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## Early Adaptive Functioning Trajectories in Preschoolers With Autism Spectrum Disorders

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**Early Adaptive Functioning Trajectories In Preschoolers  
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Review

# Early Adaptive Functioning Trajectories In Preschoolers With Autism Spectrum Disorders

## Abstract

**Background** In preschoolers with Autism Spectrum Disorders (ASD) symptom severity has a negative impact on the development of adaptive functioning, with critical consequences on the quality of life of those children. Developmental features such as reduced social interest or the presence of behavioral problems can further impede daily life learning experiences. **Objectives** The first aim of this study is to confirm the negative impact of high symptom severity on adaptive functioning trajectories in preschoolers with ASD. The second objective intends to explore whether reduced social interest and severe behavioral problems negatively affect developmental trajectories of adaptive functioning in young children with ASD. **Methods** Sixty-eight children with ASD and 48 age and gender-matched children with typical development (TD) between 1.6 and 6 years were included in our study and longitudinal data on adaptive functioning were collected (mean length of the longitudinal data collection was 1.4 years  $\pm 0.6$ ). Baseline measures of symptom severity, social interest, and behavioral problems were also obtained. **Results** We confirmed that children with ASD show parallel developmental trajectories but a significantly lower performance of adaptive functioning compared to children with TD. Furthermore, analyses within ASD children demonstrated that those with higher symptom severity, reduced social interest and higher scores of behavioral problems exhibited especially lower or faster declining trajectories of adaptive functioning. **Conclusions** These findings bolster the idea that social interest and behavioral problems are crucial for the early adaptive functioning development of children with autism. The current study has clinical implications in pointing out early intervention targets in children with ASD.

**Key Words:** autism spectrum disorders; developmental trajectories; adaptive functioning

## Introduction

Autism Spectrum Disorders (ASD) is a neurodevelopmental disorder characterized by atypical social interactions and communication, as well as the presence of restricted and repetitive behaviors (DSM–5, 5th ed., American Psychiatric Association, 2013). ASD symptoms can impair an individual's ability to function in everyday life, or adaptively, to varying degrees. This is not surprising given that adaptive functioning requires the implementation of skills that are critical to daily life, including functional communication, autonomy and the ability to get along with people and be independent (Perry, Flanagan, Geier, & Freeman, 2009). During the first years of life, young children acquire a significant number of skills that are key to their adaptive functioning, or their daily autonomy and independence (i.e., the ability to get dressed or the ability to successfully interact with their peers). Accordingly, preschoolers with ASD are frequently already behind on adaptive functioning (e.g., Milne, McDonald, & Comino, 2013), making it a significant predictor of later quality of life in children and adolescents affected by the disorder (Emily & Grace, 2015).

Adaptive skills are widely considered to be an essential intervention goal that should be prioritized as early as possible (Dawson et al., 2010; Bal, Kim, Cheong, & Lord, 2015). However, the varying degrees by which adaptive functioning is affected in individuals with ASD make it essential to explicitly define a child's profile before establishing an intervention plan based on individual strengths and weaknesses (e.g., Mandell, Novak, & Zubritsky, 2005; Perry et al., 2009). It is, therefore, necessary to understand the factors that both underlie and depend on adaptive functioning in children with ASD during the preschool years, which coincide with the period when individuals are frequently diagnosed (Daniels & Mandell, 2014).

Studies on children with ASD have examined trajectories in the context of longitudinal designs to follow the course of variables, such as adaptive functioning, at various developmental stages (Baghdadli et al., 2012; Bal et al., 2015; Szatmari, Georgiades, & Duku,

2015; Smith, Maenner & Seltzer, 2012). Despite the clinical and scientific meaningfulness of delineating developmental trajectories to predict individual outcomes, studies using longitudinal designs and dedicated to the early development are still rare. To the best of our knowledge, Szatmari et al. (2015) is the only study that has addressed distinct patterns of adaptive functioning in preschoolers with autism. In their study, Szatmari et al. (2015) showed that membership to distinct trajectories of adaptive functioning in preschoolers with ASD is influenced by symptom severity, cognitive functioning, age at the diagnosis, gender, and language (e.g., decreased symptom severity predicts membership to a trajectory showing increasing adaptive functioning over time). In the present study, we are interested in exploring whether social interest and behavioral problems in preschoolers with ASD also impact adaptive functioning trajectories, according to previous evidence sustaining their role in affecting the early development (e.g., Franchini et al., 2016; Fulton, Eapen, Črnčec, Walter, & Rogers, 2014).

Social interest is reduced in young children with ASD (for a meta-analysis see Chita-Tegmark, 2016). For example, preschoolers with ASD demonstrate reduced interest for social cues, such as faces, or biological motion (Osterling & Dawson, 1994; Pierce et al., 2015; Franchini et al., 2017). Furthermore, social interest in young children with ASD is an essential precursor to social-communicative learning (for a review see Chevallier, Kohls, Troiani, Brodtkin, & Schultz, 2012), affording it the potential to positively impact cognitive development early in life. When young children with ASD pay less attention to socially relevant cues, their social stimulation and, in turn, their social learning is reduced compared to children with typical development (TD). Social interest has been consistently and positively related to cognitive and adaptive functioning in children with ASD (e.g., Pierce et al., 2015; Shic, Bradshaw, Klin, Scassellati, & Chawarska, 2011; Franchini et al., 2016). Reduced social interest in children with ASD can thus have dramatic consequences on learning from day-to-day experiences (e.g., if a child does not watch another person washing his hands, an everyday

task, it becomes more challenging for the child to imitate the person and learn the skill).

Maladaptive behaviors are clinically significant in one in three children with autism (Hartley, Sikora, & McCoy, 2008), and represent another factor that hinders learning from everyday life in young children with ASD (Bauminger, Solomon, & Rogers, 2010; Tomanik, Harris, & Hawkins, 2009). Maladaptive behaviors can either be internalized (e.g., depression-anxiety, withdrawal) or externalized (e.g., aggressive behaviors, defiance), and may constitute one of the primary obstacles barring individuals with autism from being socially integrated (e.g., a child with aggressive behavior is less likely to be included in a group of peers; also see Grace, Llewellyn, Wedgwood, Fenech, & McConnell, 2008). A recent study (Fulton, Eapen, Črnčec, Walter, & Rogers, 2014) suggests that maladaptive behavior can hinder the impact of early interventions if it is not treated as a separate target, resulting in reduced gain in cognitive and adaptive functioning.

To the best of our knowledge, and despite their importance to daily life experiences and learning, the impact of reduced social interest and behavioral problems on the development of adaptive functioning of preschoolers with ASD has not been explored to date. Given the potential of those variables in thriving the early developmental skills of young children with ASD, a better understanding of their impact on adaptive functioning has the potential to inform about critical targets for early interventions.

