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Influence of Siblings on Adaptive Behavior Trajectories in Autism Spectrum Disorder

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Influence of Siblings on Adaptive Behavior Trajectories in Autism Spectrum Disorder

A thesis submitted in partial satisfaction of the requirements

for the degree Master of Arts in Education

by

Nicole Elizabeth Rosen

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ABSTRACT OF THE THESIS

Influence of Siblings on Adaptive Behavior Trajectories

in Autism Spectrum Disorder

by

Nicole Elizabeth Rosen

Master of Arts in Education

University of California, Los Angeles, 2021

Professor Catherine Lord, Chair

Siblings play an important role in shaping the developmental trajectories of individuals with autism spectrum disorder (ASD). Having siblings has been associated with better social communication, non-verbal communication, and theory of mind abilities in ASD. However, little is known about the impact of siblings on adaptive skill growth over time, even though adaptive behavior competencies are among the strongest predictors of positive outcomes in ASD. This study examined the influence of sibling constellation factors, including the presence of siblings, position in birth order, gender of closest-age sibling, and gender match of sibling dyad on the adaptive behavior trajectories of individuals with ASD and non-spectrum disorders from ages 9-26 years. Participants with one or more siblings, regardless of birth order position, experienced faster growth rates in adaptive behavior than participants without siblings. Additional benefits

were noted when participants' closest-age siblings were male and when participants were gender-matched with their closest-age siblings.

The thesis of Nicole Elizabeth Rosen is approved.

Connie L. Kasari

Jeffrey J. Wood

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University of California, Los Angeles

2021

TABLE OF CONTENTS

LIST OF FIGURES

LIST OF APPENDICES

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Introduction

The sibling relationship is considered one of the most transformative and meaningful relationships that an individual may have (Cicirelli, 1995). Its unique lifetime duration (Cicirelli, 1982) makes it especially important for dyads involving an individual with ASD, for whom deficits in adaptive functioning persist across the lifespan, often necessitating ongoing support from families. Adaptive functioning, or the ability to successfully meet age-appropriate demands in everyday life, is an area that is particularly impaired in ASD, in comparison to other disabilities (Lord et al., 2020; Rodrigue et al., 1991), and that has robust impacts on adult outcomes (Farley et al., 2009; Tillmann et al., 2019). An encouraging note is that adaptive skills may be malleable through the inclusion of proper supports (Freeman et al., 1999; Howlin et al., 2000; McGovern & Sigman, 2005). Thus, given the common accessibility of siblings during childhood and adolescence, siblings may represent a key medium through which individuals with ASD could gain competence leading to healthy, happy lives (Autistica, 2020).

Siblings have the potential to have a significant impact on the development of individuals with ASD (Knott et al., 2007) through various roles within the family, including play companion, nurturer, conversation partner, teacher, support system, caregiver, and lifelong friend, among many others (Olivia & Arranz, 2005; Orsmond & Seltzer, 2007). Time diary studies of typically developing and ASD sibling dyads have shown that siblings spend a meaningful amount of time together during non-school hours each day (Knott et al., 1995; McGovern & Sigman, 2005; McHale et al., 2012; Orsmond & Kuo, 2011). Siblings, particularly when included in interventions as partners, have been shown to effectively facilitate improvements for their siblings with ASD in social engagement and joint attention (Tsao & Odom, 2006; Walton & Ingersoll, 2012), interest and cooperation in play (Celiberti & Harris, 1993), verbal play behavior

(Coe et al., 1991), social responsiveness (Ferraioli & Harris, 2011), and social imitation (Shivers & Plavnick, 2015; Walton & Ingersoll, 2012). These competencies have been shown to generalize to more naturalistic settings (Celiberti & Harris, 1993; Schreibman et al., 1983) to promote continued improvements in overall social communication (Ferraioli et al., 2012) and peer relationships (Bass & Mulick, 2007).

Adaptive Behavior Trajectories in ASD

Longitudinal analyses of individuals with ASD from childhood through early adulthood reveal that skill development, particularly adaptive skill development, is not static (Freeman et al., 1999; McGovern & Sigman, 2005). Rather, adaptive behavior trajectories in ASD can show improvement over time, particularly when individuals with ASD are engaged in frequent high-level social interactions with peers (McGovern & Sigman, 2005). Given that low levels of peer engagement and poor social communication are common among individuals with ASD (Howlin et al., 2000; Orsmond et al., 2004; Shattuck et al., 2007), individuals with ASD often must rely on their sibling interactions to supplement their development of skills. Notably, research suggests that siblings engage with individuals with ASD in a way that mirrors peer interaction, yet simultaneously provides sufficient scaffolding and support (El-Ghoroury & Romanczyk, 1999). This overlap facilitates the translation of skills from sibling interactions to peer interactions (Ferraioli et al., 2012; Knott et al., 2007; Roskam et al., 2015), which may ultimately foster improvements in adaptive skills over time.

Adaptive functioning is among the strongest contributors to overall social-communication and independent living outcomes among individuals with ASD (Farley et al., 2009; Liss et al., 2001). Therefore, identifying the factors that facilitate the growth of adaptive skills is important in optimizing environments and interventions to most effectively improve outcomes (Tillmann et al., 2019). Across samples of broad ages and cognitive abilities, research suggests that IQ, age, and severity of social-communication deficits are factors that significantly influence adaptive functioning outcomes in ASD (Freeman et al., 1999; McGovern & Sigman, 2005; Szatmari et al., 1989, 2003; Tillmann et al., 2019). Notably absent from this list of factors and from the literature as a whole is the influence of siblings and sibling constellation factors on the adaptive behavior trajectories of individuals with ASD. Thus, the present study seeks to understand these relationships and explore the importance of siblings on adaptive skill development.

Role of Siblings in Adaptive Behavior Development

A review of the limited literature, consisting primarily of cross-sectional and/or small sample studies, reveals that the presence of siblings may be positively associated with stronger skill profiles across each of the three domains that comprise the Vineland adaptive behavior composite in individuals with ASD: socialization (Ben-Itzchak et al., 2016, 2019; El-Ghoroury & Romanczyk, 1999; Knott et al., 1995; Matthews et al., 2013; Matthews & Goldberg, 2018; McGovern & Sigman, 2005), communication (Ben-Itzchak et al., 2016; El-Ghoroury & Romanczyk, 1999; Knott et al., 2007), and daily living skills (Ben-Itzchak et al., 2019). Furthermore, significant positive correlations have been reported between affected siblings' and nonaffected siblings' adaptive behavior domain scores and composite score (Brewton et al., 2012). While there are various possible interpretations of these findings, as the authors note, one implication may be that siblings positively impact the adaptive behavior profiles of individuals with ASD (Brewton et al., 2012). Thus, siblings may provide a built-in social companion, communication partner, and daily living skills role model through which individuals with ASD may develop the necessary competencies to live happy and healthy lives.

