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Adaptive behavior in infants and toddlers with Down syndrome and fragile X syndrome

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Abstract

Individuals with Down syndrome (DS) experience deficits across all domains of adaptive functioning, however little is known about the emergence and age-related changes of these impairments compared to other neurogenetic disorders with similar intellectual disability impairments, such as fragile X syndrome (FXS). Adaptive behavior is key for optimal functioning in these populations. Participants aged 5–45 months comprised three age-matched groups, DS ($n = 64$), FXS ($n = 69$), and typically developing controls (TD; $n = 69$). Adaptive behavior was measured on the Vineland Adaptive Behavior Scales-II. Regressions were used to examine adaptive behavior in a cross-sectional design across age. DS infants and toddlers evidenced deficits across all areas of adaptive behaviors compared to the age-matched TD group, with clear impairments present in the first year of life. Motor skills were the area of greatest weakness in children with DS with significant impairment evident at 12 months of age that remained low through 3 years. Compared to age-matched children with FXS, children with DS showed initially lower standard scores at 12 months of age, but slower declines in standard scores across age, resulting in less impaired functioning at 36 months. This is the first study to compare adaptive behavior in infants and toddlers with DS to FXS, and demonstrate the phenotypic specificity of adaptive profiles in this diagnostic group. These findings provide evidence that adaptive behavior should be a major target of intervention in children with FXS and DS, and that these differences are potentially driven by unique etiologies attributable to each disorder.

Keywords

adaptive behavior; cross-sectional developmental trajectories; Down syndrome; neurogenetic disorders; fragile X syndrome

1 | INTRODUCTION

Adaptive behavior facilitates independent functioning across a variety of daily contexts and responsibilities (Schalock et al., 2010; Tassé, Luckasson, & Schalock, 2016). Measures of adaptive behavior typically include the areas of communication, daily living, and socialization. A well-developed adaptive behavior repertoire is a critical foundation for multiple long-term outcomes, including academic success (Bornstein, Hahn, & Suwalsky, 2013; Miller et al., 2017), decreased maladaptive behavior (Racz, Putnick, Suwalsky, Hendricks, & Bornstein, 2017), response to intervention (Washington, Thomas-Stonell, McLeod, & Warr-Leeper, 2015; Yoder, Woynaroski, Fey, & Warren, 2014), and independence in adulthood (Woolf, Woolf, & Oakland, 2010). Individuals with intellectual disability (ID) experience significant challenges in adaptive functioning, with impairment in adaptive behavior as a core diagnostic feature (DSM-5 American Psychiatric Association, 2013; Tassé et al., 2016).

Neurogenetic syndromes are characterized by areas of developmental competence and impairment (Hodapp & Dykens, 2012; Lee, Martin, Berry-Kravis, & Losh, 2016) with ID as an almost universal feature in a number of disorders. The presence of ID alone does not result in identical phenotypes across neurogenetic syndromes. Rather, individuals with specific neurogenetic syndromes are predisposed to display unique profiles of impairment and competence across various developmental domains, including adaptive behavior (Di Nuovo & Buono, 2011; Hodapp & Dykens, 2012). Down syndrome (DS) and fragile X syndrome (FXS) are two neurogenetic disorders with distinct genetic etiologies. Both DS and FXS have a phenotype predominantly characterized by impairments in adaptive behavior and moderate ID, although there is a range of ID from mild to more severe (Loesch, Huggins, & Hagerman, 2004; McCary, Machlin, & Roberts, 2013). Despite our current knowledge on adaptive functioning in these populations, the bulk of research has focused on school-age and older samples, with no cross-syndrome comparisons examining adaptive behavior during early development. Delineating early adaptive trends and phenotypic specificity between neurogenetic conditions, such as DS and FXS, can elucidate developmental patterns and ultimately improve long-term outcomes through targeted intervention.

1.1 | Down syndrome

DS is the most common genetic cause of ID, and occurs in approximately 1 in 700 individuals (Parker et al., 2010). Individuals with DS demonstrate a nuanced behavioral profile of developmental competence in visual processing, receptive language, and social relatedness, and impairments in motor planning, expressive language, and aspects of executive function (Daunhauer & Fidler, 2011). This nuanced behavioral profile observed later in development in DS relies on skill acquisition in these domains during infancy and early childhood (Edgin, Clark, Massand, & Karmiloff-Smith, 2015; Karmiloff-Smith, 2011). This dynamic process of phenotypic emergence likely extends to adaptive behavior, warranting the examination of adaptive skills emerging in infancy and toddlerhood in order to predict later outcomes and inform early intervention which is the focus of this study.

Existing studies on adaptive behavior have focused on school-age children and adults with DS and indicate a profile of strengths and impairments across areas of adaptive functioning. Communication appears to be an area of impairment relative to socialization (Coe et al., 1999; Dykens et al., 1994; Van Duijn, Dijkxhoorn, Scholte, & Berckelaer-Onnes, 2010), motor skills (van Duijn et al., 2010), and in some instances, daily living skills (Dykens et al., 1994). In general, adaptive skills tend to become more variable with age, and show general plateaus rather than consistent gains beginning between ages 7 (Dykens et al., 1994) and 12 years (van Duijn et al., 2010).

Only one study on adaptive behavior in DS has a targeted sample exclusively within an early developmental period (Fidler et al., 2006). Fidler et al. (2006) examined the early emergence of a DS adaptive behavior profile by studying toddlers with DS ($n = 18$; mean chronological age (CA) = 33 months-old) to CA and mental-age (MA) matched toddlers with developmental disabilities (DD; $n = 19$), and MA-matched, chronologically younger typically developing (TD) children ($n = 24$). Results indicated that the DS group showed stronger adaptive skills on the Socialization domain than the MA-matched DD group, but did not significantly differ from either the DD or TD group on any other adaptive behavior domain (Fidler et al., 2006). Collectively, these results indicate that toddlers with DS demonstrate lower adaptive abilities than expected given their chronological age across all domains of adaptive functioning, with socialization skills on par with their developmental status.

Individuals with DS have primarily been compared to typically developing children or children with developmental delay on adaptive performance (Coe et al., 1999; Fidler et al., 2006). Although these comparisons inform the nature of delay and competence in adaptive skills, they do not address phenotypic specificity of a DS adaptive profile. To our knowledge, there is only one study on adaptive functioning comparing individuals with DS to individuals with other neurogenetic syndromes (Di Nuovo & Buono, 2011). Di Nuovo and Buono (2011) compared individuals with DS, FXS, Prader-Willi, Angelman, and Williams syndrome ranging from four to 39 years-of-age on adaptive behavior domains. Results indicated significant differences between the DS and the highest functioning group (Prader-Willi syndrome), as well as the lowest functioning group (Angelman syndrome), but no significant differences between the FXS and Williams syndrome groups across any domain. Although these findings suggest overlap between DS and other neurogenetic syndromes in adaptive functioning, there are limitations. Primarily, each group was comprised of a considerably wide age-range (DS ranged between 4 and 39 years-old), which may mask age-related trends. In addition, the youngest children within this sample were 4 years-old, limiting the understanding of the early emergence of adaptive behavior in DS relative to another neurogenetic syndrome, such as FXS, given that no infants or toddlers were included in the sample. Additional work is necessary to understand early trends in adaptive behavior in DS in relation to typical development, as well as to other neurogenetic syndromes.