Our objectives for the present study were twofold. First, we used data from an accelerated longitudinal design to compare the developmental trajectories of adaptive functioning of a group of preschoolers with ASD with those from a group of preschoolers with TD. Szatmari et al. (2015) found differential adaptive functioning trajectories in preschoolers with ASD according to their symptom severity, but their study lacked a comparison group. Second, we explored the impact of symptom severity, behavioral problems and social interest on the adaptive functioning trajectories of preschoolers with autism, given that all three affect day-to-day learning in young children with ASD. We hypothesize that children with ASD will

demonstrate global reductions (lower group average) and slower developmental trajectories (decreased slope) during the preschool period, compared to children with TD. We also predict that children with autism with less severe symptomatology (Szatmari et al., 2015), fewer maladaptive behaviors or increased social interest will demonstrate higher functional scores and faster adaptive functioning development (increased slope) than children with ASD with greater symptom severity, more maladaptive behaviors or decreased social interest.

**Methods**

**Participants**

One hundred and sixteen participants aged 1.6 to 5.6 years old at the time of the first recruitment were included in an ongoing accelerated longitudinal study. Sixty-eight children had an Autism Spectrum Disorder (ASD) diagnosis, and 48 were children with typical development (TD). Participants with ASD were recruited through French-speaking parent associations and specialized clinical centers. Participants with TD were recruited through announcements in the Geneva community. Children with ASD were assessed every six months and up to a maximum of five times until the age of 6 (mean length of the longitudinal data collection for the children with ASD was 1.4 years  $\pm$ 0.6). Children with TD were evaluated every six months during the first year (producing three time points) and then again after 12 months, for a maximum of four total visits and up to the age of 6 (mean length of the longitudinal data collection for the children with TD was 1.3 years  $\pm$ 0.6). The current study thus comprised a total of 369 time points, corresponding to 236 visits for the 68 children with ASD, and 133 visits for the 48 children with TD. The accelerated longitudinal design provided good coverage in both ASD and TD cohorts across the developmental period spanning the age range from 1.6 to 6 years (see Figure 1 for a graphical representation of data distribution in our sample). A statistical comparison of participants with ASD and participants with TD at their first visit (Time 1, T1) revealed no significant differences in age or gender distribution (see

Table 1). The Institutional Review Board of the University of Geneva approved the study protocol for all participants, and their parents gave their informed consent before inclusion in the study.

All children in the ASD group had received a clinical diagnosis of ASD before their inclusion in the study. Additionally, the diagnoses were verified using either the Autism Diagnosis Observation Schedule (ADOS) Generic (ADOS-G; Lord et al., 2000) or Second Edition (ADOS-2; Lord et al., 2012). ADOS assessments were administered and scored by psychologists qualified for research reliability. All ADOS scores were transformed in severity scores to allow for the comparison from different modules (Gotham et al., 2008; Esler et al., 2015). The mean severity score, which can range from 1 to 10, was of  $7.7 \pm 1.8$ . Children with TD were screened for neurological or psychiatric problems and learning disabilities before their inclusion in our research protocol. The parents of potential participants with TD completed an interview over the phone and a medical developmental history questionnaire before their initial visit to ensure eligibility. Participants with TD also were evaluated with the ADOS, which definitively excluded any sign of ASD and confirmed a severity score of 1 (the minimum), except for one child who received a severity score of 2.

(Figure 1)

**Figure 1.** Current data collection distribution available for each group from our ongoing longitudinal study.

**Table 1.** Statistical comparison between children with ASD and children with TD at the first visit in terms of their demographic, clinical, cognitive, social interest and behavioral features.

(Table 1)



**Assessment instruments.** At T1, all children were assessed for adaptive functioning, problem behaviors and social interest using the instruments described below. Besides, we also estimated Developmental Quotients (DQ) for cognitive and language functioning using the PsychoEducational Profile, 3<sup>rd</sup> edition (PEP-3; Schopler et al., 2005). Adaptive functioning measurements were subsequently repeated at each successive visit (T2, T3, T4, T5 for participants with ASD and T2, T3, T5 for participants with TD).

**Adaptive functioning.** Adaptive functioning was measured using the Vineland Adaptive Behavior Scales, 2<sup>nd</sup> Edition (VABS-II; Sparrow, Cicchetti & Balla, 2005). The VABS-II is a standardized interview completed by a parent or caregiver and used to assess adaptive functioning. The interview generates Standard Scores (SS) and adaptive levels for the following subdomains: communication, daily living skills, socialization and motor skills. The scale also produces a combined adaptive behavior composite score. The VABS-II has shown good validity and reliability in children with TD (Sparrow et al., 2005), which was also confirmed in children with ASD (Perry & Factor, 1989) using the previous version of the scale (the Vineland Adaptive Behavior Scales, VABS; Sparrow, Balla & Cicchetti, 1984). The interview was completed for all participants at each visit.

**Behavioral problems.** Behavioral problems were assessed using the Child Behavior Checklist 1.5-5 (CBCL 1.5-5; Achenbach & Rescorla, 2000), a scale intended for use with children aged 18 months to 5 years. The CBCL 1.5-5 is a questionnaire completed by caregivers that provides T-scores for total problems, as well as internalizing problems and externalizing problems subscales. T-scores further enable the categorization of clinical, borderline clinical or non-clinical scores according to a cut-off of 60 for each subscale. The CBCL 1.5-5 has shown good validity and reliability in children with TD (Achenbach & Rescorla, 2000) and in children with ASD (Pandolfi, Magyar & Dill, 2009). Out of 96 total CBCL 1.5-5 questionnaires at T1, we discarded incomplete CBCL 1.5-5 data from 13 children with ASD and 7 children with TD.

**Social interest.** We quantified participants' social interest by assessing their preference for biological motion during a biological motion visual preference eye-tracking task (Franchini et al., 2017; Franchini et al., 2016) based on Pierce et al. (2011). This passive one-minute task consists of the simultaneous presentation of dynamic geometrical motion on one side of the eye-tracking screen and dynamic biological motion on the other side. For geometrical motion, we presented moving geometrical shapes, similar to the classic abstract Mac OS screensavers or those available under General Public License laws (<http://www.reallyslick.com/screensavers>). For biological motion, we recorded standardized sequences of a child moving and dancing. To minimize distraction the scenes were recorded in front of a white wall. The six sequences showed boys and girls at a similar ratio, with three sequences showing a boy and three sequences showing a girl. In contrast to the fixed sequence used by Pierce et al. (2011), we alternated the side of stimulus type presentation (left or right side of the screen) between segments. Between each segment, a turning wheel was shown to bring participants' gazes to the middle of the screen.

