age (Flannigan et al., 2018; Skorka et al., 2020). Four studies were identified that described caregiver perceptions of child strengths. One study found that caregivers of children, ages 5-21 years, recognized many positive traits (i.e., friendliness, hard-working, compassionate) and abilities (e.g., artistic, athletic) in their child (Sanders & Buck, 2010). A second study identified relative strengths in personal self-care and household chore activities compared with other adaptive skills for children ages 5-8 years (Jirikowic et al., 2008). Third, caregivers of school-age children reported a range of personal strengths in the context of students' educational experiences such as being artistic and having strong verbal skills and work habits (Duquette & Stodel, 2005). Fourth, a study by Pruner et al. (2020) noted that all caregivers were eager to share what they enjoyed most about their child during the early intervention period, including moments of affection, love, and laughter. Notably, Olson & Montague (2011) report strengths of young children with FASD based on informal reports which "are filled with descriptions of how engaging, innocent, straightforward, amusing, curious and social young children with an FASD can be". Taken together, these studies and informal reports highlight caregivers' awareness of child strengths and their willingness to communicate these strengths to others.

The present study was designed to address the following questions: 1) What are the concerns and strengths reported by caregivers regarding their infant/toddler (ages 7 – 42 months) with PAE? and 2) Are caregiver type (birth parent, foster/adoptive parent, other biological relative) or child age (less than 2 years, 2 years and older) associated with type or frequency of reported concerns and strengths? Examining caregiver-reported concerns may yield useful information regarding delays in child development or problem behaviors that warrant the attention of practitioners, signal the need for diagnostic referral and/or lend important insight into the impact of these concerns on families. Understanding how child age or caregiver status can influence the reporting of concerns may facilitate a more targeted approach for practitioners when inquiring about caregiver concerns and for knowing what kinds of child development information and education certain families might need. In addition, the identification of child strengths and positive characteristics can provide opportunities to enhance parent-child interactions, incorporate these strengths into interventions and help build caregiver-practitioner partnerships.

3.3 Methods

Data for the current study were collected as part of a larger retrospective chart review of diagnostic assessment data from 125 infants/toddlers seen at the University of Washington Fetal Alcohol Syndrome Diagnostic and Prevention Network (FASDPN) clinic between 2009 and 2019). This clinic does not require patients to present with a concern or delay, only a confirmed PAE at any level. Two linked studies were generated from this chart review including: 1) a descriptive study that examined the developmental, sensory processing, and behavioral outcomes of infants/toddlers with PAE (Pruner, et al., 2021a); and 2) the current study, which described caregivers' early concerns and perceptions of their infant/toddlers' strengths, based on data from two standardized caregiver questionnaires, the Infant/Toddler Sensory Profile (ITSP), and the Child Behavior Checklist/1½ -5 (CBCL). All study activities were conducted with University of Washington Human Subjects approval and caregiver consent at the time of their child's FASD diagnostic evaluation.

3.3.1 Participants

Caregivers were included in this study if their infant/toddler met inclusion criteria for the prior study (Pruner, 2021a) and they completed the ITSP questionnaire and/or the CBCL as part of their child’s FASD diagnostic evaluation. Child inclusion criteria for the first study were as follows: 1) age 1 month to 3.5 years - at the time of their FASD diagnostic evaluation; 2) received one of the following 5 diagnostic classifications reflecting the full continuum of FASD a) Fetal Alcohol Syndrome (FAS) or Partial Fetal Alcohol Syndrome (PFAS), b) Static Encephalopathy / Alcohol Exposed (SE/AE), c) Neurobehavioral Disorder / Alcohol Exposed (ND/AE), d) Sentinel Physical Findings/Alcohol Exposed, or e) No Physical Findings or Central Nervous System (CNS) Abnormalities Detected / Alcohol Exposed (Normal CNS/AE) (see Astley, 2004 for details about the FAS 4-digit diagnostic code and Pruner et al., 2021a for detailed demographics of the prior study sample); 3) had complete data on at least two domains of the Bayley Scales of Infant and Toddler Development (Bayley-III; Bayley, 2006). 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 (i.e., Bayley-III Social-Emotional and Adaptive Behavior domains, ITSP, and CBCL) not being fully completed.

3.3.2 Measures

Data for this study were collected as part of a standard intake and diagnostic process for the FAS DPN diagnostic clinic visit. Measures used for this study are described below.

Infant Toddler Sensory Profile (ITSP; Dunn, 2002). The ITSP is a 48-item caregiver questionnaire that measures sensory modulation abilities in daily life for infants/toddlers (7-36 months). Caregivers rate the frequency of infant/toddler sensory behaviors on a 5-point Likert scale. Caregivers also have the opportunity to respond to two open-ended questions: “What do you see as your child’s strengths?” and “What are your concerns?”. It is relevant to note that infants/toddlers older than 36 months were administered the Short Sensory Profile (SSP;

McIntosh et al., 1999) which does not have open-ended questions as part of the questionnaire. Therefore, caregivers of infants/toddlers older than 36 months were included in this study if they completed the CBCL only.