In sum, adaptive behavior in DS is associated with strengths in socialization and distinct challenges in communication and motor skills. However, our understanding of the early emergence of adaptive impairments in DS is limited by wide age ranges (Dykens, Hodapp,

& Evans, 1994; van Duijn, Dijkxhoorn, Scholte, & van Berkelaer-Onnes, 2010), small sample sizes (Fidler, Hepburn, & Rogers, 2006; Loveland & Kelly, 1991), or emphasizes middle childhood or later developmental periods (Coe et al., 1999; Jacola et al., 2015) across extant work. As such, little is known regarding adaptive behavior in the first 3 years of life in DS. Given impairments present in the toddler years (Fidler et al., 2006), it is likely that a DS adaptive behavior profile begins to emerge in infancy.

In addition to the limitations in understanding the early emergence of adaptive impairments in DS, there is also a considerable lack of understanding regarding the phenotypic specificity of adaptive impairments in DS relative to other neurogenetic disorders, such as FXS.

Differentiating patterns of adaptive impairments across neurogenetic can inform individualized (i.e., phenotype-specific) intervention, particularly within developmentally sensitive windows such as infancy and toddlerhood. FXS is an ideal neurogenetic disorder for cross-syndrome comparisons to DS for several reasons. First, DS and FXS are similar neurogenetic conditions that can be diagnosed either prenatally or early in infancy, although FXS is often diagnosed in the third year of life (Bailey, Raspa, Bishop & Holiday, 2009). Second, both disorders have moderate ID as a core feature, with developmental delays emerging early (Fidler et al., 2006; Roberts, McCary, Shinkareva, & Bailey, 2016; Tudella, Pereira, Basse, & Salvendy, 2011). Third, both DS and FXS are at heightened risk for comorbid psychopathology, including ADHD (Ekstein, Glick, Weill, Kay, & Berger, 2011; Sullivan et al., 2006) and autism spectrum disorder (ASD; Bailey, Hatton, Skinner, & Mesibov, 2001; DiGuseppi et al., 2010; Philofsky et al., 2004), which can exacerbate adaptive behavior impairments beyond those associated with ID.

1.2 | Fragile X syndrome

FXS is the most common inherited cause of intellectual disability (Crawford, Acuna, & Sherman, 2001), and is marked with both strengths and weaknesses in the behavioral profile (Hagerman, 2007). FXS is an X-linked disorder affecting more males than females, with an incidence rate of 1 in 5,000 males and 1 in 8,000 females (CDC, 2011; Coffee et al., 2009). There is greater variability in the behavioral profile associated with girls with FXS due to a protective factor afforded by the presence of an unaffected second X chromosome, and accordingly, much research is focused on males. Challenges associated with the FXS phenotype, including ID, and risk of comorbid autism spectrum disorder, anxiety, and ADHD (Cordeiro, Ballinger, Hagerman & Hessel, 2011; Klusek, Martin & Losh, 2014; Roberts, Weisenfeld, Hatton, Heath, & Kaufmann, 2007; Roberts et al., 2009), which likely lead to a greater propensity for challenges in adaptive outcomes.

In FXS, adaptive behavior has been more thoroughly characterized than in DS, yet our understanding remains limited because emphasis has also been on samples of older children and wide age ranges (Dyken, Hodapp, Ort, & Leckman, 1993; Dyken et al., 1996; Fisch et al., 1999, 2007; Hahn et al., 2015; Hatton et al., 2003; Klaiman et al., 2014), as well as a lack of emphasis on cross-syndrome comparisons. Extant work indicates age-related changes in adaptive skills in FXS, and areas of competence and impairment differ depending on developmental period and measurement. For example, research utilizing raw scores evidences adaptive skill growth through age 10, with plateaus or slight declines thereafter

(Dykens et al., 1996; Hahn et al., 2015), while studies examining standard scores show declines across time beginning in toddlerhood (McCary et al., 2013; Klaiman et al., 2014). In terms of general areas of competence and impairment, individuals with FXS tend to show greater abilities in daily living compared to socialization and communication (Fisch et al., 2007; Hatton et al., 2003). However, these patterns can vary depending on age-related changes. For example, Klaiman et al., (2014) examined longitudinal adaptive trajectories in 275 children with FXS between 2 and 18 years-old using standard scores, with few male participants younger than 5 years-old. Males with FXS under 5 years-old showed higher performance on Socialization than Communication and Daily Living, whereas Daily Living became an area of better performance compared to communication for older children and adolescents. In the only longitudinal infant study to date (Caravella & Roberts, 2017) infants with FXS evidenced delays in adaptive behavior skills across all domains as early as 9 months of age, indicating profiles of adaptive impairment emerge early in the lifespan. Collectively, this evidence suggests that early adaptive trends differ from those of children and adolescents with FXS, and understanding the nature of early impairments can inform not only later potential for skill acquisition, but also areas for targeted intervention.

In sum, adaptive behavior is a significant area of challenge for individuals with FXS across development. Although daily living skills are potentially an area of strength relative to other areas of adaptive functioning (Dykens et al., 1993, 1996; Rogers, Wehner, & Hagerman, 2001), this is not evident during early development (Klaiman et al., 2014; Caravella & Roberts, 2017). Examining adaptive behavior profiles in FXS during early development relative to DS will shed light on the emergence of adaptive impairments and the phenotypic specificity between these two distinct neurogenetic disorders with somewhat overlapping phenotypic features.

1.3 | Current study

The current study addresses several gaps in the literature and is the first to characterize the emergence of the adaptive behavior phenotype in infants and toddlers with DS compared to TD and FXS groups. The primary aim of this study was to characterize adaptive behavior within the first 3 years of life in a large sample of infants and toddlers with DS compared to CA matched TD infants and toddlers, as well as to an FXS group matched on CA and nonverbal mental age (NVMA).

1.4 | Research questions and hypotheses

1. At what age do adaptive behavior deficits emerge in infants and toddlers with DS?

We hypothesized that both composite and domain level deficits would emerge within the first year of life in a sample with DS contrasted to chronological age-matched neurotypical controls.

2. Does the profile of adaptive behavior in infants and toddlers with DS differ from a mental and chronological age-matched sample with FXS?

We hypothesized that infants and toddlers with DS would demonstrate similar age-related trends (*declining standard scores*) in adaptive behavior relative to infants and toddlers with

FXS. Additionally, we hypothesized that toddlers with DS would evidence better adaptive skills on Socialization, worse adaptive skills on Motor and Communication, and similar skills on Daily Living compared to the FXS group.