The task was administered using Tobii Studio software ([www.tobii.com](http://www.tobii.com)) with a TX300 Tobii eye-tracker. The sampling rate of the machine was 300 Hz, and the video resolution was 1920x1080 pixels. As recommended by Tobii, participants sat on a parent's lap or alone on a chair at an approximately 60cm distance from the screen to minimize the impact of head movement on the gaze data. Before administering the task, all participants completed a five-point calibration procedure adapted to toddlers to detect eye motion and eye gaze.

Data analysis also was done with Tobii Studio (version 3.1.6). An I-VT filter was enabled during analysis (Classifier: 30°/s; Velocity calculator window length: 20ms). The *merge fixations* option was further enabled (max. time between fixations: 75ms; max. angle between fixations: 0.5°). Areas Of Interests (AOIs) were drawn on the paradigm videos to delineate biological motion and geometric motion. For each kind of stimuli (biological motion and geometric motion), we calculated the total sum of fixation duration (total watching time).

The percentage of time spent on each stimulus type (biological motion and geometric motion) was calculated by dividing the total time spent on each AOI (total fixation duration) by the total time spent on the entire screen. Visual preference for biological motion was only calculated if a child looked at the video during at least 50% of the time, per Pierce et al. (2011; 2015) and Franchini et al. (2016; 2017). According to this criterion, 7 children with ASD and 4 children with TD were excluded from analyses involving measures of social interest.

**Developmental Quotients.** The children included in our study were also assessed with the PEP-3 (Schopler et al., 2005), which allows evaluating developmental skills of young children from 2 to 7.5 years old. The PEP-3 has demonstrated good validity and reliability in children with TD and in children with ASD (Schopler et al. 2005; Giacomo et al., 2015). Nine children with TD and 7 children with ASD were yet too young to perform the evaluation. From the scores provided by the Cognition Verbal-Preverbal (CVP), Expressive Language (EL) and Receptive Language (RL) domains, we estimated a DQ that we used to describe our sample (see Table 1 and Table 2). The DQ is the result of a division of the developmental age of the children by their chronological age (e.g., Kawabe et al., 2016).

**Analysis strategy**

We started with descriptive cross-sectional statistical analyses comparing children with ASD to children with TD in terms of demographic, clinical, cognitive, social interest and behavioral characteristics. Student’s *t*-tests were conducted in PRISM 6.0 for Mac OS X with diagnosis as a fixed variable.

Then, we examined trajectory differences between children with ASD and children with TD. As described in the Participants section, all children included in our longitudinal cohort were between 1.6 and 5.6 years old at the time of the first recruitment. Longitudinal data (collected every six months) were included up to an age of 6 years. The accelerated longitudinal design allowed us to obtain a concentrated distribution of data in the whole age range (see Figure 1). Trajectories of adaptive functioning were then calculated using mixed

model regression analyses (Mutlu et al., 2013, Shaw et al., 2006, Thompson et al., 2011). This analysis approach has previously been applied in samples of similar size using an accelerated design (e.g., Maeder et al. 2016; Schneider et al. 2014). Specifically, developmental trajectories were estimated by fitting random-slope models to our data, taking into account both within-subject and between-subject effects to estimate developmental trajectories. Linear random-slope models including group, age and their interaction as fixed effects were estimated using the *nlmefit* function in MATLAB R2014b (MathWorks). A likelihood ratio test was subsequently performed to quantify the significance of between-group differences in the intercept and slope of the developmental adaptive function trajectories. In the results, we reported intercept ( $\alpha$ ) and slope ( $\beta$ ) for estimated trajectories of each group, along with the corresponding *p*-values. By modeling the within-subject intercept and slope as random effects, this mixed model regression approach provides a robust estimate of the average developmental trajectory for each cohort despite multiple measurements, missing data points and variable time intervals between measurements (Shaw et al., 2006; Thompson, Hallmayer & O'Hara, 2011). Therefore, regardless of the fact that some time points in our ongoing study were missing, the remaining time points were still useful for estimating group trajectories.

Finally, we divided the group of children with ASD into subgroups according to their scores at T1 and applied the same mixed model regression approach to probe for differences in the developmental trajectories of adaptive functioning between the respective subpopulations. The group of children with ASD was divided into two subgroups according to three different variables: a. ADOS symptom severity ( $>8$ =high or  $<8$ =low to moderate), b. Social interest ( $>50\%$  of time spent on biological motion or  $<50\%$  of time spent on biological motion), and c. Behavioral problems (T-score  $>60$ =clinical or borderline clinical range T-score,  $<60$ =nonclinical range). We then applied mixed model regression as described above to compare the subgroups of children with ASD resulting from each division (variables a, b or c).

Results of the VABS-II adaptive behavior composite were considered significant at

$p \leq 0.05$ . Given that the VABS-II includes four different subdomain scores and the CBCL 1.5-5 contains two subdomains, we applied a Bonferroni correction for multiple comparisons when investigating the subdomains: an alpha-level of  $p \leq 0.0125$  for the VABS-II subdomains, an alpha-level of  $p \leq 0.025$  for the CBCL 1.5-5 subdomains.

**Results**

**Cross-sectional results (Time 1)**

Children with ASD and children with TD did not differ in age or gender distribution (Table 1). Children with ASD showed reduced preference for biological motion, more behavioral problems and decreased cognitive functioning compared to children with TD (Table 1). When we compared subgroups of children with ASD, we did not find group differences for CVP, EL and RL DQ scores from the PEP-3 between children with ASD with low or high symptom severity and low or high behavioral problems. A trend toward significance was however evident for the CVP scores between children with ASD with low or high symptom severity, i.e. children with higher symptom severity were showing lower scores. All the PEP-3 DQ scores (CVP, EL, and RL) differed between the children with ASD with low or high social interest (with children with a higher interest in geometric motion showing lower scores) (see Table 2).

**Table 2.** Differences at the PEP-3 (developmental quotients for cognitive verbal-preverbal, expressive language, and receptive language) between subgroups of children with ASD, defined according to low or high score on symptom severity, behavioral problems, and social interest.

(Table 2)

## Longitudinal results

### Adaptive functioning trajectories in children with ASD and children with TD.

Adaptive functioning trajectories were lower in children with ASD at all ages on all VABS-II domains (Table 3). For the adaptive behavior composite (see Figure 2a), daily living skills and socialization scores (see Figure 2b) participants with ASD and participants with TD did not differ in trajectories slope. In the communication and motor skills subdomains, children with ASD and children with TD displayed significant slope difference when: a. The communication scores from the ASD group increased with age while remaining constant in the group of children with TD; b. The motor skills scores from the ASD group decreased with age but increased in the group of children with TD (see Table 3, see Figure 2b).

