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## Cognitive and adaptive correlates of an ADOS-derived joint attention composite

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### Abstract

Joint attention skills have been shown to predict language outcomes in children with autism spectrum disorder (ASD). Less is known about the relationship between joint attention (JA) abilities in children with ASD and cognitive and adaptive abilities. In the current study, a subset of items from the Autism Diagnostic Observation Schedule (ADOS), designed to quantify JA abilities, were used to investigate social attention among an unusually large cross-sectional sample of children with ASD ( $n = 1061$ ). An examination of the association between JA and a range of functional correlates (cognitive and adaptive) revealed JA was significantly related to verbal (VIQ) and non-verbal (NVIQ) cognitive ability as well as all domains of adaptive functioning (socialization, communication, and daily living skills). Additional analyses examined the degree to which the relation between adaptive abilities (socialization, communication, and daily living skills) and JA was maintained after taking into account the potentially mediating role of verbal and nonverbal cognitive ability. Results revealed that VIQ fully mediated the relation between JA and adaptive functioning, whereas the relation between these adaptive variables and JA was only partially mediated by NVIQ. Moderation analyses were also conducted to examine how verbal and non-verbal cognitive ability and gender impacted the relation between JA and adaptive functioning. In line with research showing a relation between language and JA, this indicates that while JA is significantly related to functional outcomes, this appears to be mediated specifically through a verbal cognitive pathway.

### Keywords

ASD; Joint attention; Adaptive functioning; Cognition; ADOS

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## 1. Introduction

Deficits in social communication and interaction are a core feature of autism spectrum disorder (ASD; American Psychiatric Association, 2013). Although individuals with ASD commonly have a heterogeneous profile of social impairment, deficits in joint attention (JA) are consistently observed within this population (Bruinsma, Koegel, & Koegel, 2004; Dawson, Meltzoff, Osterling, & Rinaldi, 1998; Dawson et al., 2004; Mundy, Sigman, Ungerer, & Sherman, 1986) and JA is frequently examined as a prognostic indicator (Charman et al., 2003). JA behaviors consist of triadic social attentional exchanges between individuals and some object in the social world for the purpose of social sharing. JA appears to be related to a range of important functional outcomes. For example, research demonstrating a relation between JA and other early social behaviors such as social orienting and imitation (e.g., Hobson & Hobson, 2007; Johnson, Gillis, & Romanczyk, 2012; Leekam & Ramsden, 2006) aligns with theory suggesting that JA is one of the key components of a complex cluster of social impairment in ASD (Schultz, 2005). Moreover, JA behaviors have been found to predict positive social outcomes in infants at risk for autism (infant siblings of children with ASD; Malesa et al., 2013) and in children at risk for poor developmental and behavioral outcomes secondary to prenatal drug exposure (Sheinkopf, Mundy, Claussen, & Willoughby, 2004). While less research has examined the impact of JA deficits in older children, evidence from longitudinal research demonstrates that impairments in JA among infants with ASD remain stable later in childhood (Mundy, Sigman, & Kasari, 1990; Sigman et al., 1999). Such findings indicate that research examining functional outcomes related to JA impairment later in childhood would be beneficial.

Early JA skills also predict later language ability in both children with autism and in typical development (e.g., Charman et al., 2003; Mundy & Gomes, 1998; Mundy et al., 1990). Research has shown that language impairment in ASD is not only directly related to adaptive impairment across domains of communication and socialization, but also, daily living skills (Liss et al., 2001). These prior findings indicate that the relationship between JA and social outcomes may in some ways be mediated by language development. For example, in both Malesa et al. (2013) and Sheinkopf et al. (2004), the infants' initiation of JA with others was related to language outcomes, whereas responding to the JA bids of others was related to measures of social competence and skill. A review by Koegel (2000) discusses the importance of incorporating precursor skills such as JA into interventions to improve language outcomes. More recent treatment approaches have also conceptualized JA as a pivotal skill that may be related to positive language and social outcomes (e.g., Kasari, Gulsrud, Wong, Kwon, & Locke, 2010).

In spite of the pivotal role JA plays in language development, it is often not included as a key phenotypic variable in epidemiological studies or genetic investigations of ASD. Additionally, little research has conducted large-scale studies to examine how JA relates specifically to functional outcomes such as cognition or adaptive measures of communication, socialization, and daily living skills. This void in the research is driven in part by methodological challenges associated with quantifying JA. However, given that JA appears to be a pivotal early social skill, increased research is needed to understand how it relates to impairment in other functional outcomes such as adaptive skills (Szatmari et al.,

2009). In addition, we need to provide a clearer picture of this behavioral phenotype for inclusion in future genetic research. Additional investigations of the relation between JA and adaptive correlates may help to enhance efforts to tailor interventions for maximum effectiveness across these domains. For example, there is evidence that interventions targeting JA also result in improvement in other domains of social functioning, such as imitation (e.g., Ingersoll & Schreibman, 2006; Whalen, Schreibman, & Ingersoll, 2006). Furthermore, research supports that interventions designed specifically to target improvements in JA result in collateral gains in language (e.g., Jones, Carr, & Feeley, 2006; Kasari, Gulsrud, Freeman, Paparella, & Helleman, 2012; Whalen et al., 2006).

### 1.1. Importance of new methods for assessing joint attention

Specific paradigms designed to elicit and quantify JA typically require time-intensive administration and behavioral coding protocols (Mundy et al., 1986; Seibert, Hogan, & Mundy, 1982; Wetherby & Prizant, 1993). The strengths of this approach include the ability to capture: (1) moment-to-moment counts of behaviors allowing for time series analyses, (2) counts of subcategories of behaviors, and (3) estimates of rates of behaviors. However, these methods require a great deal of time and labor resulting in limited efficiency and, as a result, are infrequently used in studies with large samples of children with ASD, such as phenotypic examinations within genetic investigations. These limitations have rendered it difficult to comprehensively assess the relation between JA and functional outcomes among large samples of children with ASD and to generalize findings. Methods that would enable a more efficient examination of JA may also help facilitate large scale studies aimed at understanding the genetic underpinnings of social attention deficits.