2 | METHODS

2.1 | Participants

Participants included 64 children with DS ($MCA = 18.81$), 69 children with FXS ($MCA = 19.87$), and 69 TD children ($MCA = 19.49$; see Table 1 for participant descriptive statistics). Participants were well-matched (Kover & Atwood, 2013) on mean CA ($F(2, 201) = 0.17$; $p = 0.85$), and the DS and FXS groups were also matched on NVMA ($p = 0.771$). Participants in the sample were pooled from multiple studies examining early development in children with neurodevelopmental disorders at three different sites, Colorado State University, the University of South Carolina, and the University of North Carolina, Chapel Hill. Inclusion in the DS group was determined by parent report of DS diagnosis. Inclusion in the group with FXS was determined by a positive FXS diagnosis per genetic report (i.e., >200 repeats of the CGG sequence on the *FMRI* gene). Inclusion criteria for the TD group, included lack of developmental delay (i.e., Mullen Early Learning composite >85 confirmed through study participation) and no family history of autism spectrum disorder. The majority of the DS and TD groups was comprised of male participants (see Table 1 for gender distributions) whereas the entire FXS group was comprised of male participants. Preliminary correlations between gender and adaptive behavior domains were examined and no significant associations were identified in either group. As part of larger batteries at each site, participants completed the Mullen Scales of Early Learning (Mullen, 1995) and their parents were interviewed with the Vineland Adaptive Behavior Scales-Survey Interview Form-Second Edition (VABS-II; Sparrow, Cicchetti, & Balla, 2005). A subset of the males with FXS ($n = 28$) had Childhood Autism Rating Scale (CARS; Schopler, Bourgonien, Wellman, & Love, 2010) data. Given the high prevalence of ASD in FXS, we examined the relationship of ASD symptoms via the CARS to the VABS in this subset of participants.

3 | MEASURES

3.1 | Vineland adaptive behavior scales

The VABS-II is a semi-structured parent interview that measures adaptive functioning across 4 domains: Communication, Socialization, Daily Living Skills, and Motor Skills. Individual items are scored on a 0–2 scale which indicates the frequency with which the individual completes the skill with independence: usually (2), sometimes (1), or never (0). A composite standard score, the Adaptive Behavior Composite (ABC), is derived from the four domain standard scores. All standard scores have a mean of 100, and standard deviation of 15. Standard scores are used in all analyses.

3.2 | Mullen scales of early learning

The Mullen Scales of Early Learning (MSEL, 1995) is a standardized measure of infant and toddler development normed for children ages 0–68 months. The Mullen measures development in five domains; Expressive Language, Receptive Language, Fine Motor, Gross

Motor, and Visual Reception. Standard scores for each domain have a mean of 50 and a standard deviation of 10. In addition to the domain scores, a composite score called the Early Learning Composite (ELC) is derived from the domains (with the exception of gross motor). The ELC has a mean of 100 and a standard deviation of 15. The MSEL also provides nonverbal mental age (NVMA) by averaging the Age Equivalence scores from the Fine Motor and Visual Reception domains. This was calculated for all participants except for a subset of participants with DS that were assessed as an initial pilot sample ($n = 21$). Due to time constraints of pilot data collection, these participants only had a measure of Visual Reception and these age-equivalence scores were used as a proxy of NVMA.

3.3 | Childhood autism rating scale

The CARS is a 15-item rating scale of general autistic behavior. Items are scored on a four-point Likert scale, where 1 = within normal limits (given age) and 4 = severely abnormal (given age or developmental level). Scores across the 15 items are summed for a total score in which 30–37 indicates mild to moderate autism symptomatology or behavior. CARS data were available for only a subset of the FXS participants 24 months and older due to multiple data sources and considerations regarding the age at which presence of autism symptomatology is more reliable. Thus, total CARS scores were used in post hoc analyses for a subset of the FXS group ($n = 28$) to determine the association between autism symptomatology and adaptive behavior.

3.4 | Procedures

Ethical approval was obtained from each Institutional Review Board at Colorado State University, the University of South Carolina, and the University of North Carolina, Chapel Hill. Participants in the DS and FXS groups were recruited through local and national organizations serving each population. TD children were recruited through community based sites (e.g., pediatricians' offices) and social media. Across the data collection sites, assessments were conducted primarily in the participants' homes. Parents received a summary of their child's developmental assessment (Colorado State University) or a developmental summary and monetary compensation for participating (the University of South Carolina, the University of North Carolina, Chapel Hill).

3.5 | Analytic approach

All descriptive statistics and group difference effect sizes (i.e., Cohen's d) are presented in Table 1. Group differences and age-by-group interactions were examined using a regression framework across: Communication, Daily Living Skills, Socialization, Motor Skills, and the Adaptive Behavior Composite. Dummy coded values were computed for each group and entered into each model, such that the DS group served as the reference group. Age was centered at 24 months for primary analyses and entered into each model, along with age by group interaction terms for both the FXS and TD groups. Age was centered at 24-months as this was a relative median age of the sample and provided for probing the interaction at equidistant age points in either direction included in the sample. Thus, significant age by group interactions were probed by recentering age at 12 months and 36 months, respectively. Table 2 presents primary model results with age centered at 24 months. Figure 1 presents

graphed age-by-group trends for overall adaptive behavior, as well as each subdomain of adaptive behavior.

4 | RESULTS

4.1 | Adaptive behavior composite

4.1.1 | DS versus TD—The DS group scored significantly lower by approximately 24 standard points on general adaptive behavior than the TD group ($b = 24.02$; $p = < 0.001$; see Table 2). The lack of significant age-by-group interaction between the DS and TD groups indicated that adaptive behavior impairments emerge for infants with DS within the first year of life, and that TD infants and toddlers outperformed the DS group across all ages included in the study.

4.1.2 | DS versus FXS—There was no significant group difference on overall adaptive outcomes between 24 month-olds with DS and FXS, yet there was a significant age by group interaction ($b = -0.608$; $p = < 0.001$). Results from probing the interaction indicated that 12 month olds with DS scored significantly lower than 12 month-olds with FXS ($b = 5.19$; $p = 0.009$), but 36 month-olds with DS performed significantly higher on adaptive behavior outcomes relative to children with FXS ($b = -9.40$; $p = 0.002$). Thus, across age, participants with FXS demonstrate more rapid declines in adaptive behavior standard scores (i.e., a lesser ability to keep pace with developmental gains in adaptive skills) relative to children with DS, with the most pronounced difference in overall adaptive behavior occurring in 36 month-olds.

4.2 | Communication

4.2.1 | DS versus TD—The 24 month-olds with DS scored significantly lower than 24 month-old TD participants on the Communication domain by approximately 27 standard points ($b = 26.60$, $p = < 0.001$), and there was a significant interaction. Results from probing the interaction however, indicated that the DS group scored significantly lower than the TD group at both 12 months ($b = 19.67$; $p = < 0.001$) as well as 36 months ($b = 33.53$; $p = < 0.001$). Collectively, these results indicated that infants with DS may have similar communication skills to TD infants at a much earlier age, such as prior to 6 months-old, but clear communication deficits emerge by 12 months.

