

Theory of Mind, Language and Adaptive Functioning in ASD: A Neuroconstructivist Perspective

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Abstract

Introduction: Neuroconstructivist theories of development highlight the potential effect one developmental domain may have on constraining or facilitating another. Empirical validation of this theory requires further testing in autism spectrum disorders (ASD), and may illuminate the complex interplay of developmental trajectories, particularly in the relationship between predictor and outcome variables. In ASD, language ability is an early predictor of important functional outcomes such as communication and socialization. We aimed to investigate whether theory of mind (ToM) mediates the relation between language ability and adaptive functioning in more cognitively able children with ASD (IQ > 70). **Methods:** Thirty-nine children were followed prospectively every two years from 4-6 years to 12-14 years. Their language and theory of mind abilities and adaptive functioning were tested using the Test of Language Development-2 (the independent variable, at age 6-8 years), the "Eyes Test" (a measure of ToM, the mediator, at age 10-12) and the Vineland Adaptive Behaviour Scales (the outcome variable, at age 12-14). **Results:** ToM mediated an association between language and adaptive functioning in the communication domain, but not in the social domain. **Conclusion:** These results challenge the usefulness of ToM as a unifying theory for ASD deficits and highlight the potential usefulness of a neuroconstructivist framework for prospective studies.

Key Words: autism, Asperger Syndrome, language, theory of mind, mediation

Résumé

Introduction: Les théories neuroconstructivistes du développement soulignent l'effet potentiel de restriction ou de facilitation qu'un domaine développemental peut exercer sur un autre domaine. La confirmation empirique de cette théorie exige plus de vérifications dans les troubles du spectre de l'autisme (TSA), et peut faire la lumière sur l'interaction complexe des trajectoires développementales, en particulier sur la relation entre les variables de prédiction et celles des résultats. Dans le TSA, l'aptitude linguistique est un prédicteur précoce d'importants résultats fonctionnels comme la communication et la socialisation. Nous visons à rechercher si la théorie de l'esprit (TdE) aide la relation entre l'aptitude linguistique et le fonctionnement adaptatif chez les enfants cognitivement aptes souffrant d'un TSA (IQ > 70). **Méthodes:** Trente-neuf enfants ont été suivis prospectivement tous les deux ans, de 4 à 6 ans jusqu'à 12 à 14 ans. Leurs aptitudes de langage et à la théorie de l'esprit ainsi que leur fonctionnement adaptatif ont été vérifiés à l'aide du test du développement du langage-2 (la variable indépendante, à l'âge de 6 à 8 ans), du « Eyes Test » (une mesure de la TdE, la médiatrice, à l'âge de 10 à 12 ans) et de l'Échelle de comportement adaptatif Vineland (la variable de résultat, à l'âge de 12 à 14 ans). **Résultats:** La TdE facilitait une association entre le langage et le fonctionnement adaptatif dans le domaine de la communication, mais pas dans le domaine social. **Conclusion:** Ces résultats remettent en question l'utilité de la TdE en tant que théorie unificatrice des déficits du TSA, et font valoir l'utilité potentielle d'un cadre neuroconstructiviste pour les études prospectives.

Mots clés: autisme, syndrome d'Asperger, langage, théorie de l'esprit, médiation

SUPPLEMENTARY ONLINE MATERIALS

http://www.cacap-acpea.org/uploads/documents/Theory_of_Mind_Bennett_2013_02.pdf

Online materials include: further information on participants; figure 1 and table 1 (Bivariate correlations of study variables); measures; time measures.

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Functional outcomes in autism spectrum disorders (ASD) differ significantly across individuals over time; many continue to require significant support in adulthood, whereas others progress toward independent living. Research into early predictors of outcomes has begun to explain this heterogeneity. For example, early language abilities are important indicators of later adaptive functioning - defined as practical abilities in social and communication skills and self-care (Bennett et al., 2008; Rutter, Greenfeld, & Lockyer, 1967; Sparrow, Balla, & Cicchetti, 1984; Szatmari, Bryson, Boyle, Streiner, & Duku, 2003; Szatmari et al. 2000; Venter, Lord, & Schopler, 1992). Identifying early predictors is an important task in longitudinal research, but “how” or “why” predictors such as early language ability are associated with variation in outcomes such as adaptive functioning is an important issue.

From a theoretical perspective, constraints or growth in one developmental domain may “spill over” to influence change across other important domains. For example, the additional burden of language impairment in young children with ASD may lead to further lags in their social understanding - there is evidence that verbal children with ASD rely more upon language to understand social tasks than do typically developing children (Fisher, Happé, & Dunn, 2005). This may, in turn, constrain their social and communication skills later on. Conversely, one domain may facilitate another by opening up new opportunities for growth. For example, better language skills may facilitate more opportunities for social interaction, which may in turn lead to better social skills.

“Neuroconstructivism” (Karmiloff-Smith, 1998; Sirois et al., 2008), a particularly promising theoretical framework that can capture this dynamic interplay between developmental domains, has gained increasing recognition in cognitive and developmental neuroscience research. The neuroconstructivist approach focuses on the developmental process itself as “key to understanding developmental disabilities” (Karmiloff-Smith, 1998). Therefore particular attention is paid to early differences - genetic alterations, environmental exposures, brain trauma - that may constrain multiple interacting developmental trajectories over time (Karmiloff-Smith, 1998; Farran & Karmiloff-Smith, 2012). This approach would therefore enhance longitudinal research in ASD by providing a more complete and dynamic understanding of the developmental processes that occur in the causal chain between predictors and outcomes.