Given the high frequency of JA impairment among children with ASD, items assessing the amount or quality of both initiating and responding to JA (IJA or RJA, respectively) have been incorporated into ASD specific diagnostic instruments (e.g., the Autism Diagnostic Observation Schedule [ADOS]; Lord, Rutter, DiLavore, & Risi, 2002). Extrapolating information about JA ability from widely used measures such as the ADOS enables researchers to quantify JA in large samples of children with ASD using relatively minimal resources. Such a method would potentially help to advance ASD research across genetic and epidemiological domains. Several studies that have used factor analysis to examine ADOS subfactors have revealed an additional JA factor (Gotham et al., 2008; Gotham, Risi, Pickles, & Lord, 2007; Oosterling et al., 2010; Robertson, Tanguay, Lecuyer, Sims, & Waltrip, 1999). The JA factor identified by Gotham et al. (2007, 2008) and replicated by (Oosterling et al., 2010) was comprised of a range of discrete social behaviors including JA but also including behavioral requesting (i.e., pointing, response to joint attention, gesturing, showing, initiation of joint attention and unusual eye contact). Even more broad, the JA factor identified by Robertson et al. (1999) was comprised of items assessing overall social functioning (i.e., quality of social overtures, quality of social responses). Both of these studies are an important step forward in identifying subfactors that may make quantifying JA in larger samples more feasible; however, a JA factor containing only items that explicitly quantify JA would ensure researchers are specifically assessing these abilities rather than quantifying socialization more broadly.

Recent research has begun to investigate a more accurate JA composite derived from the ADOS (Maljaars, Noens, Scholte, & Berckelaer-Onnes, 2012; Thurm, Lord, Lee, & Newschaffer, 2007), including only items designed specifically to quantify JA ability (e.g., ADOS items labeled “Spontaneous Initiation of Joint Attention” and “Response to Joint Attention”). Although these two studies much more precisely capture the construct of JA by narrowing the ADOS items included in the composite, they are limited by the fact that they do not include the ADOS item assessing protodeclarative showing, a classic IJA behavior (Bruinsma et al., 2004), and one that is included as part of standard assessments of JA (Seibert et al., 1982; Wetherby & Prizant, 1993). Future studies using a similar ADOS-derived JA composite would benefit from including all three JA specific ADOS items (e.g., ADOS items labeled “Spontaneous IJA”, “Response to JA”, and “Showing”).

## 1.2. Joint attention in relation to adaptive and cognitive abilities

Adaptive abilities describe how an individual functions in everyday life, typically in areas of communication (both expressive and receptive), socialization, and daily living or life skills. Although individuals with ASD have a distinct pattern of adaptive functioning compared to typically developing and developmentally delayed peers (Carter et al., 1998) and research demonstrates a relation between autism symptomatology and adaptive functioning (Liss et al., 2001), minimal research has examined the relation between specific pivotal symptoms of ASD, such as JA, and functional outcomes. Within this small literature base, some research finds evidence to support a relation between JA and adaptive communication among infants with ASD (Poon, Watson, Baranek, & Poe, 2012), whereas other longitudinal research does not (Gillespie-Lynch et al., 2012). While Mundy, Sigman, and Kasari (1994) examined the relation between JA and parent reported socialization more broadly using a behavior problem focused measure (the Aberrant Behavior Checklist), Gillespie-Lynch et al. (2012) are the only known group to examine the relation between JA and both adaptive socialization and daily living skills. Yet, this study is limited by its small sample size, as it included only 20 individuals with ASD, warranting replication. As such, ambiguity remains as to the relation between JA and functional adaptive outcomes such as communication, socialization, and daily living skills. Importantly, while Poon et al. (2012) and Mundy et al. (1994) examined this relation in early childhood and Gillespie-Lynch et al. (2012) examined adults, there has been limited research documenting the relation between JA and functional outcomes among school-aged children.

In addition to examining functional outcomes such as language and adaptive skills, the supposition that social impairment may be tied to cognitive functioning (Mundy et al., 1994) calls for a more thorough understanding of the relation between JA and cognitive ability. The majority of studies have only indirectly investigated this relation by examining how IQ and JA differentially predict language ability (Anderson et al., 2007; Mundy et al., 1990). The one study reported in the literature examining the direct relation between JA and IQ in children with ASD ( $n = 30$ ) dichotomized overall IQ ability, comparing children with mild intellectual disability to those with moderate/severe intellectual disability (Mundy et al., 1994). The authors demonstrated that cognitive impairment was related to greater deficits in low-level JA behaviors (such as shift in eye gaze), but not high-level JA behaviors (such as social pointing or showing), in individuals with moderate to severe intellectual disability. No

relation was observed between cognitive scores and JA among individuals with mild intellectual disability.

Although this initial investigation of the relation between cognitive ability and JA has great merit, future research with increased power is needed to further examine this relation among individuals with a range of cognitive abilities. This somewhat ambiguous finding might also result from examining cognition as a unitary construct rather than examining verbal and non-verbal subfactors of cognition (Mundy et al., 1994). Given the specific links between JA and language ability (e.g., Mundy & Gomes, 1998; Mundy et al., 1990; Wetherby & Prizant, 1993), assessing whether JA differentially relates to verbal IQ (VIQ) and non-verbal IQ (NVIQ) may help to fully understand the intersection between cognition and socialization. Further, research shows that IQ accounts for a large amount of variance in adaptive skills among school-age children (Kanne et al., 2011); therefore, investigating whether the relation between JA and functional outcomes (e.g., language and adaptive social functioning) is mediated by cognitive functioning may be of import when trying to understand how early social impairments contribute to other functional outcomes in ASD.

### 1.3. Current investigation

The current study sought to examine how an ADOS-derived JA composite relates to cognitive and adaptive functional outcomes within a large cross-sectional sample of children diagnosed with ASD. Although there has been minimal research investigating the association between cognitive ability and JA, as a result of the clearly demonstrated relation between language and JA, we hypothesized that JA would be more highly related to VIQ than NVIQ. In addition, we anticipated JA would be significantly associated with adaptive communication. As a core social deficit, we also hypothesized that JA would be associated with adaptive socialization, but less highly so with daily living skills, given past research and the limited overlap between social ability and the ability to perform self-care routines. We also sought to determine whether JA accounted for the variability in adaptive functioning after controlling for cognition as well as whether the relation between JA and adaptive functioning was differentially mediated by VIQ as compared to NVIQ. Finally, we also examined whether VIQ and NVIQ moderated the relation between JA and adaptive functioning.