4.2.2 | DS versus FXS—There was no significant difference between 24 month-olds in the DS and FXS groups on communication (see Table 2), but there was a significant age by group interaction ($b = -0.45$; $p = 0.029$). Results from probing the interaction indicated that there was no significant difference between groups at 12 months, but at 36 months, there was a marginally significant difference ($b = -7.55$; $p = 0.064$), such that children with DS were predicted to score approximately 7.5 points higher on standard communication scores relative to FXS children. Thus, DS and FXS have similar communication profiles across the first 3 years of life that begin to diverge at 36 months.

4.3 | Daily living

4.3.1 | DS versus TD—On the Daily Living domain, DS and TD 24 month-olds significantly differed ($b = 15.71$; $p = <0.001$), such that toddlers with DS scored approximately 16 standard points lower than the TD group. There was no significant age-by-group interaction for the DS and TD groups, indicating that deficits in Daily Living skills emerge in DS within the first year of life and remain consistent across ages included in the study.

4.3.2 | DS versus FXS—There was no predicted difference between 24 month-olds in the DS and FXS groups, but there was a significant age by group interaction ($b = 5.67$; $p = 0.014$). Results from probing the interaction indicated that 12 month old children with DS scored significantly lower on Daily Living outcomes relative to children with FXS, such that children with DS were predicted to perform approximately six standard points lower than children with FXS. However, at 36 months the DS group scored significantly higher by approximately 12 standard points than the FXS ($b = -12.39$; $p = 0.001$). Collectively, this indicated infants with DS demonstrate early deficits but slower rates of decline in Daily Living *standard scores* compared to infants with FXS, resulting in higher standard scores by the third year of life relative to the FXS group.

4.4 | Socialization

4.4.1 | DS versus TD—DS and TD 24 month-olds significantly differed on socialization ($b = 16.55$; $p = 0.001$), such that the DS group scored approximately 17 standard points lower on the Socialization domain than the TD group. There was no significant age-by-group interaction between the DS and TD groups, indicating that impairments in Socialization emerge prior to 12 months and remain consistent across ages included in the study.

4.4.2 | DS versus FXS—The DS and FXS groups also significantly differed on the Socialization domain ($b = -4.14$; $p = 0.023$), such that at 24 months, the DS group scored approximately four standard points higher on socialization compared to 24-month-old with FXS. There was a significant age by group interaction ($b = -0.54$; $p = 0.001$). Results from probing the interaction indicated socialization differences between groups was not present at 12 months, and that the most pronounced difference occurred in 36-month-old, with the DS group scoring approximately 11 standard points higher on socialization than the FXS group ($b = -10.57$; $p = 0.001$).

4.5 | Motor

4.5.1 | DS versus TD—The DS and TD groups significantly differed at 24 months on motor outcomes ($b = 26.04$, $p = <0.001$). There was no age-by-group interaction, indicating that deficits in motor skills were significant and present in infants and toddlers with DS across all ages included in the study.

4.5.2 | DS versus FXS—Motor performance did not significantly differ between DS and FXS 24 month-olds. There was, however, a significant age-by-group interaction ($b = -0.67$; $p = <0.001$). Results from probing the interaction indicated that 12-month-old with

DS demonstrate significant deficits on motor compared to 12-month-old with FXS, scoring 11 standard points lower ($b = 10.51$; $p < 0.001$). There was no significant group difference evident in 36-month-old, indicating that across ages, toddlers with DS make small gains and eventually stabilize in their motor skill acquisition, converging with the declining trend in skill acquisition observed across ages in toddlers with FXS.

4.6 | Post-hoc correlations

Data were drawn from multiple sources and much of the study sample was focused on children under 24 months with ASD measures included only for the FXS studies.

Accordingly, CARS ratings for ASD severity were only available for a portion of the FXS participants 24 months and older ($n = 28$). Significant associations were found between ASD severity and the Socialization ($r = -.40$; $p = 0.036$) and Communication ($r = -.45$; $p = 0.024$) domains.

5 | DISCUSSION

Adaptive behavior is critical for successful daily functioning and is related to optimal outcomes across cognitive, academic, and mental health domains (Bornstein et al., 2013; Kanne et al., 2011; Racz et al., 2017). Impairments in adaptive behavior are characteristic of ID and, as such, are associated with a number of specific neurogenetic disorders including DS (Daunhauer, 2011). While existing research has begun to identify an adaptive behavior phenotype in DS, our understanding of this profile in the first years of life is quite limited. The current study is the first to evaluate adaptive behavior in infants and toddlers with DS compared to a CA-matched TD group, and a CA and NVMA-matched group with FXS. Findings from this study identify global impairments in adaptive behavior for infants and toddlers with DS relative to TD children, with delays emerging in the first year of life across all domains of adaptive functioning. Age-related trends identified in this study indicate that young children with DS demonstrate difficulty keeping pace in all areas of adaptive functioning, showing a deceleration in adaptive trends across ages, with the most pronounced discrepancies between DS and TD evident in motor and communication skills. In addition, results highlight the phenotypic specificity of emergent adaptive behavior in DS. Specifically, infants and toddlers with DS demonstrated differential levels and trends in adaptive behavior across ages and domain when compared to the FXS group. While both the DS and FXS group demonstrated declining trends in adaptive behavior standard scores, the DS group showed initially lower adaptive functioning in all areas, yet a slower rate of decelerated skill acquisition compared to the FXS group. Results from the current study provide insight into the emergent nature of adaptive challenges in DS and offer implications for targeted early intervention.

As hypothesized, infants and toddlers with DS demonstrated early and consistent deficits in adaptive behavior that emerged between 5 and 12-month-of-age, and were persistent through 45 months relative to the TD group. A decline in the rate of adaptive skill development for the DS group was evident as the discrepancy between standard scores became more pronounced across ages (see Table 1 and Figure 1). Based on mean difference effect sizes, communication, and motor were the two areas of greatest divergence between DS and TD

groups, and group differences persisted from 5 to 45 month-of-age. Both motor and communication skills serve as critical foundational abilities associated with both concurrent and later developmental outcomes. Specifically, early motor skills facilitate learning and exploration of one's environment, as well as overall cognitive development (Bornstein et al., 2013; Howe & Lewis, 2005). In addition, motor exploration within infancy has been linked to higher academic achievement in early adolescence (Bornstein et al., 2013). Early communication skills encompass the receptive understanding of language in the environment, as well as shared enjoyment, vocalizations, and use of gestures. These foundational skills lead to more developmentally advanced skills, including social reciprocity and joint attention (Wetherby, 2006). These early skills are therefore critical for success within one's daily environment and interactions, but also as a fundamental developmental foundation.