(Figure 2)

**Figure 2.** Developmental trajectories of adaptive functioning for children with ASD compared to children with TD in *a.* VABS-II total adaptive composite scale *b.* VABS-II subscales of communication, daily living skills, socialization and motor skills.

**Table 3.** Differences in developmental trajectories between groups of children with ASD and children with TD.

(Table 3)

**Adaptive functioning trajectories in subgroups of children with ASD.** Subgroups of children with ASD were created based on three different criteria to compare developmental trajectories of adaptive functioning: a. Children with high symptom severity (n=38) and children with low to moderate symptom severity (n=30), b. Children who preferred biological (n=21) or geometric motion (n=40), c. Children with clinical-borderline (n=28) or nonclinical (n=27) range of behavioral problems. For each category (lower or higher functioning according to symptom severity scores, social interest or behavioral problems), we compared subgroups of children with ASD.

The subgroups with higher symptom severity and higher interest in geometric motion demonstrated lower adaptive behavior composite trajectories (lower intercept with lower scores at all ages) compared to children with lower symptom severity and a preference for biological motion (see Figure 3 & Table 4). When comparing subgroups of children with high vs. low behavioral problems, both subgroups showed similar levels at youngest age. However, children with high behavioral problems presented a declining linear trajectory, while children with low behavioral problems improved their adaptive behavior composite scores with age (significant differences in intercept and slope between the two subgroups) (see Figure 3 & Table 4).

Children with ASD who showed a preference for geometric motion at our eye-tracking task showed significant group difference indicated by a lower intercept and lesser scores at all ages in all VABS-II subscales (communication, daily living skills, socialization and motor skills) compared to children with ASD who preferred biological motion. A trend toward significance was further found indicating lesser slopes in the daily living skills, socialization and motor skills subscales in children who showed a preference for geometric motion (see Table 4).

Finally, we found that children with ASD with high symptom severity and clinical-borderline behavioral problems showed faster decreasing trajectories in the VABS-II

socialization subscale, compared to children with ASD with lower symptom severity scores or non-clinical behavioral problems (with a significant difference in slope between the two subgroups) (see Table 4). Similarly, children with clinical-borderline behavioral problems showed faster declining motor skills trajectories than children with non-clinical scores (with a significant difference in slope between the two subgroups) (see Table 4). Concerning behavioral problems subscales, we also found faster declining trajectories on the VABS-II socialization subscale for the internalizing subscale in children with clinical-borderline behavioral problems (with a significantly reduced slope). We did yet not find the same result for the externalizing subscale (see Table 4).

(Figure 3)

**Figure 3.** Developmental trajectories at the adaptive composite from the VABS-II for subgroups of children with ASD. Subgroups have been created in consideration of lower or higher scores for symptom severity, social interest, and behavioral problems.

**Table 4.** Differences in developmental trajectories between subgroups of children with ASD defined according to low or high score on symptom severity, behavioral problems, and social interest.

(Table 4)

## Discussion

In this study, we first observed increased behavioral problems and reduced social interest in young children with Autism Spectrum Disorders (ASD) compared to children with typical development (TD). Compared to children with TD, children with ASD showed constantly lower adaptive functioning from 1.6 to 6 years old as measured by the adaptive behavior composite Standard Scores (SS) from the Vineland Adaptive Behavior Scales, 2<sup>nd</sup> Edition (VABS-II; Sparrow, Cicchetti & Balla, 2005). Besides, children with ASD showed



increasing trajectories over time in the communication subdomain and decreasing trajectories in the motor skills subdomain, while children with TD showed constant or increasing trajectories. Second, we observed that among participants with ASD, the subgroup with high symptom severity, high behavioral problems and showing a preference for geometric stimuli demonstrated lower or decreasing adaptive functioning over time compared to children with ASD with higher overall functioning (low to moderate symptom severity, non-clinical behavioral problems and a preference for social stimuli). Children who showed a stronger social interest also displayed higher functioning in the four VABS-II subdomains (communication, daily living skills, socialization and motor skills). Also, in the socialization subdomain, we observed that children with ASD with high symptom severity and behavioral problems showed faster decreasing trajectories over time compared to children with low to moderate symptom severity and non-clinical behavioral problems.

**Decreased developmental trajectories of adaptive functioning in children with ASD when compared to children with TD**

It is widely accepted that children on the autism spectrum exhibit reduced adaptive functioning compared to children with TD (Bal et al., 2015; Liss et al., 2001; Szatmari et al., 2015; Volkmar et al., 1987). These studies all used the VABS (either the first or second version) to measure adaptive behavior and universally observed deficits in all four adaptive subdomains: communication, daily living skills, socialization and motor skills. Within each of these four subdomains, our results show reduced trajectories of adaptive functioning in children with ASD compared to children with typical development. A difference in slope was found between the two groups in communication (participants with TD showed a constant trajectory, while SS in participants with ASD increased their score with age). A previous study by Freeman and collaborators (1999) showed that VABS SS for communication, daily living skills, and socialization increase from age 2 to 19 in individuals with ASD, but only the improvement in communication scores was correlated with IQ scores. Our results confirm

improved communication skills during development in ASD and a gradual reduction over time of the gap that separates them from their peers with TD. Communication in toddlers with ASD during the first few years of life is evaluated according to non-verbal skills (e.g., use of gestures) in addition to verbal skills, as opposed to purely verbal skills later in life. Indeed, non-verbal communication (e.g., gestures) helps to identify the communication deficits that are a hallmark of autism (Luyster, Kadlec, Carter, & Tager-Flusberg, 2008; Mundy, Sigman, Ungerer, & Sherman, 1986), increasing the gap difference when children with ASD are compared to children with TD. Therefore, the more significant gap in the VABS-II SS for communication shown in the group of children with ASD compared to children with TD very early in life might be partially explained by the non-verbal content of the evaluation. Even though this hypothesis remains speculative and has to be explicitly tested, previous evidence showed the importance of non-verbal communication in the characterization of the developmental profile in young children with ASD (e.g., Mundy, Sigman, Ungerer & Sherman, 1986; Stone, Ousley, Hogan & Hepburn, 1997). If this is the case, a separate assessment of verbal and non-verbal communication during early childhood could be particularly relevant to describe communication-related difficulties of young children with ASD.