## 2. Method

### 2.1. Participants

Participants included in this analysis were 1061 children from simplex families included in the Simons Simplex Collection (SSC). The SSC is a database designed to characterize phenotypic and genotypic information of individuals diagnosed with ASD in the United States. Individuals included in the SSC database met the following inclusion criteria: (1) surpassed clinical cutoffs for autism or autism spectrum on either the Autism Diagnostic Observation Schedule (ADOS; Lord, 2002) or the Autism Diagnostic Interview-Revised (ADI-R; Rutter, Le Couteur, & Lord, 2003), (2) were between the ages of 4 and 18 years, and (3) had a minimum nonverbal cognitive level required to participate in the testing battery (Fischbach & Lord, 2010). Participants were excluded from the SSC database based on a

range of predefined medical conditions or complications (Fischbach & Lord, 2010). We calculated a composite JA score from ADOS modules 1 or 2, which are administered to children who have not yet achieved fluent speech. As a result, only participants that completed either of these two modules were included in the current study (40% of the original sample). Probands included in this study had a mean age of 7.57 (SD = 3.34) years and were 84% male. Of this sample, 65% identified as Caucasian, 18% as Biracial, 7% as Asian, 6% as African American, and 2% or less identified as Hispanic, Native Hawaiian, Native American, or other.

## 2.2. Measures

JA scores were calculated using specific items from the ADOS modules 1 and 2 because only these two modules contain items that are operationally defined to specifically assess JA. More explicitly, in modules 3 and 4, JA behaviors are included under items assessing socialization more broadly (e.g., “Amount and Quality of Social Overtures”). A goal of this study was to include all module 1 and 2 items that explicitly operationalized JA behaviors. From module 1 the JA composite was the sum of Spontaneous IJA (item B10), Response to JA (item B11), and Showing (item B9). Likewise, from Module 2 the JA composite was the sum of Spontaneous IJA (item B6) and Response to JA (item B7), and Showing (item B5). Possible scores on items ranged from 0 to 2 (raw scores were converted to algorithm scores) and total JA composite scores ranged from 0 to 6. By including the protodeclarative showing item in addition to the ADOS items labeled RJA & IJA, this study examined a JA composite that included all ADOS items specifically assessing JA ability. The JA composite excluded items that were not operationally specific to JA, such as “quality and quantity of social overtures and responding”, to ensure the specificity of the score. Although this approach to assessing JA has not established formal reliability and validity, the coding of the ADOS itself has demonstrated excellent psychometric properties (Gotham et al., 2008; Lord et al., 2002) and previous studies have set the precedent for using such a composite (Maljaars et al., 2012; Thurm et al., 2007).

In this study, cognitive ability was assessed using one of two measures: the Differential Ability Scales – Second Edition (DAS-II; Elliott, 2007) or the Mullen Scales of Early Learning (Mullen, 1995). For whichever of the tests was administered, standard scores of verbal and non-verbal ability were generated by looking up norm-referenced scores in the corresponding test manuals (Fischbach & Lord, 2010). Although these measures more accurately assess cognitive ability, for ease of discussion, we will refer to the scores on these assessments as IQ scores. Both the DAS-II and the Mullen have established excellent reliability and validity (Bradley-Johnson, 2001; Elliott, 2012; Keith, Low, Reynolds, Patel, & Ridley, 2010) and these measures have demonstrated convergent validity (Bishop, Guthrie, Coffing, & Lord, 2011).

Additionally, parents of probands completed the Vineland Adaptive Behavior Scales, Second Edition, Survey Interview Form (VABS-II; Sparrow, Cicchetti, & Ball, 2005), a widely used semi-structured interview designed to measure adaptive behavior in children and adolescents. The VABS-II focuses on domains of Communication (Expressive, Receptive,

and Written), Socialization (Interpersonal Relationships, Play and Leisure Time, and Coping Skills), and Daily Living Skills (Personal, Domestic, and Community).

Parents of probands completed the Social Responsiveness Scale (SRS) to assess for social abilities in a range of areas including social awareness, social information processing. These references are missing the authors and align with the references added for Carter et al., (1998) and Fischbach & Lord (2010). The references missing the authors were removed from the reference list when the correct ones were added. reciprocal communication, and social avoidance (Constantino & Gruber, 2005). The SRS is a 65 item, Likert response scale where each item is rated from 1 (not true) to 4 (almost always true). The SRS is commonly used to assess social impairment among children with ASD through the generation of an overall score of autistic social impairment (SRS\_Total). Furthermore, the SRS has demonstrated excellent reliability and validity and is moderately correlated with the Autism Diagnostic Interview-Revised (ADI-R) and the ADOS (Bölte, Westerwald, Holtmann, Freitag, & Poustka, 2011; Constantino & Gruber, 2005).

### 3. Data analysis

First, we ran basic statistical analyses to obtain the general description of the dataset. To understand how JA relates to other functional domains often impaired among children with ASD, we also ran correlation analyses to investigate the relationships among the variables of interest. In particular, correlational analyses assessed the relation between JA and measures of VIQ and NVIQ, and adaptive functioning in the domains of communication (VABS\_Com), socialization (VABS\_Soc), and daily living (VABS\_DL). Considering the data collected included missing values, we used pair-wise deletion correlation analysis, which is more robust than the conventional list-wise deletion analysis (Little & Rubin, 2002). The conventional list-wise analysis deletes all the data for the participants who have missing values, resulting in a smaller sample size used for calculating the correlation matrix than the original N, and ultimately resulting in lower power. In contrast, the pair-wise analysis only deletes the data pairs with missing values. With pair-wise analysis, although the sample size used for each correlation might be different, the total sample size for calculating the correlation matrix remains the original N. To ensure the current study was specifically assessing JA and not socialization more generally, the total SRS score, which significantly correlated with the ADOS JA subdomain ( $r = 0.22, p < 0.001$ ), was included as a controlling variable in partial correlation analyses.