Child Behavior Checklist 1½−5 years (CBCL; Achenbach and Rescorla 2000). The CBCL is a 100-item caregiver questionnaire used to identify a range of emotional and behavioral problems in young children ages 1.5 – 5 years. Caregivers use a rating scale to determine the presence or absence of emotional and behavioral problems, based on the preceding 2 months. Caregivers also have to the opportunity to respond to two open-ended questions: “What concerns you most about the child?” and “Please describe the best things about your child”.

3.3.3 Descriptive Information for Participants

The two salient features from the 4-Digit Diagnostic Code and code for postnatal risk factors used in this study are described below.

4-Digit Diagnostic Code FASD Diagnosis. *FAS, PFAS, SE/AE, ND/AE, Sentinel Physical Findings/AE; No Physical or CNS Abnormalities/AE.* See Astley (2004) for a full description of diagnostic codes and features.

CNS Functional Rank. Rank 1= no dysfunction; Rank 2 = mild-to-moderate dysfunction; Rank 3 = severe dysfunction (Astley, 2004). CNS functional ranks 1-3 documents the severity of CNS dysfunction and 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.

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 perinatal complications, number of home placements, physical and/or sexual abuse, neglect, and trauma. The ranking is determined by clinical judgement at the time of the FASD evaluation and is based on available records and caregiver or other report on intake forms and/or clinical interview.

3.4 Data Analysis Plan

This study used a directed approach to content analysis (Hsieh & Shannon, 2005) to identify, categorize, and describe all instances of concerns and strengths reported by caregivers at the time of their child's diagnostic evaluation. The directed approach begins with a framework for collecting and analyzing the data but allows for new insights to emerge through a process of inductive category development (Hsieh & Shannon, 2005). In this study, researchers aimed to validate an existing framework (domains commonly assessed in early childhood) in a new context (describing caregivers' concerns and strengths). When the data did not fit into the existing framework, new categories were added to capture all possible instances of caregiver concerns and strengths (Hseih & Shannon, 2005; Kibiswa, 2019).

Written responses to the two questions from the ITSP and two questions from the CBCL were extracted verbatim, excluding any identifying information. A coding system was developed that had multiple levels. First, responses were separated into two groups based on question type - concerns versus strengths/best things. Next, the researchers read all the caregiver comments related to concerns and organized the responses into broad categories. These broad categories arose from the data to reflect general areas of function or development (i.e., Development, Behavior, General/Medical, and Caregiving). After the data related to concerns were sorted, the data within each broad category were analyzed further to create sub-categories reflecting the different examples of concerns within each broad category. Some of these sub-categories were based on domains commonly assessed in early childhood or contained in the ITSP or CBCL measures, while others arose from the data (these are identified in Tables 3.1). A similar analysis was conducted with the strengths/best things data with the broad categories being Development, Personality Traits, Interests, and Caregiving, and the sub-categories within each of these reported in Table 3.2.

Two researchers separately coded 50% of caregiver responses using the initial coding systems. They compared their results, and any discrepancies with how well the categories fit the data were discussed and adjustments to the category structure were made. This process was repeated until consensus was reached, and all responses were coded. Once the coding systems were finalized, the first author coded the remaining responses.

Frequency counts for each coded category were calculated. Responses that were left blank or completed with statements such as “no concerns at this time” or “none” were also tracked. When a response contained multiple words or phrases that were suggestive of a concern/strength, it was only coded once. For example, the description “my child is extremely social, charming and loves other kids”, was coded under Sociability one time. On the few (n = 10) occasions that a response fell under two categories, the response was coded twice. For example, the description “doesn’t seem to understand” was coded under Cognitive and Language concerns because the reason for the comprehension problems was not specified (i.e., whether it was a cognitive or language problem). Another example, “my child is easily over-stimulated”, was coded under Regulatory and Sensory Processing because of the overlapping nature of this concern. As a last step, the quantitative data were descriptively compared across age groups (i.e., < 2 years and ≥ 2 years of age) and caregiver status (i.e., biological parent, foster/adoptive parent, other biological family).