The cross-syndrome comparisons yielded some very interesting and, somewhat unexpected, findings. Overall, we found distinct age-related trends in adaptive behavior in infants and toddlers with DS contrasted to those with FXS. Across all areas of adaptive behavior, infants with DS demonstrated lower skills than infants with FXS at 12-months-of-age, though these differences were statistically significant only for the Daily Living and Motor domains and for the ABC Composite. The unanticipated age-related shifts from lower to greater competence relative to the FXS group were primarily driven by the steeper decline in rate of adaptive skill acquisition in the FXS group, despite initially stronger performance. Resulting trends indicated that groups converged on most areas of adaptive behavior at 24 months. Toddlers with DS at 36 months displayed more advanced adaptive skills across all domains with statistically significant differences present in Daily Living domain and the ABC Composite and at a trend for the Communication domain. It is important to note that decreasing trends in standard scores across age may not indicate developmental regression, but rather, a lack of keeping pace with developmental expectations regarding adaptive behavior. We informally examined mean domain raw scores trends across age between the DS and FXS group to confirm the lack of developmental regression within these groups. For both the DS and FXS groups, mean raw scores of all adaptive behavior domains increased across ages, which is consistent with prior work in infants and toddlers with FXS (Caravella & Roberts, 2017).

Despite group convergence on all domains of adaptive functioning by 24 months, toddlers with DS showed greater capabilities in keeping pace with age-expected adaptive skill acquisition than the FXS group. Resulting trends indicated significantly higher adaptive performance in daily living, communication, and socialization in DS toddlers compared to FXS toddlers. One potential influence on the differential trends identified between the DS and FXS groups is elevated ASD symptomatology. Comorbid ASD occurs at elevated rates in FXS estimated at approximately 50–70% males with FXS (Hall, Lightbody, & Reiss, 2008). Post hoc correlations examined in this study, as well as existing longitudinal work (Caravella & Roberts, 2017), support the partial influence of increased ASD symptomatology in the FXS group on differences across some areas of adaptive behavior compared to the DS group- most notably socialization and communication. Group differences in daily living skills may not be accounted for by elevated ASD severity in FXS, as a significant association was not identified in post hoc correlations. Direct assessment of

ASD comorbidity with all participants across all groups was beyond the scope of the current study and additional work is necessary to fully determine the differential effect of ASD symptomatology on adaptive behavior in children with DS, particularly compared to other neurogenetic disorders such as FXS.

Our findings identify emergent impairments across multiple domains of adaptive behavior within the first 3 years of life in both the FXS and DS groups, which are of concern given that competence in these domains is critical for optimal outcomes (Bornstein et al., 2013; Needham & Libertus, 2011; Woolf et al., 2010). Collectively, results highlight the importance of using a developmental framework to examine phenotypic profiles as the groups looked quite different across ages despite equivalent chronological and mental ages. These differences were such that the DS group demonstrated greater impairment at 12-month-of-age, few differences at 24-month-of-age and less impairment at 36-month-of-age relative to the FXS group. These phenotype specific adaptive trends suggest that DS and FXS have unique adaptive behavior profiles within the first 3 years of life and, therefore, may benefit from individualized intervention approaches to address their greatest areas of impairment and to draw on existing areas of competence.

6 | LIMITATIONS AND FUTURE RESEARCH

Although current findings contribute to a significant gap in the understanding of early adaptive behavior in DS, this study was not without limitations. Cross-sectional trajectories were examined to determine age-related trends and phenotypic specificity in infants and toddlers with DS relative to CA-matched FXS and TD groups. While this approach is highly informative regarding age-related trends and differences between groups, longitudinal methods can better elucidate adaptive trends across development. Such efforts would further characterize the nature of impairments in adaptive skills in DS as well as phenotypic specificity relative to FXS. In addition, our sample included only males with FXS, but females with DS. Although no gender associations were identified in the DS group in preliminary analyses, a more direct comparison of DS to FXS males only may demonstrate different age-related trends between groups than we identified in this study.

Future research may also consider alternative methods, such as lab-based measures to confirm or further delineate patterns of phenotypic specificity between DS and FXS across certain areas. For example, within the Communication domain, children with DS may demonstrate greater needs in expressive skills whereas children with FXS may have greater needs in the area of pragmatic language abilities (Abbeduto, Warren, & Conners, 2007; Martin et al., 2017). Another limitation is the lack of assessment of ASD features in the DS group. Individuals with DS are also at risk for increased ASD comorbidity, which may influence adaptive behavior. Future work should aim to further delineate group differences in adaptive behavior as a function of ASD symptomatology in children with DS compared to children with FXS or other neurodevelopmental disorders. Finally, future empirical efforts should also focus on evaluating long term outcomes associated with early adaptive impairments in DS, as these foundations are critical for a multitude of outcomes (Bornstein et al., 2013; Woolf, Woolf, & Oakland, 2010).

7 | SUMMARY AND IMPLICATIONS

The current study provides evidence that unique developmental profiles characterize the early emergence of adaptive behavior in DS compared to FXS, two neurogenetic disorders associated with ID and adaptive impairments. Our findings are particularly striking given that the groups were matched on both chronological and nonverbal mental age and that both groups showed significant developmental delays across age. Thus, this work challenges assumptions that adaptive behavior impairments align with ID, or with early age in a simple linear manner. Results suggest that infants and toddlers with DS and FXS are in great need of early intervention across all domains of adaptive behavior, but with syndrome specific differences.

7.1 | Implications for targeted intervention

Collectively, findings have several implications for early intervention. DS and FXS are two unique neurogenetic disorders with the potential to be identified very early in the lifespan, or even prenatally (Bailey et al., 2009; Edgin et al., 2015), although FXS is typically diagnosed around 3 years (Bailey et al., 2009). Thus, phenotypic specificity is important to inform both differential diagnosis of early impairments and to inform the individualization of treatment. Eligibility for early intervention services through Part C of IDEA (IDEA, 2004) results in the recommendation for children with DS begin receiving some intervention services within the first year of life (Davidson, 2008; Rihtman et al., 2010). Similarly, children with FXS may begin early intervention by 21 months (Hatton et al., 2000), and services for both populations may include speech, physical, and occupational therapy. Results from this study, however, suggest potential limitations to early intervention as currently provided to neurogenetic groups.

Despite nearly all participants with DS or FXS in this study having a history of early intervention, we reported consistent declines in standard adaptive behavior scores across age, with the exception of motor skill acquisition for those with DS. Infants and toddlers with FXS demonstrated consistent and steeper declines in standard scores across ages and all domains of adaptive functioning, again despite the presence of early intervention. Thus, current findings suggest that young children with neurogenetic disorders may benefit from an increased dosage of therapy, or syndrome-specific intervention to both build and maintain adaptive skills. Those with DS may need more intensive intervention targeting adaptive skills in the first year of life, particularly in the areas of motor and daily living. Infants and toddlers with FXS also need a great deal of support to develop and maintain their adaptive skills in the first 3 years of life; however, they may need more focus on maintaining skills most notably starting at 2 and 3 years of age. Innovations to current therapeutic practices to better individualize treatment across neurogenetic conditions is warranted. For example, leveraging strengths in socialization to target motor and communication delays for children with DS (Will & Hepburn, 2015), and leveraging of motor skills to target other areas of greater impairment for children with FXS, may achieve greater therapeutic impact and outcomes for each of these populations.