Next, to further understand the detailed relation between JA and adaptive ability, we conducted a series of SEM analyses. We used the maximum likelihood (ML) estimation method for these analyses so that all available information in the dataset was used. The first SEM analysis used multivariate regression to examine whether adaptive abilities were significantly associated with JA after controlling for the total SRS score (see Fig. 1). To further understand the relation between JA and adaptive ability, we ran a second SEM model to examine whether cognition was a potential mediator of these relations. This analysis was run in accordance with established mediation procedures (Baron & Kenny, 1986). Specifically, analyses were run to investigate whether each domain of cognitive functioning (VIQ or NVIQ) mediated the relation between JA and the three domains of adaptive

functioning: communication (VABS\_Com), socialization (VABS\_Soc), and daily living (VABS\_DL). We conducted the analysis separately for VIQ and NVIQ in order to distinguish their mediation effects. Fig. 2(a) and (b) illustrates the path diagrams of the mediation models 1 and 2, respectively. Model 1 focused on NVIQ and model 2 focused on VIQ. In both models, the total SRS score was included as a control variable in the analyses to account for the overall level of social impairment, thereby ensuring that observed relations were not an artifact of overall social symptom severity. Finally, we ran a third SEM model to investigate whether VIQ or NVIQ moderated the effect of JA on VABS\_Com, VABS\_Soc, and VABS\_DL.

## 4. Results

### 4.1. Preliminary analyses

A description of the functional outcomes of this sample of children with ASD is reported in Table 1. Table 2 presents sample descriptive statistics by each ADOS module. We compared participants completing module 1 and module 2 and discovered the two groups different significantly in terms of evaluation age,  $t = 3.30$ ,  $p = 0.001$ , total ADOS score,  $t = 9.44$ ,  $p < 0.001$ , JA subfactor score,  $t = 16.56$ ,  $p < 0.001$ , and SRS total,  $t = 8.22$ ,  $p < 0.001$ , but not gender,  $t = 0.39$ ,  $p = 0.70$ . Pearson correlation coefficients depicting the relation between JA and all other study variables as shown in Table 3 reveal JA was significantly negatively correlated with all measures of cognitive and adaptive abilities (VIQ, NVIQ, VABS\_Com, VABS\_Soc, VABS\_DL), such that greater impairment in JA as reflected by higher scores on the JA composite was related to greater impairment across all functional outcomes. Even after controlling for overall socialization using the total SRS score, the JA composite score remained significantly negatively correlated with all cognitive and adaptive study variables. In essence, lower amounts of JA (reflected by higher scores) were correlated with lower cognitive and adaptive scores. JA was not significantly correlated with ethnicity ( $r = -0.005$ ,  $p = 0.878$ ) but was significantly correlated with gender ( $r = -0.072$ ,  $p = 0.020$ ).

### 4.2. Multivariate regression and mediation analyses

Before mediation analyses were conducted, we ran multivariate regression models to examine the relation between JA and adaptive ability (see Fig. 1). These pathways of interest have been bolded below in Tables 4–6 for ease of interpretation. Although this step is not necessary for mediation analysis, considering cases with suppressor variables, we used this analysis to help fully understand the association between JA and adaptive abilities. Table 4 shows that after controlling for the total SRS score, JA was significantly negatively associated with adaptive socialization (JA  $\rightarrow$  VABS\_Soc:  $B = -0.250$ ,  $p < 0.001$ ), adaptive communication (JA  $\rightarrow$  VABS\_Com:  $B = -0.275$ ,  $p < 0.001$ ), and adaptive daily living ability (JA  $\rightarrow$  VABS\_DL:  $B = -0.240$ ,  $p < 0.001$ ). By analyzing the relation between JA and VIQ & NVIQ, we also found that JA was significantly negatively associated with VIQ (JA  $\rightarrow$  VIQ:  $B = -0.371$ ,  $p < 0.001$ ) and NVIQ (JA  $\rightarrow$  NVIQ:  $B = -0.306$ ,  $p < 0.001$ ).

Given the overall relation between JA and adaptive abilities, mediation models 1 and 2 (see Fig. 2) were then conducted to examine the potential mediation effects of VIQ and NVIQ. In mediation model 1, we focused on whether VIQ mediated the relation between JA and



adaptive abilities. It was observed (see Table 5) that the indirect effect of JA (JA  $\rightarrow$  VIQ  $\rightarrow$  VABS component) was significant but all of the direct effects of JA (JA  $\rightarrow$  VABS component) were non-significant (JA  $\rightarrow$  VABS\_Soc,  $p = 0.151$ , JA  $\rightarrow$  VABS\_Com,  $p = 0.226$ , and JA  $\rightarrow$  VABS\_DL,  $p = 0.496$ ); therefore, VIQ significantly mediated the relation between JA and adaptive abilities. Regarding the mediating effect of NVIQ on adaptive abilities, data were analyzed based on mediation model 2. It was determined (see Table 5) that NVIQ only partially mediated the relation between JA and each adaptive ability, because both the indirect effect of JA (JA  $\rightarrow$  NVIQ  $\rightarrow$  VABS component) and the direct effect of JA (JA  $\rightarrow$  VABS component) were significant (JA  $\rightarrow$  VABS\_Soc,  $p < 0.001$ , JA  $\rightarrow$  VABS\_Com,  $p = 0.002$ , and JA  $\rightarrow$  VABS\_DL,  $p = 0.007$ ).

### 4.3. Moderation analyses

Given the significant mediation effects of the relation between JA and adaptive abilities, moderation models 1 and 2 were also conducted to examine the moderation effect of cognitive ability (VIQ or NVIQ) on JA and adaptive abilities. Fig. 3(a) and (b) shows the plots of model 1 and model 2, respectively. We also investigated the moderation effect of gender and Fig. 3 (c) shows the plot. In model 1, VIQ and the interaction between VIQ and JA were added to explain the variance in adaptive abilities. The top part of Table 6 lists the estimation results. We observed that the interaction effect between JA and VIQ on VABS\_Com was significant ( $p = 0.009$ ); however, the effect was not significant for the other two interaction terms (JA\*VIQ  $\rightarrow$  VABS\_Soc;  $p = 0.379$  and JA\*VIQ  $\rightarrow$  VABS\_DL;  $p = 0.345$ ), which indicates that different levels of VIQ significantly changed the effect of JA on VABS\_Com but not on VABS\_Soc and VABS\_DL. The positive coefficient estimate (0.108) means with the higher VIQ level, the effect of JA on VABS\_Com was stronger; therefore, VIQ significantly positively moderated the relation between JA and the adaptive ability of VABS\_Com. The middle part of Table 6 lists the analysis results for moderation model 2 examining the moderation effect of NVIQ. None of the interaction terms between JA and NVIQ were significant (JA\*NVIQ  $\rightarrow$  VABS\_Com;  $p = 0.211$ , JA\*NVIQ  $\rightarrow$  VABS\_Soc;  $p = 0.256$ , and JA\*NVIQ  $\rightarrow$  VABS\_DL;  $p = 0.560$ ), which means NVIQ did not significantly moderate the effect of JA on all adaptive abilities.