Influence of Sibling Constellation Factors

In assessing the role of siblings on the adaptive behavior development of individuals with ASD, it is important to consider various sibling constellation factors, including birth order, gender of the sibling, and gender match of the sibling dyad, that may be influencing the relationship. Related to birth order, the few available studies largely provide support for the positive influence of both younger and older siblings on the adaptive functioning of individuals with ASD, with mixed and non-significant differences reported between the impact of younger versus older siblings (Ben-Itzchak et al., 2019; Brewton et al., 2012). The influence of sibling gender on adaptive skills, specifically in the social domain, of individuals with ASD has also been preliminarily assessed, with no significant effects noted (Ben-Itzchak et al., 2019). Lastly, the gender match of the sibling dyad has not, to our knowledge, been analyzed in relation to adaptive skills. Given the limited existing literature, insight into potential gender and gender match effects may be extrapolated from findings in the related sibling relationship literature. Research largely suggests that female siblings, compared to male siblings, may have the strongest and most positive relationships with their siblings with developmental disabilities (male or female) (Orsmond & Seltzer, 2000). Unlike female siblings, however, the level of involvement of male siblings appears to be gender-dependent, with closer relationships noted between male siblings and gender-matched males with developmental disabilities (Orsmond & Seltzer, 2000; Seltzer et al., 1991).

Various studies have analyzed the role of siblings and the sibling relationship within dyads containing an individual with ASD and an unaffected sibling, yet little is known about how siblings and sibling constellation factors may influence the adaptive behavior trajectories of individuals with ASD. Existing studies on the role of siblings in the development of adaptive skills have largely been limited by the following factors: 1) a focus on the outcomes of the unaffected siblings and the sibling relationship (Brewton et al., 2012; Hastings, 2003; Kaminsky & Dewey, 2002; Macks & Reeve, 2007); 2) an examination of the sibling influence predominantly during childhood (Orsmond & Seltzer, 2007); 3) a relatively narrow scope of outcomes including autism severity, social communication, and theory of mind; 4) a lack of diversity in samples (racial, cognitive ability, and caregiver education levels) (Orsmond & Seltzer, 2007; Orsmond et al., 2009); and 5) an absence of longitudinal studies to assess changes over time (Orsmond et al., 2009). Thus, the present study will be the first to examine longitudinally from childhood through adulthood the influence of siblings, and various sibling constellation factors, on adaptive behavior trajectories among individuals with ASD of diverse backgrounds.

Current Study

Drawing upon an ongoing longitudinal study assessing 253 individuals across 24 years, the present study will be the first to examine longitudinally from childhood through adulthood the influence of siblings, and various sibling constellation factors, on adaptive behavior trajectories from ages 9 to 26 in individuals with ASD diagnoses (139 males, 22 females) and a comparison group of individuals with neurodevelopmental disorders other than ASD (26 males, 21 females) seen at the same time points. The aim is to evaluate the effects of sibling constellation factors (presence of sibling, birth order, gender of closest-age sibling, and gender match of sibling dyad) on growth in adaptive behaviors from childhood through adulthood.

Based on the limited existing sibling literature reviewed above, I hypothesized that individuals with ASD with one or more siblings, controlling for demographic and individual differences such as verbal IQ (VIQ) and autism severity, would show stronger adaptive skills and greater growth in these skills as measured by Vineland Adaptive Behavior Scales (VABS) age equivalents across time than those without siblings. Furthermore, among individuals with ASD with siblings, I hypothesized that, in accordance with existing research, those with female closest-age siblings and those with gender-matched siblings, regardless of birth order position, would demonstrate the steepest growth in adaptive behavior age equivalents from late childhood through adulthood (Ben-Itzchak et al., 2019; Orsmond & Seltzer, 2000).

Methods

Participants

Participants were originally recruited from three sources: a) 192 children under age 3 years referred for possible ASD to two tertiary autism programs (North Carolina and Illinois); b) 21 children under age 3 years with non-ASD developmental delays identified through the referral sources of the first group (North Carolina and Illinois); and c) 40 children with ASD or neurodevelopmental delays also diagnosed at early ages who joined the study at approximately age 9 and then were followed at the same ages as the first two groups (Michigan) (see Anderson et al., 2014). Thus, the initial cohort participated in face to face assessments around ages 2, 3 (ASD referrals only), 5 (North Carolina only), with the full sample seen at ages 9 (*M* years = 9.98, *SD* = 0.89), 19 (*M* = 19.04, *SD* = 1.2), and 26 (*M* = 25.97, *SD* = 1.4), as well as a phone interview at around age 14 (*M* = 14.22, *SD* = 0.41) and biannual packets of questionnaires throughout all this time.

Of the original 253 participants, 208 were selected for the current study based on their completion of at least one face to face assessment at approximately age 9 or older. A substantial majority completed more than one face to face assessment (83.2% completed two or more and 67.3% completed three or more), with a mean number of over three VABS available per participant. Attrition patterns in our sample were similar to those of previous studies, such that attrition was higher among Black families (p = .001), but not associated with gender, recruitment site, diagnosis, caregiver education, VIQ, or ADOS calibrated severity score (see Anderson et al., 2014; Lord et al., 2006; McCauley et al., 2020). Among the 208 participants in the current sample, Black participants accounted for 23.1% with the remainder White. The sample was predominantly male (79.3%) with 50% from North Carolina, 32.2% from Illinois, and 17.8% from Michigan. Approximately half of the sample reported a caregiver education level of at least a four-year college degree (49.5%). 22.6% of the sample had never received a formal diagnosis of ASD throughout the course of the longitudinal study despite repeated blinded assessments. These participants are included in the current study because they show similar patterns in presentation and outcome across development to the participants with ASD (see Lord et al., 2020; McCauley et al., 2020).

Among the 208 participants in the current study, 160 (76.9%) reported having at least one sibling (including full biological, half, step, and adopted siblings) with whom they lived during childhood. There were approximately even splits between the gender (male or female) of the closest-age sibling (50% male) and the gender match (gender-matched or non-gender-matched) of the proband and closest-age sibling (48.1% gender-matched). Over 98% of the 160 probands with at least one sibling reported age differences of fewer than 9 years from their closest-age sibling. However, the majority had narrower age gaps (86.9% were within five years, 48.8% were within three years, and 37.5% were within two years). Thus, given approximately all (98%) probands in the sample with one or more siblings were living with their closest-age sibling by age 9, coupled with the significance of age 9 as the first assessment timepoint that included the full sample (Michigan participants joined the study at age 9), the decision was made to start data analysis at the 9-year-old visit. Related to birth order, 51.9% of probands were youngest children, 22.5% were middle children, and 25.6% were oldest children. The number of siblings reported per proband included the following breakdown: 48 (23.1%) had no siblings, 87 (41.8%) had one sibling, 39 (18.8%) had two siblings, 26 (12.5%) had three siblings, 3 (1.4%) had four siblings, and 5 (2.4%) had five siblings. The 160 probands with at least one sibling did not differ from the larger sample of 208 in race (19.4% Black), gender (79.4% male), recruitment site (44.4% referred from North Carolina, 35.6% from Illinois, and 20% from Michigan), caregiver education (57.5% had at least a four-year college degree), or ASD diagnosis (20.6% never received a formal ASD diagnosis). The 48 probands without siblings also did not differ from the larger sample of 208 in race, gender, recruitment site, and ASD diagnosis. However, samples differed in caregiver education level, with fewer caregivers of probands with no siblings (22.9%) having at least a four-year college degree (p = .005). Complete demographic and individual descriptive data are provided in Appendix 1.