Table 3.1. Final Coding System Documenting 19 Unique Concerns

Broad Categories	Sub-Categories	Definition	Example(s)
Development	Overall Development	Concerns related to development in general	child is developmentally behind despite interventions
	Cognitive*	Concerns related to comprehension, intelligence, and learning	slow processing, doesn't know shapes, colors, or letters
	Language/Communication*	Concerns related to speech, language, receptive/expressive and social communication	she can't express what she wants, babbles like an infant a lot
	Motor/Movement*	Concerns related to fine and gross motor skills, movement	not walking, doesn't sit up
	Social-emotional*		
	Regulation	Difficulty coping with discomforting emotions	cannot quiet or calm self, easily over-stimulated
	Attachment	Difficulty bonding, seeking comfort and closeness	difficulty bonding, when's he's away from us he won't engage in play
	Adaptive Behavior*	Concerns related to everyday tasks and activities of daily living, play	baths don't go well, hard to participate in everyday, normal activities
	Sleep	Sleep-related disturbances	can't fall asleep, only sleeps 2 hours at a time
	Eating/feeding	Concerns related to feeding, eating, mealtimes	reflux, poor eating habits, gagging
Behavior	Internalizing problems**	Emotionally reactive, anxious/depressed, somatic complaints and withdrawn behaviors	quick switches between fury and happy, whiney, fussy, withdrawn
	Externalizing problems**		
	Attention problems	Problems with inattention, hyperactivity, or impulsivity	unable to sit still or focus, running and tripping into objects
	Aggressive behavior	Problems managing frustration, may hurt others or self	head banging, prolonged tantrums, kicking and screaming
	Sensory Processing***	Sensory processing behaviors that interfere with daily life	sensitive to sound, high pain tolerance, doesn't respond to name
	Behavioral Inflexibility	Difficulties adapting to changes in routine	does NOT have transitioning skills, needs a structured environment
	Safety Awareness problems	Lack of safety awareness	will walk off with stranger, fearless, impulsive
General/ Medical	Prenatal alcohol & other drug exposures	Concerns related to PAE & other drugs	I drank frequently when I was pregnant and chewed tobacco
	FAS physical findings	Concerns related to growth deficiency, microcephaly, or FAS facial features	small size, poor weight gain, small head, thin lip
	Physical or health problems	Concerns related to physical or health problems	failure to thrive, low muscle tone, born premature
Caregiving	Caregiving concerns	Concerns related to parenting, questions that arise and perceptions about their child's future	exhausted, worry that child will be unable to catch up with peers, why is he delayed, it's hard to tell if this is normal for his age

Notes. *Categories based on the Bayley-III domains; **Categories based on the CBCL scales; ***Category based on the ITSP.

Table 1.2. Final Coding System Documenting 20 Unique Strengths/Best Things

Broad Categories	Sub-Categories	Definition	Example(s)
Development	Cognitive*	Capacities related to intelligence and learning	smart, bright, problem-solver
	Language/Communication*	Capacities related to speech, language, receptive/expressive and social communication	strong communicator, voice's own opinion
	Motor*	Capacities related to motor skills and movement	loves to run, athletic, strong
	Social-emotional*		
	Regulation	Capacities related to coping with discomforting emotions	self-soothes, loves to be cuddled and really likes deep pressure
	Attachment	Capacities related to bonding, seeking comfort and closeness	hooked on mama, devoted to brother
	Adaptive Behavior*	Capacities related to everyday tasks and activities of daily living (bathing, grooming, toileting, sleep, etc.).	likes to put shoes and socks on by herself
	Eating/feeding	Capacities related to feeding, eating, drinking, mealtimes	loves food, not afraid to try different foods
Personality Traits	Happiness	Always happy, joyful, cheerful	happy child
	Love	Loving and lovable	very loving
	Kindness	Eager to help and give to others, compassionate	caring, gentle with younger brother
	Affectionate	Expressing fondness to others	gives kisses, loves snuggling
	Humor	likes to laugh and bring smiles to others	jokester, always smiling and laughing
	Sociability	Social engagement and competence	charming, outgoing, friendly
	Curiosity	Explores and is interested in trying new things	inquisitive, observant
	Courage	Brave, perseverance, resilient	fearless, very determined and persistent
	Zest	Approaching life with energy and excitement	enthusiasm, larger than life, firecracker
	Adaptable	Adapts well, patient, compliant	mild-tempered, easy going
Interests	Child Interests	Interests that capture child's attention, motivating activities	she loves animals and art, loves to be outside
Caregiving	Confidence with parenting	Responses that show a caregiver's confidence with parenting and knowing their child	we've gotten used to the things he doesn't like; child responds well to structure
	Appreciation for positive change	Responses that show appreciation for child and positive changes	great to have in my life, seems to be progressing

Notes. *Categories based on the Bayley-III domains.

3.6 Results

Records from 117 caregivers of infants/toddlers with PAE (ages 7 – 42 months) met the inclusion criteria for this study. Of these caregivers, 32% were birth parents, 44% were foster/adoptive parents and 25% were extended relatives of the child (e.g., grandparent, aunt). An overwhelming majority of the sample (91%) presented with at least some level of postnatal risk, in addition to their PAE. Eighty percent of caregivers in the sample completed the ITSP (7-36 months), while 57% completed the CBCL (1.5-3.5 years). See Table 3.3 for participant characteristics.