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REFERENCES

- Abbeduto L, Warren SF, & Conners FA (2007). Language development in Down syndrome: From the prelinguistic period to the acquisition of literacy. *Developmental Disabilities Research Reviews*, 13(3), 247–261.
- Bailey DB, Hatton DD, Skinner M, & Mesibov G (2001). Autistic behavior, FMR1 protein, and developmental trajectories in young males with fragile X syndrome. *Journal of Autism and Developmental Disorders*, 31(2), 165–174. [PubMed: 11450815]
- Bailey DB, Raspa M, Bishop E, & Holiday D (2009). No change in the age of diagnosis for fragile X syndrome: Findings from a national parent survey. *Pediatrics*, 124(2), 527–533. [PubMed: 19581269]
- Bornstein MH, Hahn C, & Suwalsky JTD (2013). Physically developed and exploratory young infants contribute to their own long-term academic achievement. *Psychological Science*, 24(10), 1906–1917. [PubMed: 23964000]
- Caravella KE, & Roberts JE (2017). Adaptive skill trajectories in infants with fragile X syndrome contrasted to typical controls and infants at high risk for autism. *Research in Autism Spectrum Disorders*, 40, 1–12. [PubMed: 29170682]
- Centers for Disease Control and Prevention. (2011). Fragile X syndrome. <http://www.cdc.gov/Features/FragileX/>
- Coe DA, Matson JL, Russell DW, Slifer KJ, Capone GT, Baglio C, & Stallings S (1999). Behavior problems of children with Down syndrome and life events. *Journal of Autism and Developmental Disorders*, 29(2), 149–156. [PubMed: 10382135]
- Coffee B, Keith K, Albizua I, Malone T, Mowrey J, Sherman SL, & Warren ST (2009). Incidence of fragile X syndrome by newborn screening for methylated FMR1 DNA. *The American Journal of Human Genetics*, 85(4), 503–514. [PubMed: 19804849]
- Cordeiro L, Ballinger E, Hagerman R, & Hessler D (2011). Clinical assessment of DSM-IV anxiety disorders in fragile X syndrome: Prevalence and characterization. *Journal of Neurodevelopmental Disorders*, 3(1), 57–67. [PubMed: 21475730]
- Crawford DC, Acuna JM, & Sherman SL (2001). FMR1 and the fragile X syndrome: Human genome epidemiology review. *Genetics in Medicine*, 3(5), 359–371. [PubMed: 11545690]
- Daunhauer LA (2011). The early development of adaptive behavior and functional performance in young children with Down syndrome: Current knowledge and future directions. *International Review of Research in Developmental Disabilities*, 40, 109–131.
- Daunhauer LA, & Fidler DJ (2011). The Down syndrome behavioral phenotype: Implications for practice and research in occupational therapy. *Occupational Therapy in Health Care*, 25(1), 7–25. [PubMed: 23898980]
- Davidson MA (2008). Primary care for children and adolescents with Down syndrome. *Pediatric Clinics of North America*, 55(5), 1099–1111. [PubMed: 18929054]
- DiGuseppi C, Hepburn S, Davis JM, Fidler DJ, Miller L, Ruttenber M, & Robinson C (2010). Screening for autism spectrum disorders in children with Down syndrome: Population prevalence and screening test characteristics. *Journal of Developmental and Behavioral Pediatrics*, 31, 181–191. [PubMed: 20375732]

- Di Nuovo S, & Buono S (2011). Behavioral phenotypes of genetic syndromes with intellectual disability: Comparison of adaptive profiles. *Psychiatry Research*, 189(3), 440–445. [PubMed: 21507490]
- DSM-5 American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders*. Arlington: American Psychiatric Publishing.
- Dykens EM, Hodapp RM, & Evans DW (1994). Profiles and development of adaptive behavior in children with down. *American Journal on Mental Retardation*, 98(5), 580–587. [PubMed: 8192903]
- Dykens EM, Hodapp RM, Ort SI, & Leckman JF (1993). Trajectory of adaptive behavior in males with fragile X syndrome. *Journal of Autism and Developmental Disorders*, 23(1), 135–145. [PubMed: 8463193]
- Dykens E, Ort S, Cohen I, Finucane B, Spiridigliozzi G, Lachiewicz A, ... O'Connor R (1996). Trajectories and profiles of adaptive behavior in males with fragile X syndrome: Multicenter studies. *Journal of Autism and Developmental Disorders*, 26(3), 287–301. [PubMed: 8792261]
- Edgin JO, Clark CAC, Massand E, & Karmiloff-Smith AD (2015). Building an adaptive brain across development: Targets for neuro-rehabilitation must begin in infancy. *Frontiers in Behavioral Neuroscience*, 9, 1–15. [PubMed: 25653603]
- Ekstein S, Glick B, Weill M, Kay B, & Berger I (2011). Down syndrome and attention-deficit/hyperactivity disorder (ADHD). *Journal of Child Neurology*, 26(10), 1290–1295. [PubMed: 21628698]
- Fidler D, Hepburn S, & Rogers S (2006). Early learning and adaptive behaviour in toddlers with Down syndrome: Evidence for an emerging behavioural phenotype? *Down Syndrome Research and Practice*, 9(3), 37–44.
- Fisch G, Carpenter NJ, Holden JJ, Simensen R, Howard-Peebles PN, Maddalena A, ... Nance, W. (1999). Longitudinal assessment of adaptive and maladaptive behaviors in fragile X males: Growth, development, and profiles. *American Journal of Medical Genetics*, 83(4), 257–263. [PubMed: 10208158]
- Fisch G, Carpenter N, Howard-Peebles PN, Holden JJA, Tarleton J, Simensen R, & Nance W (2007). Studies of age-correlated features of cognitive-behavioral development in children and adolescents with genetic disorders. *American Journal of Medical Genetics, Part A*, 143A(20), 2478–2489. [PubMed: 17853466]
- Hagerman RJ, (2007). Etiology, diagnosis, and development in fragile X syndrome. In Roberts JE, Chapman RS, & Warren SF (Eds.), *Speech and language development and intervention in Down syndrome and fragile X syndrome* (pp. 27–42). Baltimore, MD: Brookes.
- Hatton DD, Bailey DB, Roberts JP, Skinner M, Mayhew L, Clark RD, Waring E, & Roberts JE (2000). Early intervention services for young boys with fragile x syndrome. *Journal of Early Intervention*, 23(4), 235–251.
- Hatton DD, Wheeler AC, Skinner ML, Bailey DB, Sullivan KM, Roberts JE, ... Clark RD (2003). Adaptive behavior in children with fragile X syndrome. *American Journal on Mental Retardation*, 108(6), 373–390. [PubMed: 14561110]
- Hahn LJ, Brady NC, Warren SF, & Fleming KK (2015). Do children with fragile X syndrome show declines or plateaus in adaptive behavior? *American Journal on Intellectual and Developmental Disabilities*, 120(5), 412–432. [PubMed: 26322389]
- Hall SS, Lightbody AA, & Reiss AL (2008). Compulsive, self-injurious, and autistic behavior in children and adolescents with fragile X syndrome. *American Journal on Mental Retardation*, 113(1), 44–53. [PubMed: 18173299]
- Hodapp RM, & Dykens EM (2012). Genetic disorders of intellectual disability: Expanding our concepts of phenotypes and of family outcomes. *Journal of Genetic Counseling*, 21(6), 761–769. [PubMed: 22918667]
- Howe ML, & Lewis MD (2005). The importance of dynamic systems approaches for understanding development. *Developmental Review*, 25(3), 247–251.
- Individuals with Disabilities Education Act of 2004. (2004). 20 U. S. C. §1400 et seq.