Procedures

Various diagnostic and psychometric instruments were administered to probands and their parents during in-person assessments and phone interviews. Clinicians conducting the assessments, generally a post-doctoral fellow or licensed clinician and a post-baccalaureate research assistant, were research reliable in the relevant measures and were blind to the probands' previous assessment results. Overall, diagnoses of ASD or other disorders were made by this team and presented to a panel of experienced clinicians who reviewed all information and, with this team, reached consensus diagnoses of ASD and other conditions. In-person assessments typically included proband diagnostic assessments, parent interviews, and proband cognitive testing. All visits and assessments were provided free of charge; feedback was provided to individuals and families. Informed consent was obtained from all participating families and individuals themselves whenever possible. This research was approved by IRBs at various institutions across the duration of the study.

Measures

Autism Severity

At each in-person assessment at approximately ages 9, 19, and 26 years, participants were administered the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000). The ADOS yields a calibrated severity score (CSS; Gotham et al., 2009), which can be used to compare ASD symptom severity across individuals of different developmental levels. Proband CSS at age 9 (if unavailable, from later years) was included in the model as a covariate representing autism symptom severity.

Cognitive Abilities

Cognitive assessments were administered at each face to face assessment. The instrument used to obtain VIQ scores at age 9 was chosen from a standard hierarchy including the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999), Wechsler Intelligence Scale for Children (WISC-III; Wechsler, 1991), and Differential Abilities Scale (DAS; Elliott, 1990, 2007). Ratio VIQs were calculated from age equivalents when raw scores fell outside deviation score ranges. Proband VIQ at age 9 (if unavailable, from later years) was included in the model as a covariate.

Demographic and Sibling Information

Parents completed several questionnaires over the years about proband demographics and about the sibling constellation including the number of siblings and the age, gender, and ASD diagnostic history of the proband's closest-age sibling, revealing a 13.1% prevalence rate of sibling ASD co-occurrence.

Adaptive Behavior

The Vineland Adaptive Behavior Scales (VABS; Sparrow et al., 1984, 2005), a standardized, semi-structured caregiver interview of adaptive functioning, was administered at all face to face assessments and at the 14-year-old phone interview. Age equivalent scores, which represent an approximation of the chronological age of typical development based on an individual's abilities, were produced for each domain by averaging the age equivalent scores of the subdomains (Bal et al., 2015; Yang et al., 2016). For this study, domain age equivalent scores were averaged to produce a VABS adaptive behavior composite age equivalent score (VABS-AE). Previous research has suggested that age equivalents, as opposed to composite scores, are more appropriate for samples consisting of participants with intellectual impairment (in our case, 60.6%) because standard composite scores are subject to basal effects, which can obscure differences between adaptive domains (Carter et al., 1998).

Analytic Plan

Preliminary Analyses

Sibling constellation factors were coded into four variables for analyses: a) dichotomous variable representing the presence of siblings (yes or no); b) trichotomous variable representing the proband's position in the sibling birth order, comparing probands with only older siblings (youngest children), probands with younger and older siblings (middle children), and probands with only younger siblings (oldest children); c) dichotomous variable representing the gender of the closest-age sibling, comparing probands with a male closest-age sibling to probands with a female closest-age sibling; and d) dichotomous variable representing the gender match of the closest-age sibling and the proband, comparing gender-matched sibling dyads to non-gender-matched sibling dyads.

Analyses of group differences in sibling constellations based on demographic and individual descriptive factors are provided in Appendix 1. As seen in Appendix 1, the presence of a sibling factor differed in race, caregiver education, site, and VIQ. Within the birth order factor, participants varied in site, number of children in the family, and CSS. Related to the gender of the closest-age sibling, participants differed in site and VIQ. Participants did not significantly differ within the sibling dyad gender match factor. Sibling constellation descriptive information separated by race is provided in Appendix 2. As described below, demographic and individual descriptive factors were addressed and controlled in further analyses.

Primary Analyses

Change in VABS-AEs from late childhood into adulthood was examined using multilevel models via the MIXED procedure in Stata version 16. First, a null model with no predictors was used to test whether random effects capturing between-participant and between-recruitment site were appropriate (Luke, 2020). Second, an unconditional growth model was developed to examine the rate of change of VABS-AEs as a function of participant age as a fixed effect. Linear and quadratic models of age were compared using log-likelihood values. Third, I tested whether allowing age slopes to vary between recruitment sites and between individuals improved model fit. Fourth, the demographic and individual descriptive covariates including gender, race, caregiver education, VIQ, CSS, and history of ASD of the closest-age sibling were tested to examine whether they interacted with age in the baseline model.

The factors were tested individually, comparing the new model to the original baseline model. If a factor significantly interacted with age, it was included as a covariate in the full baseline model. Finally, sibling constellation factors were entered as interaction terms to test the hypotheses. From the full baseline model, a series of four mixed models were developed to investigate whether the presence of a sibling in the family, the position of the proband in the birth order, the gender of the closest-age sibling, and the gender match of the sibling dyad interacted with the rate of change in proband VABS-AEs. These factors and their interaction with age were entered into separate models for each constellation factor. A likelihood ratio test was used to examine the goodness of fit of the models. Post hoc Scheffe's tests were used for slope contrasts on sibling constellation factors. Missing data across time was assumed to be missing at random and estimated using restricted maximum likelihood estimation.

Results

Primary Baseline Model Analyses

The null model without predictors revealed that significant variation in VABS-AEs could be attributed to recruitment site differences ($ICC = 0.25$) nested in between-participant differences ($ICC = 0.44$). Linear and quadratic models of age as a fixed effect were then tested, revealing no significant improvement in fit through the addition of the quadratic component ($p = .27$); thus, the parsimonious linear model was selected. Allowing the slope to vary between recruitment sites significantly improved model fit ($p < 0.001$).