Table 3.3. Demographic and clinical characteristics of the 117 participants

Caregiver and child characteristic	N	(valid %)
Respondent		
Biological mother	34	(29.1)
Biological father	3	(2.6)
Other biological family member	29	(24.7)
Foster parent	44	(37.6)
Adoptive parent	7	(6.0)
Total caregiver sample size	**117**	
Completed ITSP	94	(88.7)
Completed CBCL	67	(82.7)
Completed both ITSP & CBCL	54	(46.2)
Age of child described (years)		
0.58 to 0.99	14	(12.0)
1 - 1.99	45	(38.5)
2 - 2.99	44	(37.6)
3 - 3.5	14	(12.0)
Mean (SD)	**1.99**	(0.78)
Sex of child		
Female	60	(51.3)
Male	57	(48.7)
FASD Diagnosis (Diagnostic category)		
FAS	3	(2.6)
PFAS	4	(3.4)
SE/AE	13	(11.1)
ND/AE	72	(61.5)

Sentinel physical findings/AE	5	(4.3)
No evidence of CNS abnormalities/AE	20	(17.1)
CNS Functional Rank		
Rank 1, no dysfunction	27	(23.1)
Rank 2, moderate dysfunction	86	(73.5)
Rank 3, severe dysfunction	4	(3.4)
Postnatal Risk: Rank		
1. No risk	10	(8.5)
2. Unknown risk	1	(0.9)
3. Some risk	69	(59.0)
4. High risk	37	(31.6)

Notes: Infant toddler sensory profile 7-36 months (ITSP); child behavior checklist 1.5-5 years (CBCL); fetal alcohol spectrum disorder *(*FASD); fetal alcohol syndrome (FAS); partial FAS (PFAS); static encephalopathy/ alcohol exposed (SE/AE); neurobehavioral disorder/alcohol exposed (ND/AE).

3.6.1 Concerns Identified

The coding process used for this study generated a list of 19 unique concerns expressed by caregivers (Table 3.1). Caregivers reported an average of 2.5 concerns per child, ranging from 0 - 7 concerns per child. A total of 293 concerns were reported across the study sample. The five most frequently reported concerns were related to developmental and behavioral challenges and included aggressive behavior (27%), language/communication (22%), sensory processing (21%), internalizing problems (19%), and regulation (18%). Twenty-four (24%) caregivers did not report a concern for their child on either measure. The proportion of caregivers who did not report a concern was comparable across all three caregiver groups. The frequency of reported concerns across categories are presented in Table 3.4.

Concerns were explored across age groups. For younger infants/toddlers (< 2 years), caregiver concerns expressed most often were aggressive behavior (25%; *screams at high pitches, extreme temper*), sensory processing behaviors (24%; *sensitivity to sounds, lights, clothes*), and motor skills (20%; *isn't sitting up on his own*). For older infants/toddlers (≥ 2 years), caregivers reported the most concerns with language/communication skills (29%; *slow speech, doesn't talk in more than 3-word sentences),* aggressive behavior (29%; *can throw a fit that lasts for some time*) and internalizing problems (19%; *whiny, fussy, sudden mood changes*). Notably, aggressive behaviors were reported with the most frequency across both age groups.

Concerns across caregiver status were also explored. Birth parents had the most concerns for PAE & other drug exposures (23%; *he was born addicted*), aggressive behavior (20%; *abusive*) and language/communication (18%; *worried about speech development*). The most common concerns reported by foster/adoptive parents were aggressive behavior (34%; *head butts, pulls out gobs of her own hair and even tries to pull mine out*), language /communication (25%; excessively repeats herself, doesn't talk in more than 3-word sentences), and sensory processing behaviors (26%; *has extremes in responses to stimuli*). Top concerns noted among other biological family members were language /communication (27%; *speech, constant chatter*), aggressive behavior (27%; *tantrums that are hard to calm down from*) and sensory processing (22%; *becomes inconsolable as soon as caregiver…introduces new sensation*). Aggressive behaviors were a top concern that was common across all three caregiver types.

3.6.1 Strengths Identified

The coding process generated a list of 20 unique strengths (Table 3.2). Caregivers identified an average of 3.0 perceived strengths per child, ranging from 0-7 coded strengths each. A total of 352 strengths were coded across the study sample. The most frequent strengths or best things reported were reflective of personality Traits: happiness (33%), sociability (30%), love (28%), and curiosity (26%) and developmental competencies: cognitive (22%). In contrast, strengths related to adaptive behavior (1%), eating/feeding (3%) and regulation (3%) were rarely reported. Strengths in the child interests (14%) and caregiver experience (3-5%) categories were also endorsed less frequently. Twenty percent of caregivers did not report a strength on either measure. The proportion of caregivers who did not report a strength was comparable across all three caregiver groups. Table 3.5 shows the frequency of reported strengths across categories.

Perceived strengths were explored across two age bands. For infants/toddlers (< 2 years), caregivers frequently reported the following personality traits: curiosity (37%; *observant, curious*), happiness (31%; *very happy, happy most of the time*), and love (29%; *child is lovable*). For older infants/toddlers (2 - 3.5 years), many caregivers described their child as happy (35%; *very happy girl*), social (35%; *loves to interact with me and other children*) and loving/able (30%; *very loving*).