Our results also show that participants with TD displayed increased trajectories over time in the motor skills subdomain, whereas the SS from the participants with ASD decreased with age, thereby increasing the developmental gap between groups. A decline in motor skills in infants and preschoolers with autism has previously been observed and is associated with worse outcomes (Dawson et al., 2010; Landa & Garrett-Mayer, 2006). Our results suggest that motor skills progressively decrease during the preschool period. This downturn can be explained by a qualitative difference in the way motor skills are evaluated in toddlers during the first years of life, compared to children at the end of their preschool years. Motor skills during early infancy are assessed on context-independent abilities (e.g., running abilities), whereas, later in life, motor skills are more often evaluated according to context-dependent

skills (e.g., correct scissors use). This difference makes them more closely tied to academic experiences and learning opportunities, which are often disparate among children with ASD and children with TD. The divergence in context-dependent learning opportunities between children with ASD and children with TD can also, at least in part, support findings relating motor skills abilities to ASD symptomology (e.g., Sutura et al., 2007; MacDonald, Lord, Ulrich, 2014). Finally, it is important to acknowledge that the VABS-II SS for motor skills are only provided up to 6 years old. Despite the VABS-II has shown excellent reliability and internal consistency (Sparrow et al., 2005; also see Perry & Factor, 1989), the observed decreased in SS of the motor skills subdomain could underlie approaching a plateau effect.

**Symptom severity is related to early trajectories of adaptive functioning in children with autism**

We found similar results as Szatmari et al. (2015) by showing that symptom severity influences the development of adaptive functioning in children with ASD. Children with autism with more severe symptomatology show reduced adaptive functioning at baseline, as well as lower adaptive functioning throughout the preschool years. Moreover, participants with ASD with more severe symptoms exhibited a decreasing slope in the socialization subdomain, whereas children with lower symptom severity showed a constant slope. This downturn means that the severity of symptomatology in infancy may substantially contribute to the evolution of social skills in preschoolers with autism.

Per Perry et al. (2009), we found that the adaptive functioning profile of preschoolers with autism is characterized by impairments that are particularly evident in their socialization skills and consistent with core features of an ASD diagnosis. Symptom severity, as measured by the Autism Diagnosis Observation Schedule (ADOS), is largely based on impaired social affect (Gotham, Pickles, & Lord, 2009). By consequence, our results suggest that differences in the developmental trajectories of socialization behavior in preschoolers with ASD (e.g., in

skills like Emotion Recognition and Expressions or Friendship Development) may be principally driven by abilities that influence social affect scores on the ADOS (e.g., items of Quality of Social Overtures or Shared Enjoyment in Interactions). However, this close association may be something that is specifically related to the age of our subjects. A study by Klin and collaborators (2007) showed that socialization skills tend to decrease with age, whereas social affect scores from the ADOS tend to stay stable in children with ASD. This finding points to the possibility that impaired social affect may prevent social behaviors from developing, perhaps due to diminished social opportunities. This could underscore the importance of stimulating social behavior and social interest in young individuals with ASD during early interventions. Increasing the social learning opportunities in children with ASD has, in turn, the potential of positively impact their development (see Dawson et al., 2009; Dawson et al., 2004; Chevallier et al., 2012).

### **Social interest is related to early trajectories of adaptive functioning in children with autism**

Commensurate with previous research, we found that preschoolers with ASD orient less towards social stimuli (biological) than children with TD (Pierce et al., 2011; 2015). Social interest has been positively associated with cognitive and adaptive functioning in preschoolers with ASD (Pierce et al. 2015), as well as with decreased symptom severity in young children with ASD (Franchini et al., 2016). Furthermore, social interest is thought to be critical for creating learning opportunities and for bolstering social learning (Chevallier et al., 2012; Dawson et al., 2004). Results from the present study add to previous research by linking social interest with developmental increases in adaptive functioning. Moreover, the current study showed a trend in declining trajectories for daily living skills, socialization and motor skills subdomains in preschoolers with ASD who showed a stronger interest in non-social stimuli compared to children with ASD who showed a stronger preference for social stimuli. These

1 results may suggest that decreased social interest negatively impacts the developmental course  
2 of early acquisitions in preschoolers with ASD. Measures of social interest are particularly  
3 promising while defining the clinical profile of young children with ASD because of their  
4 potential of informing about clinical signs very early in life (Pierce et al., 2011; Pierce et al.,  
5 2015; Bacon et al., 2017). Moreover, as previously suggested, social interest appears to be a  
6 critical intervention target very early in life. Increasing children's interest in their social world  
7 can positively impact their further development (Dawson et al., 2009; Dawson et al., 2004). It  
8 is yet important to note that social interest preference was associated with cognitive functioning  
9 and language skills in our sample of young children with ASD, where children with ASD who  
10 showed reduced social interest were also more impaired in their cognitive and language skills.  
11 This confirms, at least in part, the association between social interest and cognitive functioning  
12 very early in the life of children with ASD (e.g., Pierce et al., 2011). Besides, it could also  
13 suggest that cognitive functioning plays a role in the association between social interest and  
14 adaptive functioning in those children (e.g., Matthews et al., 2015).

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33 **Behavioral problems are related to early trajectories of adaptive functioning in**  
34 **children with autism**

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38 The description of our sample at baseline confirms elevated behavioral problems.  
39 Within our sample of preschoolers with autism, about a half (50.9%) met criteria for behavioral  
40 problems that fell in the clinical or borderline clinical range according to the CBCL 1.5-5. The  
41 prevalence of maladaptive behaviors in both the internalizing and externalizing subdomains in  
42 our sample matches those reported in previous studies (Eisenhower, Baker, & Blacher, 2005;  
43 Hartley et al., 2008; Muratori et al., 2011; Pandolfi, Magyar, & Dill, 2009). A study by Fulton  
44 and collaborators (Fulton et al., 2014) showed that improvements in maladaptive behavior in  
45 preschoolers with ASD improved their overall outcomes from intensive early intervention.

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55 Furthermore, we found that children with ASD with overall behavioral problems and

1 internalizing behavioral problems (but not externalizing problems) in the clinical range  
2 demonstrate faster decreasing developmental trajectories over time, in particular in the  
3 socialization subdomain. It has been similarly shown that two groups of children with ASD  
4 divided according to membership to symptom severity trajectories (higher or lower) differed on  
5 internalizing but not externalizing problems on the CBCL 1.5-5 (Szatmari et al. 2015). Taken  
6 together, these results suggest that internalizing problems may be more related to social  
7 engagement aspects of ASD symptomatology (e.g., withdrawal) than externalizing problems.  
8 Also, a previous study of high functioning adolescents with ASD indicated that internalizing  
9 symptoms are inversely related to emotional functioning; individuals with autism with more  
10 outgoing behavior and friendships with their peers seem to be more protected from  
11 internalizing disorders (Schwartz et al., 2009). Commensurate with these results, we found that  
12 internalizing disorders influence the development of adaptive functioning early on in children  
13 on the autism spectrum. We suggest that a specific evaluation of those behaviors should be  
14 included in clinical assessments in young children with ASD, given their potential to impact  
15 their further development. Finally, as discussed in Fulton et al. (2014), targeting the child  
16 arousal level during early interventions, and thus modulating a maladaptive child response, can  
17 be preventive of future behavioral problems (e.g., see the Early Start Denver Model; Rogers &  
18 Dawson, 2010).