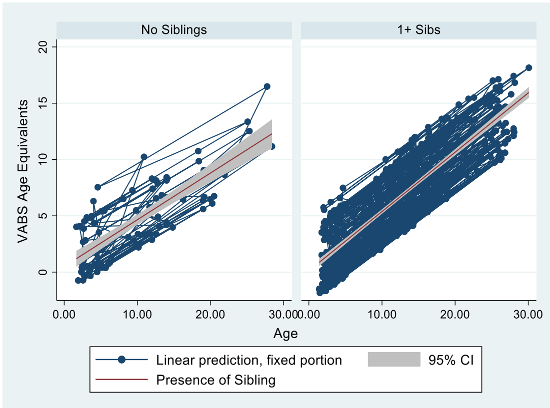
The baseline model showed a positive trajectory in VABS-AEs from late childhood into adulthood as a function of age. On average, participants grew approximately 6 months in VABS-AEs every year ($p < .001$), although there was significant variability in the growth rates. Demographic and individual descriptive factors were then individually added to the model to assess their contributions to the model. Neither participant gender ($p = .17$), caregiver education ($p = .54$), nor closest-age sibling's history of ASD ($p = .52$) significantly interacted with age or contributed to explaining the variation in proband VABS-AEs beyond age. Participant race ($p = .003$), VIQ ($p < .001$), and CSS ($p < .001$) all had significant interactions with age and were retained as covariates in the full sibling constellation models.

Primary Sibling Constellation Models

Developmental trajectories of VABS-AEs were characterized for the following four sets of group contrasts: 1) only child vs. at least one sibling; 2) youngest vs. middle vs. oldest child; 3) female vs. male closest-age sibling; and 4) gender-matched vs. non-gender-matched sibling dyad. Given VABS-AEs were used, the changes in the trajectories within each group reflect the rate of yearly change in adaptive functioning from ages 9 to 26. Each sibling constellation was first tested in a two-way interaction between sibling constellation factor and age to examine potential differential growth patterns while including VIQ, CSS, and race as covariates. Subsequent post-hoc interaction analyses revealed an unexpected three-way interaction between various sibling constellation factors, age, and race; thus, the three-way interaction with VIQ and CSS included as covariates was further explored.

Presence of a Sibling

The model examining the presence of a sibling compared to no siblings resulted in a main effect, such that participants with siblings presented with higher VABS-AEs across all times and experienced significantly steeper growth trajectories through adulthood ($B = .49$; $SE = .02$) compared to those with no siblings ($B = .39$; $SE = .04$; $p = .005$; see Figure 1a). The magnitude of this effect is evident upon assessing the increasing discrepancy in VABS-AEs in years between participants with and without siblings from ages 9 (0.75 years) to 14 (1.25 years) to 19 (1.76 years) to 26 (2.36 years; see Appendix 3).

a) 

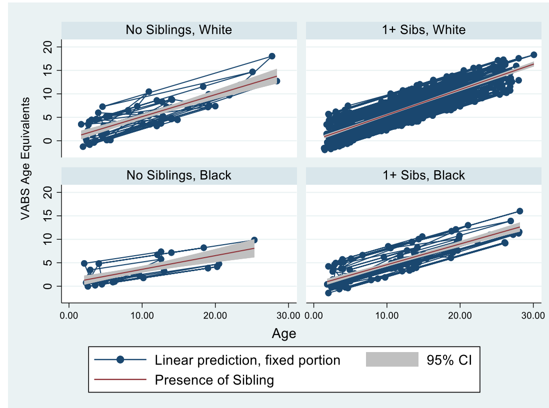
b) 

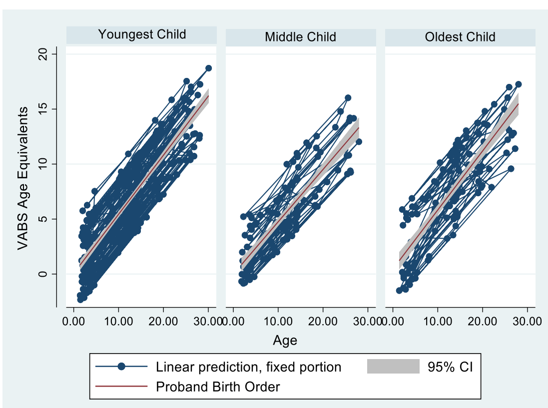
Figure 1. Trajectories of VABS-AEs by a) presence of a sibling and by b) presence of a sibling + race.

Analyses also revealed a significant three-way interaction in the model between the presence of a sibling, age, and race ($p = .02$). Black participants without siblings ($B = .24$; $SE = .06$) had significantly slower growth on VABS-AEs compared to Black participants with siblings ($B = .45$; $SE = .04$; $p = .001$), White participants with siblings ($B = .49$; $SE = .02$; $p < .001$), and White participants without siblings ($B = .46$; $SE = .04$; $p = .001$; see Figure 1b). There were no significant slope differences between Black participants with siblings and White participants with and without siblings.

While White and Black participants with siblings similarly demonstrated steeper growth on VABS-AEs compared to those without siblings, the magnitude of the difference over time was greater among Black participants. Whereas the discrepancy in age equivalents in years between White participants with and without siblings increased slightly (nonsignificant) from ages 9 (0.37 years) to 26 (0.84 years), the discrepancy in age equivalents in years between Black participants with and without siblings widened significantly from ages 9 (1.2 years) to 26 (4.53 years; see Appendix 3). Thus, differences in the rate of adaptive growth between participants with and without siblings were more pronounced among Black compared to White participants.

Position in Birth Order

The model investigating the influence of birth order position on adaptive skill growth trajectories revealed no significant differences between youngest (*B* = .51; *SE* = .03), middle (*B* = .44; *SE* = .04), and oldest children (*B* = .47; *SE* = .04; *p* = .078; see Figure 2a). Analyses revealed a significant three-way interaction between birth order position, age, and race (*p* = .008), though no significant specific contrasts were found after adjusting for multiple comparisons. More specifically, there were no significant differences between White youngest (*B* = .50; *SE* = .03), middle (*B* = .48; *SE* = .04), and oldest child probands (*B* = .50; *SE* = .04), or between Black youngest (*B* = .54; *SE* = .05), middle (*B* = .26; *SE* = .08), and oldest child probands (*B* = .37; *SE* = .06) on growth of VABS-AEs (see Figure 2b).

a)


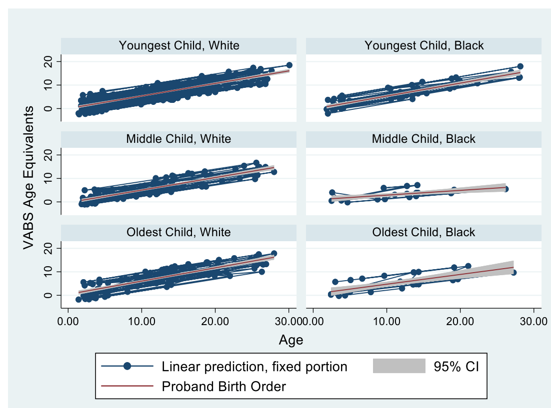
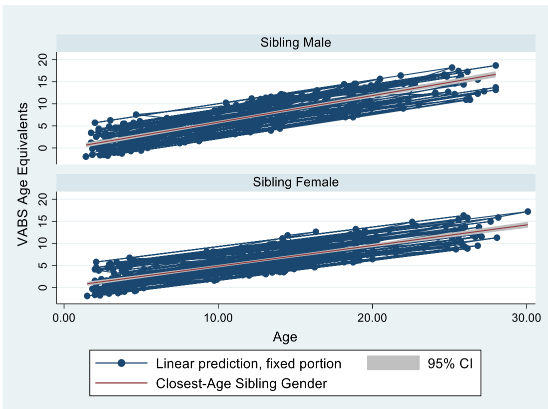
b)


Figure 2. Trajectories of VABS-AEs by a) proband position in birth order and by b) proband position in birth order + race.