Perceived strengths were also explored across caregiver status. Birth parents reported the most child strengths in the categories of happiness (27%; *happy baby*), love (24%; *she is so loving*), and sociability (21%; *he's a charmer*). Likewise, the most common strengths reported by foster/adoptive parents were happiness (43%; *she brings a lot of happiness to our* lives), Sociability (32%; *friendly and outgoing*) and love (27%; *loves her siblings*). Strengths expressed most often by other biological family members were love (37%; *loving boy*), sociability (33%; *her smile and ability to get along with others*) and curiosity (33%; *tries everything, observant*). Sociability and love were perceived strengths common across all three caregiver types.

Table 3.4. Number and frequency of 19 Unique Concerns Reported by 117 Caregivers

		Age Bands		**Caregiver Type**		
Child Concerns	Total sample	< 2 years	2-3.5 years	Birth parent	Foster/Adoptive parent	Other biological family
Broad and Sub-Categories	n (%)	n (%)		*n* (%)		
Developmental Concerns						
Overall development	12 (10.3)	10 (16.9)	2 (3.4)	2 (5.7)	7 (12.5)	3 (11.5)
Cognitive	14 (12.0)	5 (8.5)	9 (15.5)	3 (8.5)	7 (12.5)	4 (15.4)
Language/Communication	**26 (22.2)**	9 (15.3)	**17 (29.3)**	**6 (18.2)**	**14 (25.0)**	**7 (26.9)**
Motor/Movement	13 (11.1)	**12 (20.3)**	1 (1.7)	5 (14.3)	4 (7.1)	3 (11.5)
Social-emotional						
Regulation	**21 (17.9)**	11 (18.6)	10 (17.2)	1 (2.9)	13 (23.2)	1 (3.8)
Attachment	7 (6.0)	3 (5.1)	4 (6.9)	1 (2.9)	2 (3.8)	1 (3.8)
Adaptive Behavior	9 (7.7)	4 (6.8)	5 (8.6)	1 (2.9)	4 (7.1)	3 (11.5)
Sleep	9 (7.8)	7 (11.9)	2 (3.4)	1 (2.9)	4 (7.1)	1 (3.8)
Eating/feeding	15 (12.8)	9 (15.3)	6 (10.3)	2 (5.7)	8 (14.3)	2 (7.7)
Behavior Concerns						
Internalizing problems	**22 (18.8)**	11 (18.6)	**11 (19.0)**	2 (5.7)	13 (23.2)	5 (19.2)
Externalizing problems						
Attention problems	12 (10.3)	7 (11.9)	5 (8.6)	5 (14.3)	4 (7.1)	5 (19.2)
Aggressive behavior	**32 (27.4)**	**15 (25.4)**	**17 (28.8)**	**7 (20.0)**	**19 (33.9)**	**7 (26.9)**
Sensory Processing	**24 (20.5)**	**14 (23.7)**	10 (17.2)	4 (11.4)	**14 (25.0)**	**6 (22.2)**
Behavioral inflexibility	11 (9.4)	4 (6.8)	7 (12.1)	1 (2.9)	8 (14.3)	2 (7.7)
Safety awareness	11 (9.4)	4 (6.8)	7 (12.1)	1 (2.9)	5 (9.4)	4 (15.4)
Child Concerns (in general)						
PAE & Other Drug Exposures	16 (13.7)	9 (15.3)	7 (12.1)	**8 (22.9)**	4 (7.1)	3 (11.5)
FAS Physical Findings	10 (8.5)	6 (10.2)	4 (6.9)	1 (2.9)	4 (7.1)	5 (19.2)
Physical or health problems	12 (10.2)	9 (15.3)	3 (5.2)	3 (8.5)	7 (12.5)	2 (7.7)
Caregiving Experience	17 (14.5)	6 (10.2)	7 (12.1)	5 (14.3)	8 (14.3)	3 (11.5)
No concerns reported	28 (23.9)	16 (27.1)	13 (22.4)	10 (30.3)	11 (31.4)	7 (26.9)
Total number of caregivers	117	59	58	33	56	27

Notes. Bolded numbers indicate top 5 (total sample) or top 3 concerns (age and caregiver categories).

Table 3.5. Number and Frequency of 20 Unique Strengths/Best Things Reported by 117 Caregivers