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43 The present study has several limitations. First, while we are convinced that our results  
44 capture a critical period (the preschool years) for the development of social orienting and  
45 behavioral problems, we were only able to explore the outcomes of preschoolers with ASD  
46 during a short time. Future investigations should aim to examine the influence of early social  
47 interest and behavioral problems over a more extended developmental period to understand  
48 their effects on later outcome. Second, our results explore the developmental impact of only  
49 three selected ASD behavioral features (symptom severity, social interest, behavioral  
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problems) on adaptive functioning. We chose these specific features because they previously showed strong associations with adaptive functioning. However, other behavioral characteristics also have been found to be related to autistic symptomatology, and adaptive functioning (e.g., executive functioning, Pugliese et al., 2016 or cognitive functioning, Szatmari et al. 2015) and should be taken into account. Third, we observed considerable variability in adaptive functioning between participants with ASD. This heterogeneity suggests that it may be possible to identify subgroups of children with ASD with differential adaptive functioning trajectories. The current study would indicate that behavioral features of individuals with ASD, such as symptom severity, social interest, and behavioral problems, could be used to understand this heterogeneity (e.g., subgroup identification). Finally, in consideration of the data collection strategy used in this study, the rate of missing data in our sample was significant. For this reason, we chose to apply mixed model regression analyses (Mutlu et al., 2013), which allow considering both within- and between-subject effects and are especially well suited for the analysis of distributed data collections. However, the mixed nature of estimated trajectories, which contain both cross-sectional and longitudinal effects, has to be considered in the interpretation of the results. Moreover, due to our sample size, we chose only to consider linear trajectories. A more significant amount of data could have yet provided non-linear trajectories (e.g., quadratic), which should be tested in future studies.

**Conclusions**

The current longitudinal study shows that in addition to high symptom severity, both reduced social interest and greater behavioral problems are associated with lower adaptive functioning in young children with ASD. These findings reinforce the previously proposed idea that social interest and behavioral problems are critical to early adaptive functioning in children with autism and serve to identify intervention targets that can help improve adaptive behavior in preschoolers with ASD.

## Conflict of interest and ethical standards

The authors declare no conflicts of interest. The local ethical commission of Geneva approved this study.

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	<u>ASD - Mean (SD)</u>	<u>TD – Mean (SD)</u>		
	n=68	n=48	test statistic	<i>p</i> -value
Gender	11♀/57♂	12♀/36♂	$\chi^2=1.38$	0.240
Age (years old)	2.94 (0.10)	2.71 (0.15)	$t=1.34$	0.182
ADOS Symptom Severity	7.69 (1.82)	1.02 (0.02)	$t=25.16$	<0.0001*
Preference for Biological Motion (%)	42.53 (2.24)	68.67 (2.37)	$t=7.82$	<0.0001*
CBCL 1.5-5 Total score (T-score)	59.60 (1.28)	42.05 (1.84)	$t=8.10$	<0.0001*
CBCL 1.5-5 Internalizing score (T-score)	59.09 (1.13)	44.71 (1.95)	$t=5.36$	<0.0001*
CBCL 1.5-5 Externalizing score (T-score)	60.04 (1.30)	38.73 (1.56)	$t=10.54$	<0.0001*
PEP-3 Cognitive Verbal-Preverbal (DQ)	70.04 (3.04)	122.10 (3.89)	$t=10.63$	<0.01*
PEP-3 Expressive Language (DQ)	46.08 (2.03)	89.05 (2.82)	$t=12.66$	<0.01*
PEP-3 Receptive Language (DQ)	50.52 (2.41)	117.40 (4.87)	$t=13.57$	<0.01*

\*Indicates significant results. Alpha-level was considered as significant at  $p \leq 0.025$  for the CBCL 1.5-5 subdomains and at  $p \leq 0.017$  for the PEP-3 domains according to Bonferroni correction.

Subgroup definition	ASD-Low - Mean (SD)	ASD-High – Mean (SD)	test statistic	p-value
<b>Low or High Symptom Severity (ADOS)</b>				
ASD-High, n=38 / ASD-Low, n=30				
PEP-3 Cognitive Verbal-Preverbal (DQ)	78.19 (3.58)	63.81 (3.58)	<i>t</i> =2.44	0.018
PEP-3 Expressive Language (DQ)	50.22 (3.25)	42.91 (2.50)	<i>t</i> =1.82	0.075
PEP-3 Receptive Language (DQ)	55.03 (3.61)	47.08 (3.16)	<i>t</i> =1.66	0.103
<b>Low or High Interest on Geometric Motion</b>				
ASD-High, n=40 / ASD-Low, n=21				
PEP-3 Cognitive Verbal-Preverbal (DQ)	84.92 (5.31)	63.53 (3.37)	<i>t</i> =3.42	<0.01*
PEP-3 Expressive Language (DQ)	54.35 (4.48)	42.61 (2.14)	<i>t</i> =2.67	<0.01*
PEP-3 Receptive Language (DQ)	62.69 (4.99)	45.45 (2.49)	<i>t</i> =3.43	<0.01*
<b>Low or High Behavioral Problems</b>				
<b>Total (CBCL 1.5-5)</b>				
ASD-High, n=28 / ASD-Low, n=27				
PEP-3 Cognitive Verbal-Preverbal (DQ)	75.55 (4.78)	65.17 (4.59)	<i>t</i> =1.11	0.271
PEP-3 Expressive Language (DQ)	44.81 (2.49)	46.61 (3.30)	<i>t</i> =0.43	0.669
PEP-3 Receptive Language (DQ)	50.62 (2.98)	49.84 (3.99)	<i>t</i> =0.15	0.879

\*Indicates significant results. Alpha-level was considered as significant at  $p \leq 0.017$  for the PEP-3 domains according to Bonferroni correction.