- Jacola LM, Edelstein K, Liu W, Pui CH, Hayashi RJ, Kadan-Lottick N, ... Krull KR (2015). Cognitive, behavior and academic problems in adolescent survivors of childhood acute lymphoblastic leukemia (ALL): A report from the childhood cancer survivor study.
- Kanne SM, Gerber AJ, Quirnbach LM, Sparrow SS, Cicchetti DV, & Saulnier CA (2011). The role of adaptive behavior in autism spectrum disorders: Implications for functional outcome. *Journal of Autism and Developmental Disorders*, 41(8), 1007–1018. [PubMed: 21042872]
- Karmiloff-Smith A (2011). Static snapshots versus dynamic approaches to genes, brain, cognition, and behavior in neurodevelopmental disabilities. *International Review of Research in Developmental Disabilities*, 40, 1–15.
- Klaiman C, Quintin E, Jo B, Lightbody A, Hazlett HC, Piven J, ... Reiss A (2014). Longitudinal profiles of adaptive behavior in fragile X syndrome. *Pediatrics*, 134(2), 315–324. [PubMed: 25070318]
- Klusek J, Martin GE, & Losh M (2014). Consistency between research and clinical diagnoses of autism among boys and girls with fragile x syndrome. *Journal of Intellectual Disability Research*, 58(10), 940–952. [PubMed: 24528851]
- Kover ST, & Atwood AK (2013). Establishing equivalence: Methodological progress in group-matching design and analysis. *American Journal on Intellectual and Developmental Disabilities*, 118(1), 3–15. [PubMed: 23301899]
- Lee M, Martin GE, Berry-Kravis E, & Losh M (2016). A developmental, longitudinal investigation of autism phenotypic profiles in fragile X syndrome. *Journal of Neurodevelopmental Disorders*, 8(1), 47. [PubMed: 28050218]
- Loesch DZ, Huggins RM, & Hagerman RJ (2004). Phenotypic variation and FMRP levels in fragile X. *Developmental Disabilities Research Reviews*, 10(1), 31–41.
- Loveland KA, & Kelley ML (1991). Development of adaptive behavior in preschoolers with autism or down syndrome. *American Journal on Mental Retardation*, 96(1), 13–20. [PubMed: 1831619]
- Martin GE, Barstein J, Hornickel J, Matherly S, Durante G, & Losh M (2017). Signaling of noncomprehension in communication breakdowns in fragile X syndrome, Down syndrome, and autism spectrum disorder. *Journal Of Communication Disorders*, 65, 6522–6534.
- McCary LM, Machlin L, & Roberts JE (2013). The development of adaptive behaviour in toddlers and preschoolers with fragile X versus autism. *International Journal of Developmental Disabilities*, 59(2), 67–79. [PubMed: 25191537]
- Miller LE, Burke JD, Troyb E, Knoch K, Herlihy LE, & Fein DA (2017). Preschool predictors of school-age academic achievement in autism spectrum disorder. *The Clinical Neuropsychologist*, 31(2), 382–403. [PubMed: 27705180]
- Mullen EM (1995). Mullen scales of early learning. Circle Pines, MN: AGS, (pp. 58–64).
- Needham A, & Libertus K (2011). Embodiment in early development. *Wiley Interdisciplinary Reviews: Cognitive Science*, 2(1), 117–123. [PubMed: 26301917]
- Parker SE, Mai CT, Canfield MA, Rickard R, Wang Y, Meyer RE, ... Correa A (2010). Updated national birth prevalence estimates for selected birth defects in the United States, 2004–2006. *Birth Defects Research Part A: Clinical and Molecular Teratology*, 88(12), 1008–1016. [PubMed: 20878909]
- Philofsky A, Hepburn SL, Hayes A, Hagerman R, & Rogers SJ (2004). Linguistic and cognitive functioning and autism symptoms in young children with fragile x syndrome. *American Journal on Mental Retardation*, 109(3), 208–218. [PubMed: 15072521]
- Racz SJ, Putnick DL, Suwalsky JT, Hendricks C, & Bornstein MH (2017). Cognitive abilities, social adaptation, and externalizing behavior problems in childhood and adolescence: Specific cascade effects across development. *Journal of Youth and Adolescence*, 46(8), 1688–1701. [PubMed: 27815666]
- Rihtman T, Tekuzner E, Parush S, Tenebaum A, Bachrach SJ, & Ornoy A (2010). Are the cognitive functions of children with Down syndrome related to their participation? *Developmental Medicine and Child Neurology*, 52(1), 72–78. [PubMed: 19758365]
- Roberts JE, Mankowski JB, Sideris J, Goldman BD, Hatton DD, Mirrett PL, ... Bailey DB Jr. (2009). Trajectories and predictors of the development of very young boys with fragile X syndrome. *Journal of Pediatric Psychology*, 34, 827–836. [PubMed: 19074489]