The bottom part of Table 6 lists the moderation analysis results for gender. As for NVIQ, although all the three interaction estimates are positive, none of the interaction terms between JA and gender were significant (JA\*Gender  $\rightarrow$  VABS\_Com,  $p = 0.262$ ; JA\*Gender  $\rightarrow$  VABS\_Soc,  $p = 0.174$ ; JA\*Gender  $\rightarrow$  VABS\_DL,  $p = 0.362$ ); therefore, gender did not significantly moderate the effect of JA on the three adaptive abilities examined. In summary, based on the data we analyzed and the models we considered in this article, only VIQ significantly positively moderated the effect between JA and adaptive communication (VABS\_Com).

## 5. Discussion

Theorists posit that impairments in specific social behaviors, such as JA, contribute to a cascade of pervasive and pronounced social deficits later in development (Klin, Jones, Schultz, Volkmar, & Cohen, 2002) and there is substantial evidence to demonstrate the

negative downstream effects of JA deficits on language development (e.g., Mundy & Gomes, 1998; Mundy et al., 1990; Wetherby & Prizant, 1993). Yet, the majority of research examines early childhood and does not investigate whether the negative impact of JA impairments persists into late childhood. The current study used an ADOS-derived JA composite score to investigate the relation between JA and both, adaptive functioning and cognition. However, unlike previous studies involving young children, this study was conducted in a large sample of school-aged children diagnosed with ASD. Importantly, the use of such a JA composite may serve as a model for expanding research on JA to larger samples through increased feasibility. Contrary to our hypotheses we did not find that JA was differentially related to subdomains of either cognitive or adaptive functioning. Instead, this study found that JA was significantly related to both VIQ and NVIQ, as well as significantly associated with all 3 domains of adaptive functioning (Socialization, Communication, and Daily Living). Given that cognitive, social, and adaptive abilities are often interwoven (e.g., Liss et al., 2001), additional analyses were conducted to examine whether VIQ or NVIQ mediated the relation between JA and adaptive abilities. We observed that VIQ fully mediated the relation between JA and all three adaptive domains but that NVIQ only partially mediated this association.

This research demonstrates that JA deficits may have collateral effects not only on language and other basic social behaviors (e.g., imitation and social orientation), but may also have more pervasive consequences in terms of broader functional, cognitive and adaptive domains. Within a large and diverse sample of children with ASD, this study replicated previous research demonstrating a positive relation between JA and cognitive functioning (Mundy et al., 1994). More specifically this study demonstrated that JA was related to both subdomains of cognitive functioning, VIQ and NVIQ. Given that the relation between JA and adaptive functioning has received only minimal attention and heterogeneity exists within published research findings, this study further examined this relation. Previous research among a small sample of individuals with ASD found JA was related to only some aspects of adaptive functioning (e.g., Socialization but not Communication or Daily Living Skills; Gillespie-Lynch et al., 2012). Poon et al. (2012) found a significant relation between JA and adaptive Communication but did not assess Socialization or Daily Living subdomains. The large sample size of the current study and observed relation between JA and all three adaptive subdomains may help to bring some clarity to this body of research and indicates that the negative impact of JA deficits commonly observed in infancy is also present in school-aged children.

In addition to examining the relation between JA and subgroups of adaptive functioning and cognition, we also assessed whether the relation between JA and adaptive skills was mediated by either VIQ or NVIQ. A better understanding of the pathway by which JA impairments contribute to functional impairments may have important implications in tailoring specific interventions to specific groups. For example, whether this social disruption emerges differently in high- and low-functioning subgroups of individuals with ASD. We found that cognition did in fact mediate the relation between JA and all three subdomains of adaptive functioning, but that this was specific to VIQ, and not NVIQ. In line with research supporting the link between JA and language development, this study provides additional evidence to support that JA also leads to adaptive impairment across domains

through a particular verbal cognitive pathway. One hypothesis is that the mediation of the relation between JA and adaptive skills by VIQ is the result of JA being an essential prerequisite skill needed for the development of cognitive linguistic skills in addition to basic language ability. When examining the moderating effects of NVIQ and VIQ, we again found that VIQ, but not NVIQ, impacted the relation between JA and adaptive skills, but for adaptive communication only.

The use of an ADOS-derived JA composite in this study enhances the existing literature in two meaningful ways. First, this study provides guidance for strengthening a previously used ADOS-derived metric of JA ability (Maljaars et al., 2012; Thurm et al., 2007) by recommending that the composite contain all items from the ADOS explicitly assessing JA (e.g., Showing in addition to RJA and IJA items). Second, by deriving JA ability from the widely administered ADOS, it is possible to investigate the relation between JA and multiple other phenotypes among a broad range of individuals with ASD and increase the strength of the phenotypic component of genetic investigations of ASD. Further, the increased power as a result of using this ADOS-derived JA measure increases the confidence with which we report the current findings. There has been variability in JA factors used across different studies, from overly inclusive (Gotham et al., 2007, 2008; Oosterling et al., 2010) to potentially slightly narrow (Maljaars et al., 2012; Thurm et al., 2007). This variability may impact the pattern of results observed, but it can be remedied by including only items that adhere specifically to the operational definition of JA and by ensuring that all valid items in the ADOS are included in the composite.

Although the SSC data set utilized in this study provides access to phenotypic information for a large number of children, it is important to note that the data were not collected specifically for this study. As a result, although this dataset allows us to measure JA globally within a large sample, other measures designed to assess JA more rigorously were not available. Further, the brevity of the ADOS-derived JA composite score utilized in this study precludes the investigation of IJA and RJA behaviors independently. Given that many past studies have utilized measures such as the Early Social Communication Scales (ESCS; Mundy et al., 1986; Seibert et al., 1982) or the Communication and Symbolic Behavior Scales (CSBS; Wetherby & Prizant, 1993), it may be difficult to compare results from these studies to the current study. As such, future research should investigate the relation between the ADOS-derived JA composite utilized in this study and more commonly used, yet more time-intensive, measures of JA. One other limitation of the ADOS-derived JA composite is that it can only be generated from ADOS modules 1 and 2, thereby limiting its use to primarily non-verbal samples and limiting its generalizability. Finally, it is important to note the cross-sectional nature of the data at present. Given that language and socialization develop on a continuum, longitudinal designs will help to unequivocally identify whether these relations are uni- or bi-directional.