Gender of Closest-Age Sibling

The model examining the impact of the gender of the closest-age sibling on growth in adaptive skills revealed significantly steeper growth among participants with a male (*B* = .54; *SE* = .03) compared to a female (*B* = .44; *SE* = .03) closest-age sibling (*p* = .001; see Figure 3a). Analyses of increasing discrepancies in VABS-AEs between those with a male versus female closest-age sibling at each age further illustrate this pattern (age 9: 0.86 years; age 14: 1.35 years; age 19: 1.83 years; age 26: 2.42 years), revealing that participants with a male closest-age sibling presented with greater adaptive skills and experienced faster rates of growth over time than participants with a female closest-age sibling (see Appendix 5).

a) 

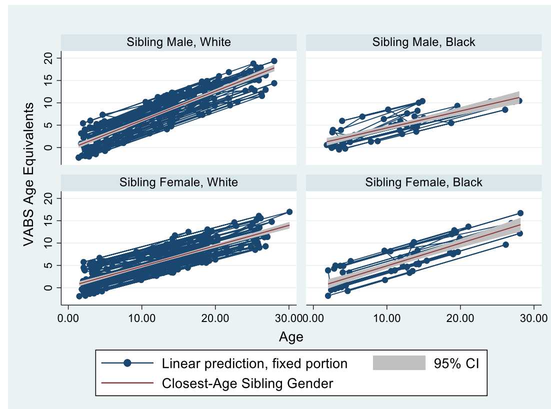
b) 

Figure 3. Trajectories of VABS-AEs by a) gender of closest-age sibling and by b) gender of closest-age sibling + race.

Results again demonstrated a significant three-way interaction between the gender of the closest-age sibling, proband age, and proband race on growth in VABS-AEs (p = .002) from late childhood through adulthood. While no significant within-race differences were found between White participants with a male (B = .58; SE = .03) versus a female closest-age sibling (B = .43; SE = .03) and between Black participants with a male (B = .41; SE = .05) versus a female closest-age sibling (B = .48; SE = .05), a significant across-race difference emerged between White and Black participants with a male closest-age sibling suggesting greater growth among White participants (p = .04; see Figure 3b). Increasing discrepancies in VABS-AEs between White and Black participants with a male closest-age sibling from ages 9 (1.39 years) to 14 (2.24 years) to 19 (3.1 years) to 26 (4.13 years) demonstrate this growing effect over time (see Appendix 5).

Gender Match of Sibling Dyad

The model revealed a significant interaction between participant age and gender match of the participant and closest-age sibling (p = .023), such that participants with a gender-matched sibling (B = .52; SE = .03) demonstrated significantly steeper adaptive skill growth trajectories than participants with a non-gender-matched sibling (B = .46; SE = .03; see Figure 4). The increasing discrepancies in VABS-AEs between gender-matched and non-gender-matched participants from ages 9 (0.66 years) to 14 (1 year) to 19 (1.34 years) to 26 (1.75 years) further elucidate this effect (see Appendix 6). The three-way interaction between gender match of the sibling dyad, proband age, and proband race was not significant.

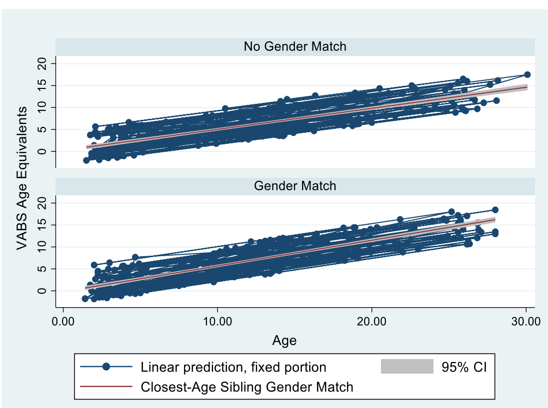


Figure 4. Trajectories of VABS-AEs by gender match of proband and closest-age sibling.

Discussion

This study was the first to examine the influence of siblings and various sibling constellation factors on adaptive behavior trajectories among individuals with ASD from late childhood through adulthood. Findings revealed that while all participants showed a positive trajectory of adaptive skill development as a function of age, with an average growth of six months per year, there was significant variability in rates of growth based on group membership within the following sibling constellation factors: presence of a sibling, position in birth order, gender of the closest-age sibling, and gender match (same or different genders) between the proband and the closest-age sibling.

Presence of a Sibling

Participants with siblings experienced significantly steeper adaptive skill growth trajectories from childhood through adulthood compared to participants without siblings, even after controlling for demographic and individual descriptive factors. Findings provide preliminary support for the importance of siblings on adaptive skill development in ASD and are consistent with existing literature detailing the positive influence of siblings on development, specifically in the areas of theory of mind (Matthews et al., 2013; Matthews & Goldberg, 2018; O'Brien et al., 2011), social communication (Ben-Itzchak et al., 2019), and non-verbal communication (Ben-Itzchak et al., 2016).

White and Black participants demonstrated similar patterns of greater growth in adaptive skills among those with one or more siblings compared to those without siblings, though differences in intensity were noted showing a magnified effect among Black participants. These racial differences in VABS-AEs across time should be interpreted with caution given the various confounding variables that likely contribute to the findings. Upon analyzing participants based on within-race demographic differences, both White and Black participants with no siblings were significantly more likely to have caregivers with lower levels of education (p = .002; p = .038) and to live in single-caregiver homes (p = .001; p = .005) compared to participants with one or more siblings (see Appendix 2), with Black participants with no siblings having significantly lower caregiver education (p = .038) and a higher proportion of single-caregiver homes (p = .002) compared to White participants with no siblings. Furthermore, although I controlled for VIQ in analyses, it is worth noting that participants with no siblings had significantly lower VIQs across both races than participants with one or more siblings (p = .018), and Black participants with no siblings had significantly lower VIQs than White participants with no siblings (p = .03). Given that these children were primarily recruited at age 2 through early diagnoses of autism, and knowing that children with higher VIQs and children from Black families for many years received diagnoses later than children with lower VIQs and children from White families, it is possible that the observed differences were related to recruitment effects (Maenner et al., 2020). The substantial number of both Black and White families in our sample allowed for these comparisons, though replication with larger, more diverse samples will be important.