- Roberts JE, McCary LM, Shinkareva SV, & Bailey DB, Jr (2016). Infant development in fragile X syndrome: Cross-syndrome comparisons. *Journal of Autism and Developmental Disorders*, 46(6), 2088–2099. [PubMed: 26864160]
- Roberts JE, Weisenfeld LAH, Hatton DD, Heath M, & Kaufmann WE (2007). Social approach and autistic behavior in children with fragile X syndrome. *Journal of autism and developmental disorders*, 37(9), 1748–1760. [PubMed: 17180715]
- Rogers SJ, Wehner EA, & Hagerman R (2001). The behavioral phenotype in fragile X: symptoms of autism in very young children with fragile X syndrome, idiopathic autism, and other developmental disorders. *Journal of developmental & behavioral pediatrics*, 22(6), 409–417. [PubMed: 11773805]
- Schalock RL, Borthwick-Duffy SA, Bradley VJ, Buntinx WH, Coulter DL, Craig EM, ... Shogren KA (2010). Intellectual disability: Definition, classification, and systems of supports. American Association on Intellectual and Developmental Disabilities. 444 North Capitol Street NW Suite 846, Washington, DC 20001.
- Schopler E, Van Bourgondien ME, Wellman GJ, & Love SR (2010). The childhood autism rating scale, (CARS2). Los Angeles: WPS.
- Sparrow SS, Cicchetti DV & Balla DA (2005). Vineland adaptive behavior scales: Survey forms manual (2nd ed.). Circles Pines, MN: American Guidance Service.
- Sullivan K, Hatton D, Hammer J, Sideris J, Hooper S, Ornstein P, & Bailey D (2006). ADHD symptoms in children with FXS. *Am J Med Genet Part A*, 140A, 2275–2288.
- Tassé MJ, Luckasson R, & Schalock RL (2016). The relation between intellectual functioning and adaptive behavior in the diagnosis of intellectual disability. *Intellectual and Developmental Disabilities*, 54(6), 381–390. [PubMed: 27893317]
- Tudella E, Pereira K, Basso RP, & Savelsbergh GJP (2011). Description of the motor development of 3–12 month old infants with Down syndrome: The influence of the postural body position. *Research in Developmental Disabilities*, 32, 1514–1520. [PubMed: 21367575]
- Van Duijn G, Dijkxhoorn Y, Scholte EM, & Berckelaer-Onnes V (2010). The development of adaptive skills in young people with Down syndrome. *Journal of Intellectual Disability Research*, 54(11), 943–954. [PubMed: 20704634]
- Washington KN, Thomas-Stonell N, McLeod S, & Warr-Leeper G (2015). Outcomes and predictors in preschoolers with speech-language and/or developmental mobility impairments. *Child Language Teaching and Therapy*, 31(2), 141–157.
- Wetherby AM (2006). Understanding and measuring social communication in children with autism spectrum disorders. *Social and Communication Development in Autism Spectrum Disorders: Early Identification, Diagnosis, and Intervention*, 3–34.
- Will E, & Hepburn S (2015). Applied behavior analysis for children with neurogenetic disorders. *International Review of Research in Developmental Disabilities*, 49, 229–259.
- Woolf S, Woolf CM, & Oakland T (2010). Adaptive behavior among adults with intellectual disabilities and its relationship to community independence. *Intellectual and Developmental Disabilities*, 48(3), 209–215. [PubMed: 20597731]
- Yoder P, Woynaroski T, Fey M, & Warren S (2014). Effects of dose frequency of early communication intervention in young children with and without Down syndrome. *American Journal on Intellectual and Developmental Disabilities*, 119(1), 17–32. [PubMed: 24450319]

FIGURE 1.
Age-by-group trends in adaptive behavior

TABLE 1

Participant descriptives by group and age

	TD (<i>n</i> = 30)		(TD vs. DS)		DS (<i>n</i> = 26)		(DS vs. FXS)		FXS (<i>n</i> = 30)	
	<i>M</i>	<i>SD</i>	<i>d</i>		<i>M</i>	<i>SD</i>	<i>d</i>		<i>M</i>	<i>SD</i>
5–12 months										
CA ^a	9.63	1.10			9.23	2.03			9.57	1.48
Male	22	76%	–		14	54%	–		30	100%
MA ^b	11.17	2.21	1.74		7.04	2.52	–0.08		7.22	1.92
Communication DSS ^c	100.03	12.83	1.48		77.92	16.78	–0.45		85.60	17.29
Daily living DSS	97.77	11.60	1.42		80.31	13.00	–0.77		90.80	14.10
Socialization DSS	101.30	8.01	1.68		85.38	10.77	–0.52		91.37	12.31
Motor DSS	96.23	9.88	2.73		67.65	11.02	–1.22		81.63	11.83
Adaptive composite DSS	95.60	15.70	1.55		74.81	10.75	–0.82		84.67	13.11
	TD (<i>n</i> = 18)		(TD vs. DS)		DS (<i>n</i> = 19)		(DS vs. FXS)		FXS (<i>n</i> = 13)	
	<i>M</i>	<i>SD</i>	<i>d</i>		<i>M</i>	<i>SD</i>	<i>d</i>		<i>M</i>	<i>SD</i>
13–24 months										
CA	19.39	3.84			19.00	4.30			19.46	4.20
Gender	12	67%	–		12	63%	–		13	100%
MA	20.72	4.23	1.87		13.61	3.34	0.56		11.77	3.20
Communication DSS	95.61	8.15	2.36		80.26	4.30	1.00		73.31	8.88
Daily living DSS	93.11	9.14	1.15		81.74	10.52	0.78		73.69	10.10
Socialization DSS	98.50	7.29	1.24		87.11	10.81	0.54		81.46	9.96
Motor DSS	98.06	6.91	2.89		74.26	9.36	–0.27		76.92	10.21
Adaptive composite DSS	94.94	7.71	2.18		77.95	7.86	0.76		71.77	8.25
	TD (<i>n</i> = 21)		(TD vs. DS)		DS (<i>n</i> = 19)		(DS vs. FXS)		FXS (<i>n</i> = 26)	
	<i>M</i>	<i>SD</i>	<i>d</i>		<i>M</i>	<i>SD</i>	<i>d</i>		<i>M</i>	<i>SD</i>
25–45 months										
CA	33.67	2.90			31.74	6.67			31.96	5.80
Male	19	91%	–		16	84%	–		26	100%
MA	31.76	2.90	4.00		18.53	3.67	0.20		17.73	4.44
Communication DSS	106.90	10.08	3.66		73.10	8.30	0.59		67.85	9.36
Daily living DSS	90.67	6.86	2.17		73.79	8.63	0.96		66.08	7.33
Socialization DSS	98.71	10.35	2.05		79.84	7.93	0.98		71.50	9.00

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Motor DSS	95.10	11.92	2.59	68.79	8.04	0.20	67.04	9.62
Adaptive composite DSS	96.52	8.42	3.49	69.21	7.19	0.79	63.31	7.95

^aCA, chronological age.
^bMA, nonverbal mental age.
^cDSS, Vineland adaptive behavior scales domain standard score.

TABLE 2

Multiple regression results

	Communication			Daily living			Socialization			Motor skills			ABC		
	<i>b</i>	SE(<i>b</i>)	<i>p</i>	<i>b</i>	SE(<i>b</i>)	<i>p</i>	<i>b</i>	SE(<i>b</i>)	<i>p</i>	<i>b</i>	SE(<i>b</i>)	<i>p</i>	<i>b</i>	SE(<i>b</i>)	<i>p</i>
Intercept	75.69	1.74	0.000	77.42	1.52	0.000	82.75	1.34	0.000	70.19	1.43	0.000	72.61	1.31	0.000
FXS	-2.14	2.37	0.368	-3.36	20.60	0.104	-4.14	1.82	0.023	2.48	1.94	0.202	-2.10	1.78	0.240
TD	26.60	2.40	0.000	15.71	2.08	0.000	16.55	1.83	0.000	26.04	1.95	0.000	24.02	1.80	0.000
Age	-0.29	0.15	0.060	-0.27	0.13	0.045	-0.29	0.12	0.013	0.05	0.12	0.714	-0.28	0.11	0.014
FXS×Age	-0.45	0.21	0.029	-0.75	0.18	0.000	-0.54	0.16	0.001	-0.67	0.17	0.000	-0.61	0.15	0.000
TD×Age	0.58	0.21	0.006	-0.02	0.18	0.932	0.18	0.16	0.257	-0.08	0.17	0.659	0.23	0.16	0.167
	$R^2 = 0.51$			$R^2 = 0.48$			$R^2 = 0.52$			$R^2 = 0.59$			$R^2 = 0.64$		