In sum, by using a relatively novel measure of JA ability to assess a large group of children diagnosed with ASD, the current study provides further evidence to show how JA skills are interwoven with cognitive, linguistic, and adaptive functioning. A greater appreciation of how JA relates to functional outcomes among individuals with ASD may help to provide insight into the heterogeneous impairment observed among individuals with ASD as well as

the mechanisms by which this impairment develops. Further, by understanding how JA differentially relates to functional outcomes and the potential mediators of these relations, it may be possible to tailor interventions to specific groups of individuals. Knowledge about how JA relates to functional outcomes may help to explain the positive collateral impact JA specific interventions have on social development at large. As a whole, this investigation provides a wealth of knowledge concerning how to tailor JA interventions, which populations to target, and how to measure the outcome. It also furthers our understanding of the heterogeneous patterns of cognitive, linguistic, and adaptive functioning seen in children with ASD.

## Acknowledgements

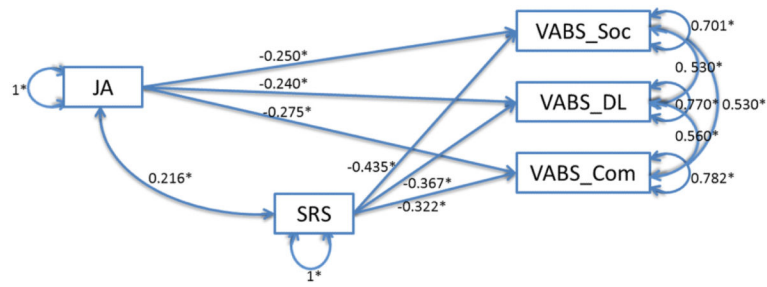
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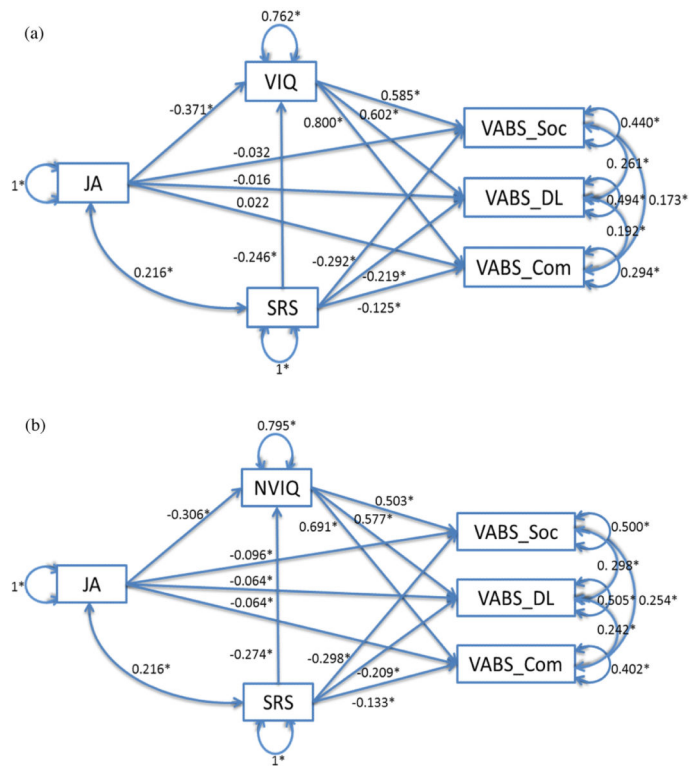
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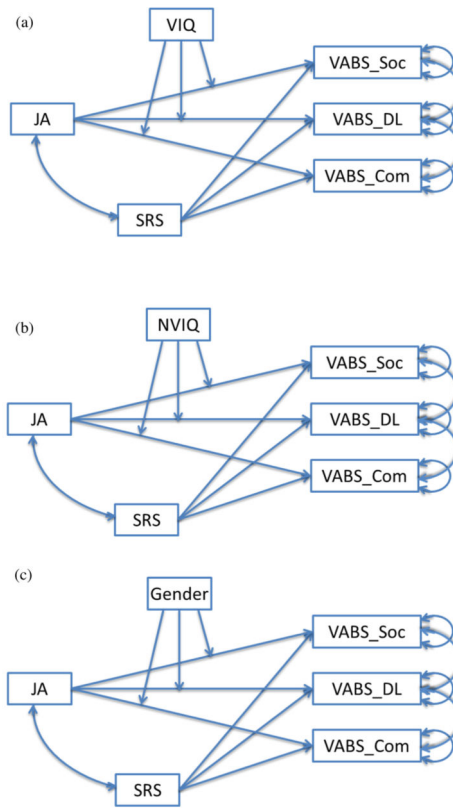


**Fig. 1.** Path diagrams of the multivariate regression depicting how the functional outcomes are associated with JA after controlling for the total SRS score.



**Fig. 2.** Path diagrams of the mediation analysis depicting how the relation between JA and functional outcomes is mediated by cognitive ability (VIQ & NVIQ) while controlling for the total SRS score. (a) Path diagram of the model testing the mediation effect of VIQ. (b) Path diagram of the model testing the mediation effect of NVIQ.





**Fig. 3.** Path diagrams of the moderation analysis depicting how the relation between JA and functional outcomes is moderated by cognitive ability (VIQ & NVIQ) and gender while controlling for the total SRS score. (a) Path diagram of the model testing the moderation effect of VIQ. (b) Path diagram of the model testing the moderation effect of NVIQ. (c) Path diagram of the model testing the moderation effect of Gender.

**Table 1**

Sample Functional Outcome Descriptives.

	<b>Mean</b>	<b>SD</b>
SRS_Total	81.62	9.28
VIQ	53.45	27.34
NVIQ	66.88	25.49
VABS_Com	70.12	15.45
VABS_Socialization	65.52	12.76
VABS_DailyLiving	69.43	13.05

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**Table 2**

Descriptive Statistics by ADOS Module.