		Child Age Bands		**Caregiver Type**		
Child Strengths/Best Things	Total sample	< 2 years	2 - 3.5 years	Birth parent	Foster/Adoptive parent	Other biological family
Broad and Sub-Categories	*n* (%)	*n* (%)		*n* (%)		
Developmental Competencies						
Cognitive	**26 (22.2)**	12 (20.3)	13 (22.4)	5 (15.1)	14 (25.0)	6 (22.2)
Language/Communication	11 (9.4)	7 (11.9)	4 (6.9)	1 (3.0)	8 (14.3)	2 (7.4)
Motor/Movement	12 (10.3)	7 (11.9)	5 (8.6)	3 (9.1)	5 (8.9)	4 (14.8)
Social-emotional						
Regulation	3 (2.6)	3 (5.1)	0	1 (3.0)	2 (3.6)	0
Attachment	12 (10.3)	9 (15.3)	3 (5.2)	3 (9.1)	8 (14.3)	1 (3.7)
Adaptive Behavior	1 (0.9)	1 (1.7)	0	0	0	1 (3.7)
Eating/feeding	4 (3.4)	3 (5.1)	1 (1.7)	1 (3.0)	1 (1.8)	2 (7.4)
Personality Traits						
Happiness	**38 (32.5)**	**18 (30.5)**	**20 (34.5)**	**9 (27.3)**	**24 (42.9)**	5 (18.5)
Love	**33 (28.2)**	**17 (28.8)**	**16 (29.6)**	**8 (24.2)**	**15 (26.8)**	**10 (37.0)**
Kindness	25 (21.4)	10 (17.0)	15 (25.9)	6 (18.2)	14 (25.0)	6 (22.2)
Affectionate	10 (8.5)	7 (11.9)	3 (5.2)	3 (9.1)	7 (12.5)	0
Humor	23 (19.7)	12 (20.3)	11 (19.0)	7 (21.2)	11 (19.6)	5 (18.5)
Sociability	**35 (29.9)**	15 (25.4)	**20 (34.5)**	**7 (21.2)**	**18 (32.1)**	**9 (33.3)**
Curiosity	**30 (25.6)**	**22 (37.2)**	8 (13.8)	7 (21.2)	13 (23.2)	**9 (33.3)**
Courage	15 (12.8)	8 (13.6)	7 (12.1)	4 (12.1)	5 (8.9)	6 (22.2)
Autonomy	12 (10.3)	9 (15.3)	3 (5.2)	3 (9.1)	4 (7.1)	6 (22.2)
Zest	18 (15.4)	6 (10.1)	12 (20.7)	6 (18.2)	6 (10.7)	6 (22.2)
Adaptable	19 (16.2)	13 (22.0)	6 (10.3)	4 (12.1)	13 (23.2)	2 (7.4)
Child Interests	16 (13.7)	7 (11.9)	9 (15.5)	4 (12.1)	10 (17.9)	1 (3.7)
Caregiver's Experience						
Confidence with parenting	3 (2.6)	2 (3.4)	1 (1.7)	1 (3.0)	2 (3.6)	0
Appreciation for positive change	6 (5.1)	2 (3.4)	4 (6.9)	0	5 (8.9)	1 (3.7)
No strengths reported	23 (19.7)	12 (20.3)	11 (19.0)	7 (21.2)	10 (17.9)	5 (18.5)
Total number of caregivers	117	59	58	33	56	27

Notes. Bolded numbers in the total sample column indicate top 5 concerns, and bolded numbers in age and caregiver columns indicate top 3 concerns.

3.7 Discussion

In this retrospective study of clinical data, caregivers of infants/toddlers with PAE described a diversity of concerns and strengths in the context of two developmental questionnaires administered as part of their child’s FASD diagnostic evaluation. Our primary findings were that (a) caregivers predominate concerns fell into categories of Aggressive Behavior and Language/Communication, while Sensory Processing and Internalizing Behaviors were also commonly reported; (b) caregiver perceived strengths spanned across numerous categories, with positive personality traits such as Happiness, Sociability, and Love expressed most often; and (c) the type and frequency of reported concerns and strengths were relatively consistent across age and caregiver status. Findings from this study recognize the value of caregivers’ perspectives and offer an important reminder to practitioners that infants/toddlers with PAE and their caregivers have strengths that can be harnessed, in addition to a range of challenges that must be addressed.

Caregivers reported a broad array of concerns, reflecting the diversity of neurodevelopmental and behavioral outcomes known to be associated with PAE during early childhood (Garrison et al., 2019; Subramoney et al., 2018). Aggressive Behaviors such as kicking and screaming, head banging, and prolonged temper tantrums raised the most concerns across both age groups and were relatively consistent across caregiver groups. Previous studies that examined behavior functioning found greater negative affect among infants (Bakhireva et al., 2018) and preschool age children with PAE (O’Conner & Paley, 2006; O’Connor et al., 2002), as well as difficult temperament (Alvik et al., 2011), and conduct problems (Alvik et al., 2013) among preschoolers. Caregivers’ frequent concerns about Language/Communication are substantiated by studies that described delayed language abilities in infants with PAE (Coles et al., 2000; Davies et al., 2011; Autti-Rämö & Granström, 1991). Atypical Sensory Processing Behaviors and Internalizing Problems were commonly described by caregivers in this study and likewise, have been reported in the literature among infants/toddlers with PAE (Fjeldsted & Hanlon-Dearman, 2009; Fjeldsted & Xue, 2019; Molteno et al., 2014; Savonlahti et al., 2005). Concerns related to Self-Regulation such as difficulty soothing, or sleep complaints were also consistent with self-regulatory difficulties seen this population (Reid & Petrenko, 2018).

Overall, it appears that caregivers raised concerns that correspond closely to outcomes from standardized measures of development, sensory processing, and behavior in the existing literature. Findings demonstrate that caregivers can be an important source of information on their child. Explicitly asking caregivers about their concerns may aid in the earlier identification of delays or problems that may arise from PAE, especially when incorporated with routine screening or comprehensive clinical assessment (Bright Futures, 2006).