Position in Birth Order

Our finding of no effects of position in the birth order among participants with siblings is interesting, given the existing literature on sibling interactions shows that, irrespective of birth order position, role asymmetries develop across time for sibling dyads involving an individual with developmental disabilities (Stoneman et al., 1989). Across birth order positions, typically developing siblings may adopt the older sibling “teacher” role while individuals with developmental disabilities adopt the younger sibling “learner” role (Knott et al., 1995; Orsmond & Seltzer, 2000; Stoneman et al., 1989). The similar improvement in adaptive skills among individuals with ASD across birth order positions may also reflect unique benefits derived from both younger and older positions in dyads. For example, individuals with ASD with older siblings may benefit from more mature, scaffolded, and overall supportive interactions, while individuals with ASD with younger siblings may experience growth through interacting with siblings who more closely match their mentalizing abilities (at least for a short time), which may allow for naturalistic interactions without significant scaffolding that are more representative of peer interactions (Matthews et al., 2013).

Despite a significant three-way interaction with race and large slope differences, no significant contrasts were found after adjusting for multiple comparisons; results showed consistent unexplained error that could not be attributed to variables included in the present analyses. Given the paucity of research assessing the impact of race and birth order position on adaptive functioning in ASD, the current findings emphasize the need for future research to analyze the influence of birth order, specifically among Black participants with multiple children in the family, on adaptive skill trajectories in ASD.

Gender of Closest-Age Sibling

Given the lack of research on the influence of the gender of the closest-age sibling on adaptive skill trajectories in ASD, results identifying a significant advantage of having a male compared to a female closest-age sibling should be interpreted as preliminary, requiring replication. Though the magnitude of the yearly growth difference may initially appear small (1.2 more months of growth per year), the practical significance is large, equating to almost 3 years (33.6 months) more growth in adaptive skills across the 28-year study. While the connection between quality of sibling relationships and adaptive skill development in ASD is unknown, findings from the current study appear to run counter to the female sibling advantage observed in the sibling relationship literature related to caregiving, companionship, and positive affect (Orsmond & Seltzer, 2000; Seltzer et al., 1991), though Orsmond and Seltzer (2007) importantly note additional factors including greater similarity among siblings (i.e., in education level, physical proximity, functional abilities) that may drive sibling closeness and potentially confound gender influence. Future studies are therefore needed to replicate current findings, assess the connection between sibling relationship and growth in adaptive skills, and analyze the role of sibling similarity on adaptive functioning trajectories.

The influence of race requires further examination. The present study noted significantly greater growth in adaptive skills among White participants with a male closest-age sibling compared to Black participants with a male closest-age sibling. Notably, while not significant, within-race differences were found that suggest a slight advantage of having a male compared to a female closest-age sibling among White participants, and the opposite slight advantage of having a female compared to a male closest-age sibling among Black participants. These differences in race are not easily understood and have not previously been explored in research, and thus require further examination with larger samples across races to better understand these patterns.

Gender Match of Sibling Dyad

Findings of significantly faster adaptive skill growth among participants with a same-gender compared to a different-gender closest-age sibling are consistent with existing sibling relationship literature favoring same-gender sibling dyads. Specifically, previous research involving individuals with developmental disabilities found that mothers reported more warmth in the sibling relationship among same-gender sibling dyads (Begum & Blacher, 2011). The gender match advantage has also been observed in the typical development literature, revealing that same-gender dyads experience greater social learning through modeling than different-gender dyads (McHale et al., 2012).

Further research on the gender match of the sibling dyad and the gender of the closest-age sibling is required. An interesting pattern was noted by Orsmond and Seltzer (2000), such that while sisters show patterns of high involvement in the sibling relationship regardless of the gender of the individual with disabilities, brothers' level of involvement appears to be gender match dependent. Specifically, brothers with male siblings with developmental disabilities showed greater involvement in and reported more positive feelings about the sibling relationship than brothers with female siblings with developmental disabilities (Orsmond & Seltzer, 2000). Given the small number of female compared to male probands in the current study, future research should replicate findings using larger samples of females with ASD to better understand the generalizability of the gender match findings.

Limitations and Implications for Future Research

This study has several limitations, some of which have been described previously. Characteristic of most longitudinal studies, attrition has affected the sample across the 28-year study, with increased participant dropout noted among Black participants. Further, the sample described here is relatively small given the number of analyses, with fewer Black compared to White participants. The representativeness of the diversity in the sample is limited with only White and Black participants, and observed racial differences likely reflect differences in a host of variables that are nested within race in our study including maternal education (our measure of social class), marital status, and age of parent (on the birth order factor). Thus, it will be important for future studies to include sufficient samples of participants from different backgrounds to better disentangle race from other confounds. Lastly, participants were primarily referred for diagnostic evaluations 28 years ago and may not represent referrals and amount of early intervention available today in the U.S.

Future studies are needed to replicate the present findings in diverse racial and ethnic groups, as well as to explore the influence of sibling constellation factors on the specific domains that comprise adaptive behavior to further understand the relationship in ASD. Additionally, given the importance of siblings on adaptive functioning, more research is needed to assess if there are specific qualities of siblings (e.g., adaptive, social, and/or emotional functioning) or sibling relationships that drive improvement in adaptive functioning in ASD. Furthermore, while our study included siblings closest in age to probands in analyses because age differences have been shown to impact the frequency of sibling contact (Ben-Itzchak et al., 2019; Tomeny et al., 2012), future research should replicate the study using other siblings (non-closest-age) in the family. Lastly, future work should assess the impact of siblings on the adaptive functioning profiles of individuals with other non-ASD developmental disorders.

Conclusion

This longitudinal study aimed to assess the influence of sibling constellation factors on adaptive skill trajectories from late childhood through adulthood among individuals with ASD. Results highlight the importance of siblings on development in ASD; participants with siblings, regardless of birth order position, experienced significantly greater rates of adaptive skill growth than participants without siblings. Additional benefits were noted when closest-age siblings were male and when probands were the same gender as their closest-age siblings. Our study also revealed potential race effects; findings were largely similar for White and Black participants, though there were differences in the size of effects which require replication. This study may be helpful for family planning decisions because many families wonder about the impact of having multiple children on the development of the child with ASD given the recurrence risk of ASD in siblings; this study suggests that having a sibling, regardless of birth order position and ASD diagnosis, has positive effects on adaptive behavior development in ASD. Findings may also inform intervention planning, when it is appropriate for siblings to be involved, to ultimately maximize adaptive skill development and optimize long-term outcomes among individuals with ASD.

Appendices

Appendix 1. Sibling constellation descriptives and group difference analyses.