	<b>Module 1</b>			<b>Module 2</b>		
	<b>Mean</b>	<b>SD</b>	<b>Range</b>	<b>Mean</b>	<b>SD</b>	<b>Range</b>
Age (months)	95.27	41.8	48–214	87.11	38.1	48–215
JA Factor Score	3.93	1.33	0–7	2.58	1.31	0–6
ADOS Total Score	19.63	3.93	8–28	17.11	4.78	7–28
SRS Total Score	110.25	23.64	43–177	97.56	25.94	13–169
Gender	84.5% (male)	15.5% (female)		83.7% (male)	16.3% (female)	

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**Table 3**

Pearson correlation coefficients depicting the relation of all study variables.

	1.	2.	3	4.	5.	6.	7.	8.	9.	10.	11.
1. JA	1.00										
2. Verbal IQ	-0.43**	1.00									
3. Non-verbal IQ	-0.36**	0.83**	1.00								
4. VABS Soc	-0.34**	0.70**	0.64**	1.00							
5. VABS Comm	-0.35**	0.83**	0.76**	0.78**	1.00						
6. VABS Daily Living	-0.32**	0.68**	0.67**	0.79**	0.78**	1.00					
7. SRS Total	0.22**	-0.35**	-0.37**	-0.53**	-0.42**	-0.46**	1.00				
8. ADOS Total	0.66**	-0.43**	-0.38**	-0.39**	-0.36**	-0.34**	0.27**	1.00			
9. Gender	-0.03	-0.01	-0.06*	-0.03	-0.03	-0.03	0.03	0.02	1.00		
10. Ethnicity	-0.01	0.04	0.04	0.01	-0.01	-0.01	-0.01	-0.06	-0.05	1.00	
11. Age (mos)	0.10**	-0.56**	-0.45**	-0.56**	-0.55**	-0.45**	0.25**	0.23**	0.05	-0.06	1.00

\* Significant at the 0.05 level.

\*\* Significant at the 0.01 level.

**Table 4**

Multivariate regression analysis results testing the relation between JA and the three components of VABS while controlling for the overall SRS\_total.

			Beta (Std. Estimate)	Estimate	Std. Error	<i>t</i> _ratio	<i>p</i>
VABS_Soc	←	JA	-0.250	-2.205	0.232	-9.490	0.000
VABS_Com	←	JA	-0.275	-2.938	0.297	-9.883	0.000
VABS_DL	←	JA	-0.240	-2.165	0.249	-8.691	0.000
VABS_Soc	←	SRS	-0.435	-0.599	0.036	-16.535	0.000
VABS_Com	←	SRS	-0.322	-0.536	0.046	-11.576	0.000
VABS_DL	←	SRS	-0.367	-0.516	0.039	-13.292	0.000
JA	↔	JA	1.000	2.090	0.091	23.022	0.000
SRS	↔	SRS	1.000	86.023	3.737	23.022	0.000
SRS	↔	JA	0.216	2.895	0.421	6.871	0.000
VABS_Soc	↔	VABS_Soc	0.701	114.069	4.955	23.022	0.000
VABS_Com	↔	VABS_Com	0.782	186.707	8.110	23.022	0.000
VABS_DL	↔	VABS_DL	0.770	131.102	5.695	23.022	0.000
VABS_DL	↔	VABS_Soc	0.530	88.098	4.629	19.031	0.000
VABS_DL	↔	VABS_Com	0.559	112.769	5.924	19.037	0.000
VABS_Soc	↔	VABS_Com	0.530	104.468	5.513	18.951	0.000

**Table 5**

Mediation analysis results testing the mediation effects of VIQ and NVIQ between JA and the three components of VABS while controlling for the overall SRS\_total score.

		Beta (Std. Estimate)	Estimate	Std. Error	t_ratio	p
Mediation Model 1: Testing the mediation effect of VIQ						
VIQ	← JA	-0.371	-7.025	0.519	-13.529	0.000
VABS_Soc	← JA	<b>0.032</b>	<b>-0.287</b>	<b>0.200</b>	<b>-1.437</b>	<b>0.151</b>
VABS_Com	← JA	<b>0.022</b>	<b>0.239</b>	<b>0.197</b>	<b>1.210</b>	<b>0.226</b>
VABS_DL	← JA	<b>-0.016</b>	<b>-0.147</b>	<b>0.216</b>	<b>-0.680</b>	<b>0.496</b>
VABS_Com	← VIQ	0.800	0.452	0.011	41.935	0.000
VABS_Soc	← VIQ	0.585	0.273	0.011	25.084	0.000
VABS_DL	← VIQ	0.602	0.287	0.012	24.340	0.000
VIQ	← SRS	-0.246	-0.725	0.081	-8.955	0.000
VABS_Soc	← SRS	-0.292	-0.401	0.030	-13.473	0.000
VABS_Com	← SRS	-0.125	-0.209	0.029	-7.079	0.000
VABS_DL	← SRS	-0.219	-0.308	0.032	-9.547	0.000
JA	↔ JA	1.000	2.090	0.091	23.022	0.000
SRS	↔ SRS	1.000	86.023	3.737	23.022	0.000
SRS	↔ JA	0.216	2.895	0.421	6.871	0.000
VIQ	↔ VIQ	0.762	569.529	24.739	23.022	0.000
VABS_Soc	↔ VABS_Soc	0.440	71.580	3.110	23.022	0.000
VABS_Com	↔ VABS_Com	0.294	70.216	3.050	23.022	0.000
VABS_DL	↔ VABS_DL	0.494	84.099	3.653	23.022	0.000
VABS_DL	↔ VABS_Soc	0.261	43.409	2.731	15.897	0.000
VABS_DL	↔ VABS_Com	0.192	38.772	2.644	14.666	0.000
VABS_Soc	↔ VABS_Com	0.173	34.115	2.417	14.118	0.000
Mediation Model 2: Testing the mediation effect of NVIQ						
NVIQ	← JA	-0.306	-5.388	0.495	-10.894	0.000
VABS_Soc	← JA	<b>-0.096</b>	<b>-0.849</b>	<b>0.207</b>	<b>-4.103</b>	<b>0.000</b>
VABS_Com	← JA	<b>-0.064</b>	<b>-0.680</b>	<b>0.225</b>	<b>-3.027</b>	<b>0.002</b>
VABS_DL	← JA	<b>-0.064</b>	<b>-0.574</b>	<b>0.213</b>	<b>-2.695</b>	<b>0.007</b>
VABS_Com	← NVIQ	0.691	0.419	0.013	31.660	0.000
VABS_Soc	← NVIQ	0.503	0.252	0.012	20.656	0.000
VABS_DL	← NVIQ	0.577	0.295	0.013	23.571	0.000
NVIQ	← SRS	-0.274	-0.753	0.077	-9.762	0.000
VABS_Soc	← SRS	-0.298	-0.410	0.032	-12.825	0.000
VABS_Com	← SRS	-0.133	-0.221	0.035	-6.375	0.000
VABS_DL	← SRS	-0.209	-0.294	0.033	-8.950	0.000
JA	↔ JA	1.000	2.090	0.091	23.022	0.000
SRS	↔ SRS	1.000	86.023	3.737	23.022	0.000
SRS	↔ JA	0.216	2.895	0.421	6.871	0.000
NVIQ	↔ NVIQ	0.795	516.632	22.441	23.022	0.000