Although most caregiver-reported concerns were coded using categories from the core developmental domains, ITSP, and CBCL scales, there were a few exceptions. For example, concerns related to parenting were identified by 15% of caregivers, yet the ITSP or CBCL did not explicitly prompt caregivers to consider these concerns. Along the same lines, other concerns (e.g., PAE and Other Drug Exposures, Physical and Health Problems) were reported that were not captured on the questionnaires caregivers had previously completed. While many of caregivers' developmental, behavioral, and clinical concerns would have been assessed and/or detected at the time of their child's FASD diagnostic evaluation, it is possible that concerns related to parenting may have been missed if not for the open-ended questions. Caregivers who are struggling with the day-to-day stressors of raising an infant/toddler with PAE often require additional support to engage in sensitive and responsive parenting. With this goal in mind, findings offer a reminder to practitioners that a combination of assessment approaches is needed to ensure that caregivers have more than one avenue to share concerns. Practitioners need to understand the complex problems facing caregivers, as well as their priorities and desired outcomes, to design treatment plans that are congruent with caregiver goals.

An overwhelming majority of caregivers (88%) shared one or more strengths/best things about their infant/toddler. Happy, Sociability and Love were strengths reported most often, suggesting many caregivers believed these personality traits were worth knowing about and communicating to others. The finding that caregivers endorsed Happiness and Sociability among their top two strengths is consistent with a large meta-analysis examining the benefits of frequent positive affect in individuals across multiple life domains, which found happiness to be positively correlated with sociability (Lyubomirsky et al., (2005). Based on previous research with parents of neurotypical children (ages 3 - 9), love was also a frequently endorsed character trait (Park & Peterson, 2006). For early childhood practitioners, this is useful information for initiating a working relationship with all types of caregivers, regardless of child age. Recognized strengths such as Happiness, Sociability and Love may be perceived as a healthy indicator of parent-child connectedness and a starting point for noticing and exploring these perceptions, which is especially important for family-centered care and relationship-focused approaches (Feldman, 2007; Hollman, 2010). Alternatively, when caregivers struggle to identify strengths in their child, this may signal to practitioners a need to promote attuned and positive exchanges between caregiver and child. A strengths-based approach is particularly important given the stigma associated with FASD. Both biological, as well as non-biological parents of children with PAE experience stigmatization when they are perceived as responsible for their child’s negative behavior or delayed development (Roozen et al., 2020). Cultivating child and caregiver strengths is in alignment with guiding principles of early intervention practice (Guralnick, 2011), as well as practice guidelines specific to families impacted by substance use (SAMHSA, 2012).

Caregivers identified a few characteristics in both a positive and negative light. For example, the personality trait "Zest" was reported by 16% of caregivers, when they used phrases such as "my child's personality is larger than life" or "he's a firecracker." In contrast, it appeared some caregivers perceived their child's high energy and excitement as a problem related to attention or hyperactivity (i.e., she is very driven…very hyperactive causing her to fall or run into things often). Furthermore, caregivers reported Sociability as frequent strength, yet they also identified Language/Communication problems as a frequent concern. While concerns related to the use of language/communication are fundamentally different than the personality trait of sociability, practitioners can play a role in leveraging a child's strong social skills towards the goal of developing language and communication skills. Indeed, building on strengths to compensate for child difficulties is a central intervention principle used with families raising children with FASD [Bertrand, 2009 (study #5)].

The following are study limitations. Since this was a retrospective chart review, data were limited to the written responses reported on the assessment forms. As such, we were not able to probe for further detail or ask for clarification about any of the caregiver responses. Responses to these questions were optional, and thus there may be bias or differences among caregivers who responded to the open-ended questions compared with those who did not. To gain a more thorough understanding of caregivers' concerns and child strengths and to ensure every participant had an equal opportunity to contribute, future research could take the form of guided interviews or focus groups posing similar open-ended questions. Lastly, our analysis did not account for concerns or strengths discussed during the caregiver interview, which is a standard and extensive part of the FASDPN diagnostic evaluation. Future research could explore the extent to which information gathered at the caregiver interview aligned or diverged from written responses on the assessment forms.

3.7 Conclusion

Caregivers identified concerns that warrant the attention and action of early childhood providers, demonstrating their attunement to early challenges faced by their child. Caregivers also perceived their child to have many strengths across multiple areas. These findings suggest the importance of understanding the range of concerns and strengths that caregivers perceive in their day-day interactions with their child, which can enhance the development of family-centered interventions, strengthen parent-child connectedness, and build effective working relationships between early childhood practitioners and families impacted by PAE.