	Presence of a Sibling			Proband Position in Birth Order				Closest-Age Sibling Gender			Sibling Dyad (Proband + Closest-Age Sibling) Gender Match		
	No Sibling	1+ Sibling	χ2 / *F*	Youngest Child	Middle Child	Oldest Child	χ2 / *F*	Male Sibling	Female Sibling	χ2 / *F*	No Gender Match	Gender Match	χ2 / *F*
N	48	160	---	83	36	41	---	80	80	---	83	77	---
Male *N* (%)	38 (79.2%)	127 (79.4%)	0.01	65 (78.3%)	29 (80.6%)	33 (80.5%)	0.12	62 (77.5%)	65 (81.3%)	0.34	65 (78.3%)	62 (80.5%)	0.12
White *N* (%)	31 (64.6%)	129 (80.6%)	5.35*	67 (80.7%)	29 (80.6%)	33 (80.5%)	0.01	62 (77.5%)	67 (83.8%)	1	69 (83.1%)	60 (77.9%)	0.69
Caregiver Education - College Degree+ *N* (%)	11 (22.9%)	92 (57.5%)	17.67***	48 (57.8%)	18 (50%)	26 (63.4%)	1.42	47 (58.8%)	45 (56.3%)	0.1	48 (57.8%)	44 (57.1%)	0.01
Site North Carolina *N* (%)	33 (68.8%)	71 (44.4%)	8.8*	42 (50.6%)	14 (38.9%)	15 (36.6%)	18.53***	34 (42.5%)	37 (46.3%)	6.05*	32 (38.6%)	39 (50.6%)	3.44
Proband ASD dx *N* (%)	34 (70.8%)	127 (79.4%)	1.54	64 (77.1%)	33 (91.7%)	30 (73.2%)	4.55	60 (75%)	67 (83.8%)	1.87	65 (78.3%)	62 (80.5%)	0.12
Sibling ASD dx *N* (%)	---	21 (13.1%)	---	11 (13.3%)	2 (5.6%)	8 (19.5%)	3.28	14 (17.5%)	7 (8.8%)	2.69	10 (12%)	11 (14.3%)	0.18
VIQ *M (SD)*	47.31 (36.52)	61.99 (37.67)	5.68*	64.02 (36.93)	51.11 (33.06)	67.41 (41.76)	2.08	68.31 (39.2)	55.66 (35.2)	4.61*	58.16 (36.93)	66.12 (38.27)	1.79
ADOS CSS *M (SD)*	5.98 (2.85)	5.94 (2.97)	0.01	5.83 (3.03)	6.94 (2.81)	5.27 (2.82)	3.25*	5.75 (3.11)	6.13 (2.83)	0.64	5.84 (3.02)	6.04 (2.93)	0.17
# of Siblings - Includes Proband *M (SD)*	1 (0)	2.75 (1)	---	2.48 (0.86)	3.67 (0.83)	2.49 (0.95)	25.33***	2.74 (0.92)	2.76 (1.08)	0.03	2.67 (0.98)	2.83 (1.03)	0.97

Note: ASD dx: autism spectrum disorder diagnosis (diagnostic history); VIQ: verbal intelligence quotient; ADOS CSS: Autism Diagnostic Observation Schedule calibrated severity score. Significant group differences within each sibling constellation factor are shown with *$p < .05$, **$p < .01$, and ***$p < .001$.

Appendix 2. Racial breakdown of sibling constellation descriptives.

	Presence of a Sibling		Proband Position in Birth Order			Closest-Age Sibling Gender		Sibling Dyad (Proband + Closest-Age Sibling) Gender Match	
	No Sibling	1+ Sibling	Youngest Child	Middle Child	Oldest Child	Male Sibling	Female Sibling	No Gender Match	Gender Match
N	31 17	129 31	67 16	29 7	33 8	62 18	67 13	69 14	60 17
Male *N* (%)	24 (77.4%) 14 (82.4%)	103 (79.8%) 24 (77.4%)	53 (79.1%) 12 (75%)	23 (79.3%) 6 (85.7%)	27 (81.8%) 6 (75%)	48 (77.4%) 14 (77.8%)	55 (82.1%) 10 (76.9%)	55 (79.7%) 10 (71.4%)	48 (80%) 14 (82.4%)
Caregiver Education - College Degree+ *N* (%)	10 (32.3%) 1 (5.9%)	82 (63.6%) 10 (32.3%)	42 (62.7%) 6 (37.5%)	17 (58.6%) 1 (14.3%)	23 (69.7%) 3 (37.5%)	41 (66.1%) 6 (33.4%)	41 (61.2%) 4 (30.8%)	41 (59.4%) 7 (50%)	41 (68.4%) 3 (17.7%)
Site - North Carolina *N* (%)	20 (64.5%) 13 (76.5%)	45 (34.9%) 26 (83.9%)	28 (41.8%) 14 (87.5%)	8 (27.6%) 6 (85.7%)	9 (27.3%) 6 (75%)	20 (32.3%) 14 (77.8%)	25 (37.3%) 12 (92.3%)	21 (30.4%) 11 (78.6%)	24 (40%) 15 (88.2%)
Proband ASD dx *N* (%)	22 (71%) 12 (70.6%)	99 (76.7%) 28 (90.3%)	49 (73.1%) 15 (93.8%)	27 (93.1%) 6 (85.7%)	23 (69.7%) 7 (87.5%)	45 (72.6%) 15 (83.3%)	54 (80.6%) 13 (100%)	52 (75.4%) 13 (92.9%)	47 (78.3%) 15 (88.2%)
Sibling ASD dx *N* (%)	---	16 (12.4%) 5 (16.1%)	6 (9%) 5 (31.3%)	2 (6.9%) 0 (0%)	8 (24.2%) 0 (0%)	11 (17.7%) 3 (16.7%)	5 (7.5%) 2 (15.4%)	9 (13%) 1 (7.1%)	7 (11.7%) 4 (23.5%)
VIQ *M (SD)*	55.71 (38.54) 32 (27.32)	65.35 (37.72) 48 (34.65)	65.81 (37.53) 56.56 (34.42)	54.93 (33.54) 35.29 (27.6)	73.58 (40.42) 42 (39.77)	74.18 (38.9) 48.11 (33.98)	57.18 (34.93) 47.85 (36.95)	59.83 (36.96) 49.93 (37)	71.7 (37.89) 46.41 (33.65)
ADOS CSS *M (SD)*	5.84 (2.63) 6.24 (3.27)	5.74 (3.05) 6.77 (2.47)	5.55 (3.17) 7 (2.03)	6.83 (2.84) 7.43 (2.82)	5.15 (2.82) 5.75 (2.96)	5.45 (3.17) 6.78 (2.73)	6 (2.94) 6.77 (2.17)	5.72 (3.12) 6.43 (2.5)	5.75 (3.0) 7.06 (2.49)
# of Siblings - Includes Proband *M (SD)*	1 (0) 1 (0)	2.7 (0.93) 2.97 (1.25)	2.39 (0.65) 2.88 (1.41)	3.76 (0.87) 3.29 (0.49)	2.39 (0.79) 2.88 (1.46)	2.74 (0.96) 2.72 (0.83)	2.66 (0.91) 3.31 (1.65)	2.62 (0.93) 2.93 (1.21)	2.78 (0.94) 3 (1.32)

Note: ASD dx: autism spectrum disorder diagnosis (diagnostic history); VIQ: verbal intelligence quotient; ADOS CSS: Autism Diagnostic Observation Schedule calibrated severity score.