	ASD <i>intercept, <math>\beta^a</math>, [95% CIs<sup>b</sup>]</i>	TD <i>intercept, <math>\beta</math>, [95% CIs]</i>	Group effect		Slope	
			Likelihood ratio, <i>df</i>	Group effect ( <i>p</i> -value)	Likelihood ratio, <i>df</i>	<i>p</i> -value
VABS-II Adaptive Behavior	75.87[±2.37], 0.15[±0.55]	103.83[±2.87], -0.17[±0.77]	118.25, 2	<0.001*	0.11, 1	0.74
Composite (SS)						
Communication	63.80[±2.99], 3.47[±0.71]	104.44[±3.63], 0.30[±0.95]	109.35, 2	<0.001*	6.68, 1	<0.01*
Daily Living Skills	85.02[±2.75], -1.37[±0.68]	104.58[±3.29], -2.07[±0.91]	87.14, 2	<0.001*	0.41, 1	0.52
Socialization	80.26[±2.93], -1.14[±0.57]	105.67[±2.72], -1.37[±0.75]	133.96, 2	<0.001*	0.06, 1	0.81
Motor Skills	93.24[±2.67], -2.52[±0.66]	97.27[±3.18], 1.51[±0.88]	68.05, 2	<0.001*	12.99, 1	<0.001*

<sup>a</sup> $\beta$ =trajectory gradient. <sup>b</sup>CIs= Confidence Intervals. \*Indicates significant results. Alpha-level was considered as significant at  $p \leq 0.0125$  for the VABS-II subdomains according to Bonferroni correction.

				Group effect		Slope	
1 Subgroup definition		ASD-Low	ASD-High	Likelihood ratio, <i>df</i>	<i>p</i> -value	Likelihood ratio, <i>df</i>	<i>p</i> -value
2		intercept, $\beta^a$ , [95% <i>CI</i> s <sup>b</sup> ]	intercept, $\beta$ , [95% <i>CI</i> s]				
3							
4 Low or High Symptom Severity	VABS-II Adaptive Behavior	76.72[±3.28], 0.85[±0.70]	72.32[±3.36], 0.10[±0.80]	7.45, 2	0.02*	0.50, 1	0.47
5 (ADOS)	Composite (SS)						
6	Communication	67.60[±4.42], 3.74[±0.96]	56.50[±4.53], 4.09[±1.09]	7.64, 2	0.02	0.06, 1	0.81
7	Daily Living Skills	89.82[±4.02], -1.60[±0.81]	77.02[±4.13], -0.14[±1.02]	7.35, 2	0.03	1.27, 1	0.26
8	Socialization	76.59[±3.17], 0.73[±0.73]	83.77[±3.25], -5.18[±0.81]	18.89, 2	<0.001*	11.01, 1	<0.001*
9	Motor Skills	90.35[±3.71], -1.19[±0.85]	93.74[±3.81], -3.29[±0.95]	5.29, 2	0.07	2.68, 1	0.12
10							
11							
12 Low or High Interest on Geometric	VABS-II Adaptive Behavior	79.36[±4.18], 0.15[±0.90]	73.50[±2.79], 1.31[±0.65]	12.22, 2	<0.01*	3.79, 1	0.09
13 Motion	Composite (SS)						
14	Communication	69.37[±5.80], 4.10[±1.28]	59.35[±3.88], 3.83[±0.92]	7.78, 2	<0.01*	0.03, 1	0.87
15	Daily Living Skills	78.45[±5.05], 1.96[±1.14]	82.03[±3.38], -1.09[±0.82]	10.34, 2	<0.01*	4.66, 1	0.03
16	Socialization	77.42[±4.23], 1.03[±0.98]	79.66[±2.83], -1.54[±0.70]	12.90, 2	<0.01*	4.48, 1	0.03
17	Motor Skills	89.75[±4.54], -0.12[±1.03]	92.98[±3.03], -3.02[±0.73]	11.79, 2	<0.01*	5.17, 1	0.02
18							
19 Low or High Behavioral Problems	VABS-II Adaptive Behavior	72.74[±3.53], 1.90[±0.81]	77.36[±3.87], -1.17[±0.90]	10.43, 2	<0.01*	6.35	0.01*
20	Composite (SS)						
21 Total (CBCL 1.5-5)							
22	Communication	63.02[±4.65], 4.53[±1.06]	60.82[±5.09], 3.26[±1.18]	3.39, 2	0.18	0.64, 1	0.42
23	Daily Living Skills	81.75[±4.41], 0.24[±1.08]	85.30[±4.84], -2.27[±1.19]	5.18, 2	0.08	2.43, 1	0.12
24	Socialization	76.42[±3.50], 0.99[±0.86]	85.75[±3.85], -3.27[±0.94]	14.83, 2	<0.001*	10.82, 1	<0.01*
25	Motor Skills	90.74[±4.09], -0.99[±1.02]	97.82[±4.49], -4.71[±1.11]	10.44, 2	<0.01*	6.01, 1	0.01*
26							
27 Low or High Behavioral Problems	VABS-II Adaptive Behavior	75.45[±4.11], -0.06[±0.98]	73.83[±3.56], 0.89[±0.79]	0.88, 2	0.64	0.57, 1	0.45
28 Internalizing (CBCL 1.5-5)	Composite (SS)						
29	Communication	60.47[±5.30], 3.79[±1.27]	62.50[±4.60], 4.05[±1.01]	0.62, 2	0.73	0.03, 1	0.87
30	Daily Living Skills	80.94[±5.06], -0.51[±1.28]	84.42[±4.41], -1.14[±1.04]	0.29, 2	0.86	0.14, 1	0.71
31	Socialization	76.41[±3.89], 0.07[±0.94]	85.33[±3.50], -2.85[±0.92]	4.78, 2	0.03	3.91, 1	0.01*
32	Motor Skills	91.20[±4.13], -1.91[±0.99]	97.38[±4.71], -3.96[±1.21]	1.84, 2	0.40	1.70, 1	0.12
33							
34							
35 Low or High Behavioral Problems	VABS-II Adaptive Behavior	70.29[±3.40], 1.31[±0.78]	80.73[±4.28], -0.72[±0.98]	3.63, 2	0.16	2.60, 1	0.11
36 Externalizing (CBCL 1.5-5)	Composite (SS)						
37	Communication	57.34[±4.40], 4.92[±1.00]	68.04[±5.54], 2.50[±1.26]	2.49, 2	0.29	2.22, 1	0.14
38	Daily Living Skills	78.37[±4.20], 0.05[±1.03]	90.13[±5.26], -2.36[±1.28]	3.03, 2	0.22	2.14, 1	0.14
39	Socialization	79.29[±3.44], -1.05[±0.84]	81.34[±4.31], -0.89[±1.05]	0.92, 2	0.63	0.01, 1	0.91
40	Motor Skills	97.05[±4.20], 0.05[±1.03]	89.35[±3.95], -1.96[±0.98]	3.40, 2	0.18	1.50, 1	0.22
41							
42							

<sup>a</sup>  $\beta$ =trajectory gradient. <sup>b</sup> *CI*s= Confidence Intervals. \*Indicates significant results. Alpha-level was considered as significant at  $p \leq 0.0125$  for the VABS-II subdomains according to Bonferroni correction.