			<b>Beta (Std. Estimate)</b>	<b>Estimate</b>	<b>Std. Error</b>	<b>t_ratio</b>	<b>p</b>
VABS_Soc	↔	VABS_Soc	0.500	81.331	3.532	23.022	0.000
VABS_Com	↔	VABS_Com	0.402	95.963	4.168	23.022	0.000
VABS_DL	↔	VABS_DL	0.505	86.017	3.736	23.022	0.000
VABS_DL	↔	VABS_Soc	0.298	49.680	2.988	16.626	0.000
VABS_DL	↔	VABS_Com	0.242	48.807	3.168	15.407	0.000
VABS_Soc	↔	VABS_Com	0.254	49.964	3.117	16.027	0.000

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**Table 6**

Results of testing the moderation effects of VIQ and NVIQ on the relation between JA and the three components of VABS while controlling for the overall SRS\_total score.

		Beta (Std. Estimate)	Estimate	Std. Error	t_ratio	p
Moderation Model 1: Testing the moderation effect of VIQ						
vabs_Soc	← JA	0.003	0.029	0.410	0.070	0.944
vabs_Com	← JA	-0.064	-0.689	0.405	-1.701	0.089
vabs_DL	← JA	-0.057	-0.514	0.445	-1.155	0.248
vabs_Com	← VIQ	0.703	0.397	0.024	16.831	0.000
vabs_Soc	← VIQ	0.625	0.292	0.024	12.212	0.000
vabs_DL	← VIQ	0.556	0.266	0.026	10.252	0.000
<b>vabs_Com</b>	<b>← JA*VIQ</b>	<b>0.108</b>	<b>0.018</b>	<b>0.007</b>	<b>2.620</b>	<b>0.009</b>
<b>vabs_Soc</b>	<b>← JA*VIQ</b>	<b>-0.045</b>	<b>-0.006</b>	<b>0.007</b>	<b>-0.879</b>	<b>0.379</b>
<b>vabs_DL</b>	<b>← JA*VIQ</b>	<b>0.051</b>	<b>0.007</b>	<b>0.007</b>	<b>0.943</b>	<b>0.345</b>
vabs_Soc	← SRS	-0.292	-0.401	0.030	-13.483	0.000
vabs_Com	← SRS	-0.125	-0.208	0.029	-7.088	0.000
vabs_DL	← SRS	-0.219	-0.308	0.032	-9.545	0.000
Moderation Model 2: Testing the moderation effect of NVIQ						
vabs_Soc	← JA	-0.031	-0.269	0.551	-0.489	0.625
vabs_Com	← JA	-0.129	-1.374	0.599	-2.295	0.022
vabs_DL	← JA	-0.097	-0.880	0.567	-1.551	0.121
vabs_Com	← NVIQ	0.636	0.386	0.030	12.939	0.000
vabs_Soc	← NVIQ	0.559	0.280	0.027	10.189	0.000
vabs_DL	← NVIQ	0.548	0.281	0.028	9.940	0.000
<b>vabs_Com</b>	<b>← JA*NVIQ</b>	<b>0.072</b>	<b>0.011</b>	<b>0.008</b>	<b>1.250</b>	<b>0.211</b>
<b>vabs_Soc</b>	<b>← JA*NVIQ</b>	<b>-0.073</b>	<b>-0.009</b>	<b>0.008</b>	<b>-1.135</b>	<b>0.256</b>
<b>vabs_DL</b>	<b>← JA*NVIQ</b>	<b>0.038</b>	<b>0.005</b>	<b>0.008</b>	<b>0.582</b>	<b>0.560</b>
vabs_Soc	← SRS	-0.298	-0.409	0.032	-12.826	0.000
vabs_Com	← SRS	-0.133	-0.221	0.035	-6.386	0.000
vabs_DL	← SRS	-0.209	-0.294	0.033	-8.954	0.000
Moderation Model 3: Testing the moderation effect of gender						
vabs_Soc	← JA	-0.263	-2.317	0.253	-9.172	0.000
vabs_Com	← JA	-0.287	-3.068	0.324	-9.484	0.000
vabs_DL	← JA	-0.248	-2.243	0.271	-8.275	0.000
vabs_Com	← Gender	-0.051	-2.135	2.752	-0.776	0.438
vabs_Soc	← Gender	-0.040	-1.408	2.149	-0.655	0.512
vabs_DL	← Gender	-0.025	-0.877	2.306	-0.380	0.704
<b>vabs_Com</b>	<b>← JA*Gender</b>	<b>0.074</b>	<b>0.901</b>	<b>0.803</b>	<b>1.123</b>	<b>0.262</b>
<b>vabs_Soc</b>	<b>← JA*Gender</b>	<b>0.084</b>	<b>0.852</b>	<b>0.627</b>	<b>1.359</b>	<b>0.174</b>
<b>vabs_DL</b>	<b>← JA*Gender</b>	<b>0.059</b>	<b>0.613</b>	<b>0.673</b>	<b>0.911</b>	<b>0.362</b>
vabs_Soc	← SRS	-0.441	-0.607	0.037	-16.477	0.000
vabs_Com	← SRS	-0.324	-0.540	0.047	-11.448	0.000



			<b>Beta (Std. Estimate)</b>	<b>Estimate</b>	<b>Std. Error</b>	<b>t_ratio</b>	<b>p</b>
vabs_DL	←	SRS	-0.372	-0.523	0.040	-13.232	0.000

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