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Chapter 4. Conclusion

4.1 Summary of The Dissertation

The work presented in this dissertation focuses on the early developmental and behavioral outcomes of infants/toddlers with prenatal alcohol exposure (PAE) based on results from 10 years of retrospective clinical data. Early identification and diagnosis of FASD and associated developmental challenges can bolster access to comprehensive and family-centered interventions that can help optimize early and long-term outcomes. Our findings, considered with similar studies reported in the literature, suggest that several domains of child functioning are vulnerable to the teratogenic impact of PAE and that these delays are evident in the first years of life. Our findings also showed that caregivers were attuned to the early challenges faced by their child as they identified concerns that warrant the attention and evaluation of early childhood practitioners. Lastly, caregivers highlighted a variety of child strengths that provide direction for a strengths-based approach consistent with best practices in early childhood assessment and intervention. Overall, these dissertation findings offer important implications for the early identification of infants/toddlers with PAE, as well as a reminder to practitioners to identify and utilize child strengths for planning interventions.

4.2 Implications for Clinical Practice

Based on key findings from this research, early childhood practitioners from multiple systems of care (i.e., primary health care, child welfare, early intervention, infant mental health) should consider the following enhancement (Figure 4.1) to an existing system of developmental surveillance and screening to improve capacity for the early identification of infants/toddlers with PAE. See the American Academy of Pediatrics (AAP) clinical report “Promoting Optimal Development: Identifying Infants and Young Children with Developmental Disorders Through

Developmental Surveillance and Screening" (Lipkin & Macias, 2020) for the original resource and guidance on developmental screening.

Elicit & attend to caregivers' concerns

- Listen closely for concerns related to aggressive behavior, language/communication, sensory processing and parenting.

Obtain & document developmental history

- Be sure to "ask the question" about maternal alcohol use history.

Make accurate & informed observations of the child

- Use standardized measures to screen for development, sensory processing, and challenging behaviors.

Identify risks & strengths

- Remember to ask about child and caregiver strengths. Utilize strengths for planning interventions.

Maintain an accurate record of the findings

Sharing and obtaining opinions and findings from other professionals

- Make sure to consider professionals from multiple systems of care.

Figure 4.1. Proposed Enhancement to Developmental Surveillance Components (AAP, 2020) for the early identification of infants/toddlers with prenatal alcohol exposure.

4.2.1 *Elicit and attend to caregivers' concerns about their child's development*.

Findings from Chapter 3 showed that caregivers' most frequently reported concerns were related to aggressive behavior, language/communication, sensory processing, and internalizing behaviors. It is possible that these early concerns could serve as "red flags" that warrant further investigation; however, more evidence is needed to confirm and extend these findings. Listening for and monitoring concerns related to parenting is important for practitioners working with infants/toddlers with PAE and their families.

4.2.2 Obtain and document a developmental history.

In addition to gathering information about child development and behavior that does not appear typical, early childhood practitioners should be implementing universal screenings or structured identification processes for maternal substance use history [i.e., “asking the question (s)”]. Through sensitive inquiry and positive communication strategies, practitioners can play a critical role in identifying prenatal alcohol exposures as part of early developmental histories (Watson et al., 2011).

4.2.3 Make accurate and informed observations of the child.

Findings from Chapter 2 provided evidence that standardized measures of neurodevelopment, sensory processing and emotional and behavioral functioning may be useful for identifying early indicators of delay among infants/toddlers with PAE. Given that infants/toddlers from our sample showed considerable individual variability in their development, it is essential that practitioners look at, but also beyond the core developmental domains, and screen for sensory processing differences and challenging behaviors as indicators of early regulatory concerns.

4.2.4 Identify risks and strengths and protective factors.

Findings from Chapter 3 highlight caregivers’ awareness of their child’s strengths and their willingness to communicate these strengths in the context of parent-report questionnaires that were completed as a part of a clinical diagnostic evaluation. Although the 2020 AAP clinical report recommends identifying strengths as well as risk factors of young children with developmental disorders, the findings of this study, considered with similar research investigating strengths of individuals with FASD (Petrenko et al., 2014; Skorka et al., 2020), brings new attention to the importance of a strengths-based approach to assessment and intervention in this population. Understanding child and caregiver strengths is essential to counterbalancing factors that may place them at risk and for developing holistic and family-centered interventions.

4.2.5 Maintain an accurate record of the process and findings

The identification and care of infants/toddlers with PAE can be facilitated by effective collection and documentation of information related to intake, investigation, and family needs assessment (Ingoldsby, 2021). These action steps, which are intended to improve child welfare agencies' approach to working with children with PAE, are applicable to all relevant systems of care (i.e., primary health care, early intervention, infant mental health) and are in alignment with the fifth component of developmental surveillance outlined in the 2020 AAP clinical report

4.2.6 Sharing and obtaining opinions and findings with other professionals

Bidirectional communication with a wide range of other professionals including childcare providers, home visitors and developmental therapists (i.e., speech language pathologists, physical therapists, occupational therapists, social workers) is a critical component to developmental surveillance for young children with developmental disabilities (AAP, 2020). Indeed, this group of other professionals will have important observations and contributions to share as they work with additional systems of care (i.e., primary health care, child welfare, early intervention, infant mental health) to ensure optimal care coordination of efforts for infants/toddlers with PAE and their families.

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