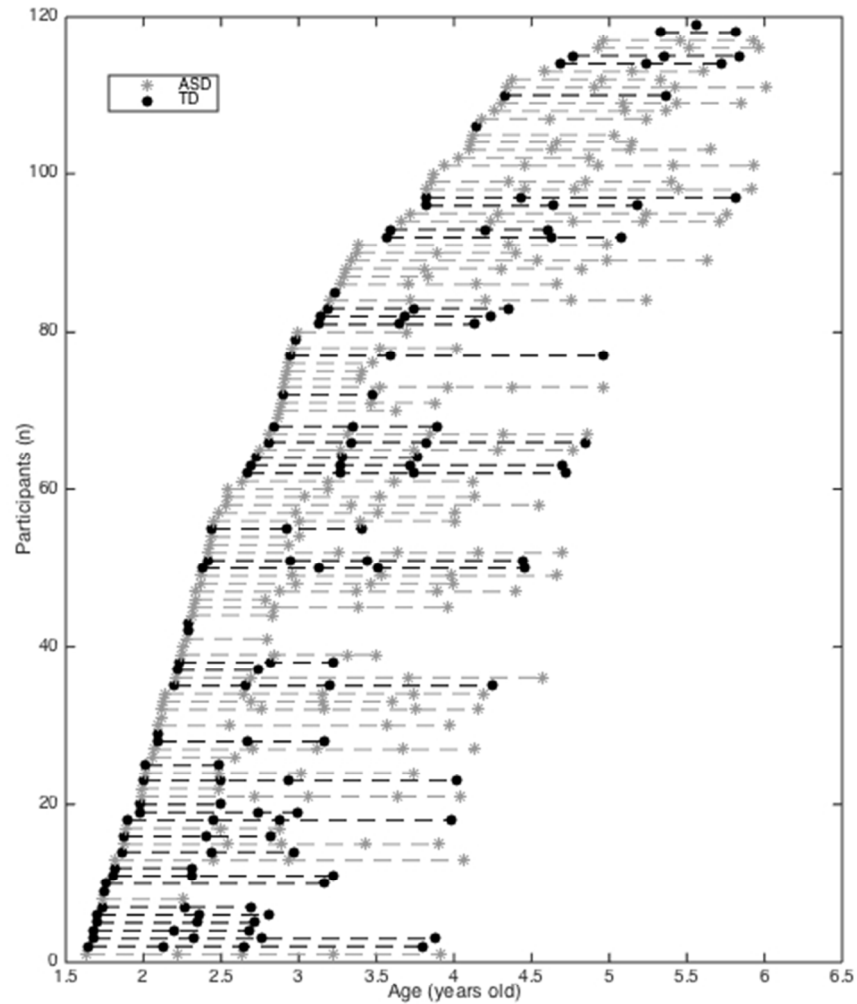


Figure 1. Current data collection distribution available for each group from our ongoing longitudinal study.

197x228mm (72 x 72 DPI)

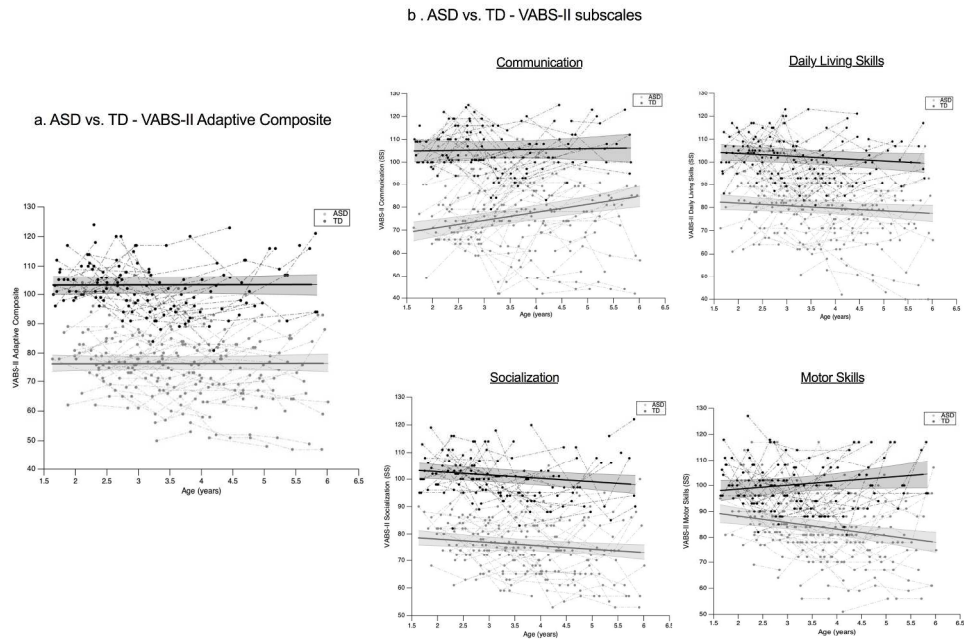


Figure 2. Developmental trajectories of adaptive functioning for children with ASD compared to children with TD in a. VABS-II total adaptive composite scale b. VABS-II subscales of communication, daily living skills, socialization and motor skills.

272x176mm (300 x 300 DPI)

ASD subgroups (lower vs. higher functioning) - VABS-II Adaptive Composite

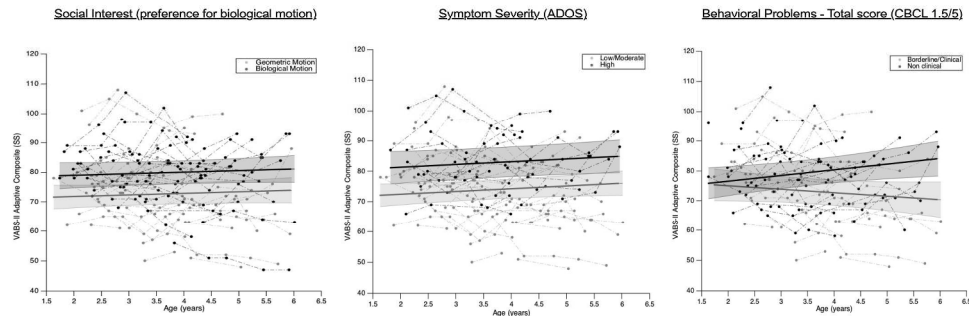


Figure 3. Developmental trajectories at the adaptive composite from the VABS-II for subgroups of children with ASD. Subgroups have been created in consideration of lower or higher scores for symptom severity, social interest, and behavioral problems.

272x113mm (300 x 300 DPI)