Two data points are listed within each descriptive characteristic of each sibling constellation factor: the top value represents White participants, and the bottom value represents Black participants.

Appendix 3. Mean age equivalents across time by presence of a sibling and by presence of a sibling + race.

Chronological Age	Presence of a Sibling	Mean Age Equivalents	Standard Error
9	No Siblings	4.01	0.25
	White + No Siblings	4.50	0.28
	Black + No Siblings	3.01	0.36
9	1+ Siblings	4.76	0.17
	White + 1+ Siblings	4.87	0.17
	Black + 1+ Siblings	4.21	0.26
12	No Siblings	5.17	0.31
	White + No Siblings	5.90	0.35
	Black + No Siblings	3.72	0.45
12	1+ Siblings	6.22	0.22
	White + 1+ Siblings	6.35	0.21
	Black + 1+ Siblings	5.55	0.31
14	No Siblings	5.94	0.36
	White + No Siblings	6.83	0.41
	Black + No Siblings	4.20	0.53
14	1+ Siblings	7.19	0.25
	White + 1+ Siblings	7.34	0.24
	Black + 1+ Siblings	6.44	0.35
19	No Siblings	7.87	0.52
	White + No Siblings	9.15	0.58
	Black + No Siblings	5.39	0.77
19	1+ Siblings	9.63	0.35
	White + 1+ Siblings	9.81	0.34
	Black + 1+ Siblings	8.67	0.49
21	No Siblings	8.64	0.58
	White + No Siblings	10.08	0.66
	Black + No Siblings	5.86	0.87
21	1+ Siblings	10.60	0.39
	White + 1+ Siblings	10.80	0.38
	Black + 1+ Siblings	9.56	0.56
26	No Siblings	10.19	0.72
	White + No Siblings	11.94	0.81
	Black + No Siblings	6.81	1.08
26	1+ Siblings	12.55	0.47
	White + 1+ Siblings	12.78	0.46
	Black + 1+ Siblings	11.34	0.68

Appendix 4. Mean age equivalents across time by proband position in birth order and by proband position in birth order + race.

Chronological Age	Position in Birth Order	Mean Age Equivalents	Standard Error
9	Youngest Child	4.82	0.21
	White + Youngest Child	4.75	0.27
	Black + Youngest Child	4.85	0.38
9	Middle Child	4.32	0.26
	White + Middle Child	4.52	0.32
	Black + Middle Child	2.91	0.52
9	Oldest Child	5.06	0.26
	White + Oldest Child	5.28	0.32
	Black + Oldest Child	4.12	0.47
12	Youngest Child	6.35	0.26
	White + Youngest Child	6.26	0.30
	Black + Youngest Child	6.47	0.41
12	Middle Child	5.65	0.31
	White + Middle Child	5.97	0.34
	Black + Middle Child	3.69	0.59
12	Oldest Child	6.46	0.29
	White + Oldest Child	6.78	0.33
	Black + Oldest Child	5.24	0.51
14	Youngest Child	7.37	0.30
	White + Youngest Child	7.26	0.32
	Black + Youngest Child	7.55	0.46
14	Middle Child	6.53	0.35
	White + Middle Child	6.93	0.38
	Black + Middle Child	4.21	0.68
14	Oldest Child	7.39	0.34
	White + Oldest Child	7.78	0.36
	Black + Oldest Child	5.98	0.57
19	Youngest Child	9.91	0.41
	White + Youngest Child	9.77	0.41
	Black + Youngest Child	10.24	0.62
19	Middle Child	8.74	0.50
	White + Middle Child	9.33	0.51
	Black + Middle Child	5.52	0.98
19	Oldest Child	9.73	0.47
	White + Oldest Child	10.27	0.48
	Black + Oldest Child	7.84	0.80
21	Youngest Child	10.93	0.46
	White + Youngest Child	10.78	0.45
	Black + Youngest Child	11.32	0.70

21	Middle Child	9.63	0.56
	White + Middle Child	10.29	0.57
	Black + Middle Child	6.05	1.11
21	Oldest Child	10.66	0.53
	White + Oldest Child	11.27	0.54
	Black + Oldest Child	8.59	0.90
26	Youngest Child	12.96	0.56
	White + Youngest Child	12.79	0.54
	Black + Youngest Child	13.47	0.86
26	Middle Child	11.39	0.69
	White + Middle Child	12.22	0.69
	Black + Middle Child	7.10	1.39
26	Oldest Child	12.53	0.66
	White + Oldest Child	13.26	0.67
	Black + Oldest Child	10.07	1.13

Appendix 5. Mean age equivalents across time by gender of closest-age sibling and by gender of closest-age sibling + race.

Chronological Age	**Closest-Age Sibling Gender**	**Mean Age Equivalents**	**Standard Error**
9	Sibling Male	5.15	0.27
	White + Sibling Male	5.41	0.31
	Black + Sibling Male	4.02	0.40
9	Sibling Female	4.29	0.27
	White + Sibling Female	4.21	0.31
	Black + Sibling Female	4.47	0.41
12	Sibling Male	6.77	0.30
	White + Sibling Male	7.14	0.33
	Black + Sibling Male	5.24	0.44
12	Sibling Female	5.62	0.30
	White + Sibling Female	5.51	0.33
	Black + Sibling Female	5.92	0.44
14	Sibling Male	7.85	0.33
	White + Sibling Male	8.29	0.35
	Black + Sibling Male	6.05	0.49
14	Sibling Female	6.50	0.33
	White + Sibling Female	6.38	0.35
	Black + Sibling Female	6.88	0.49
19	Sibling Male	10.54	0.42
	White + Sibling Male	11.18	0.44
	Black + Sibling Male	8.08	0.66
19	Sibling Female	8.71	0.42
	White + Sibling Female	8.55	0.43
	Black + Sibling Female	9.29	0.64
21	Sibling Male	11.62	0.47
	White + Sibling Male	12.33	0.48
	Black + Sibling Male	8.89	0.74
21	Sibling Female	9.59	0.46
	White + Sibling Female	9.41	0.47
	Black + Sibling Female	10.26	0.72
26	Sibling Male	13.78	0.56
	White + Sibling Male	14.64	0.57
	Black + Sibling Male	10.51	0.92
26	Sibling Female	11.36	0.55
	White + Sibling Female	11.15	0.56
	Black + Sibling Female	12.19	0.88

Appendix 6. Mean age equivalents across time by gender match of proband and closest-age sibling.

Chronological Age	**Sibling Dyad Gender Match**	**Mean Age Equivalents**	**Standard Error**
9	No Gender Match	4.40	0.27
	Gender Match	5.06	0.27
12	No Gender Match	5.77	0.31
	Gender Match	6.64	0.31
14	No Gender Match	6.68	0.34
	Gender Match	7.68	0.34
19	No Gender Match	8.96	0.44
	Gender Match	10.30	0.45
21	No Gender Match	9.87	0.48
	Gender Match	11.35	0.49
26	No Gender Match	11.70	0.58
	Gender Match	13.45	0.59

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