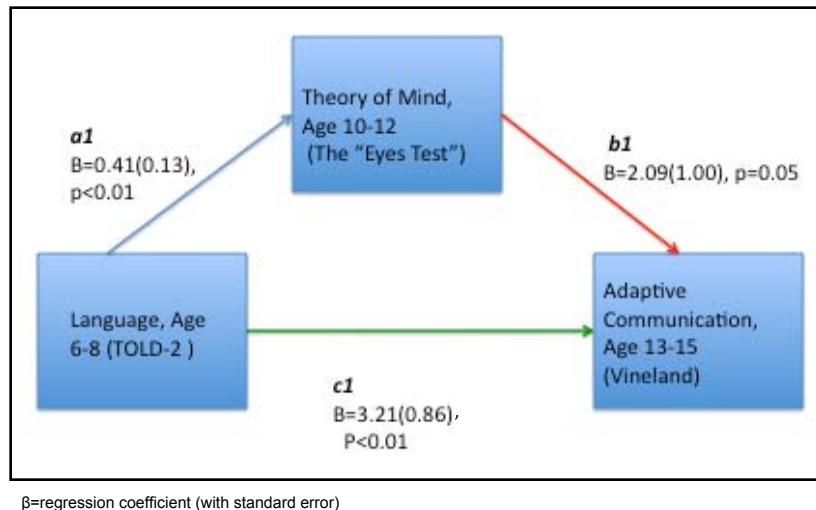
Well-designed prospective cohort studies also allow us to more thoughtfully address “how” predictors lead to particular outcomes. This requires an understanding of mediating factors and their underlying mechanisms. Mediation explains how such associations may “flow through” an intervening process in a presumably causal sequence; statistically, the predictor “X” affects the mediator “M”, which then affects the outcome “Y” (Baron & Kenny, 1986; Kraemer, Stice, Kazdin, Offord, & Kupfer, 2001;

MacKinnon, 2008). Understanding mediating pathways is important both to understanding developmental processes and to designing process-oriented clinical treatment; for example, learning how more advanced early language leads to improved adaptive functioning helps to understand brain processes in ASD and to develop more effective interventions targeted at neurocognitive mediating mechanisms rather than more “distant” predictors.

Theory of mind (ToM) - the ability to reason about the mental states of self and others - may be an important mediator in the association between early language abilities and later developmental outcomes in ASD. First, deficits in ToM have consistently distinguished children and adults with ASD from controls (e.g., Baron-Cohen, Leslie, & Frith, 1985; Baron-Cohen, Tager-Flusberg, & Cohen, 2000). Secondly, studies of typically developing children and individuals with ASD have shown ToM to be strongly associated with language abilities across different ages (Astington & Baird, 2005; see review by Milligan, Astington, & Dack, 2007). Some researchers have reported that, in fact, these two developmental abilities (language and ToM) may be more strongly related in people with ASD compared to typically developing children (Fisher et al., 2005; Tager-Flusberg & Joseph, 2005). Language skills may facilitate particularly important compensatory pathways given the social impairment associated with ASD. Children who demonstrate early language advantages will likely be exposed to a wider range of social situations and be better equipped to decode them than those who have poorer verbal abilities. This in turn would confer learning advantages in a range of ToM skills, including reading facial expressions of others, which would enhance the development of adaptive communication and social skills. ToM may therefore act as an important link mediating early language abilities to long-term outcomes in social and communication skills in ASD.

Several questions remain, however. First, we do not know how language and ToM are related over longer periods of time, and more specifically across developmental stages in individuals with ASD, for example from preschool to adolescent years. Furthermore, we do not know how ToM may relate to both early language and later functional outcomes. Understanding the ways in which children with ASD develop these cognitive abilities will help us to understand why individuals with ASD embark on different trajectories and also to design more effective interventions matched to specific trajectories.

Previous published work by our group found significant relationships between early language abilities and adolescent adaptive functioning in more cognitively able children with ASD (Bennett et al., 2008; Szatmari et al. 2003). Therefore, in this study, we aimed to determine whether ToM would mediate the association between language abilities in children assessed at ages 6-8 years old, and communication and social adaptive functioning skills assessed at age 12-14

Figure 1. Mediation Model 1

years old, using data from a longitudinal inception cohort study of 39 children with ASD and nonverbal intelligence quotients (IQ) of greater than 68 standard score points (autistic disorder and Asperger Syndrome). We hypothesized that:

1. Language abilities, measured at age 6-8 years via the Test of Language Development-2 (TOLD-2), would be significantly associated with both ToM measured at age 10-12 years (using a modified version of the "Reading the Mind in the Eyes Test") and later communication and social functioning (age 12-14) indexed on the Vineland Adaptive Behaviour Scales (VABS); and, that
2. ToM would mediate the relationship between early language and later adaptive functioning in communication and socialization.

Methods

For a detailed description of the participant sample and measures used in this study, as well as a graphic description of the mediation approach outlined in the analyses section, please see the supplementary online materials (SOM).

Analyses

The aim of these analyses was to determine whether ToM, as measured by the "Reading the Mind in the Eyes Test" at T2, mediated, or might account for, an association between the predictor variable - language abilities measured at Time 1 - and the two outcome variables, socialization and communication adaptive behaviours at T3. The effects of language and ToM on each adaptive functioning outcome variable were tested in two separate mediation models for assessing and comparing indirect effects, as outlined in Preacher and Hayes (2008) and depicted in Supplementary Figure 1. The indirect effects approach to testing simple mediation (i.e. one proposed mediating variable) apportions an independent variable X's hypothesized causal effect into

direct effects on the dependent variable Y (labeled path c) and indirect effects of X on Y through the proposed mediator M. Each path is quantified by an unstandardized regression coefficient. The effect of X on M is represented by path a, and path b represents the effect of M on Y while accounting for the effect of X. The indirect effect is then quantified by multiplying $a \times b$. Bootstrapping procedures were used to test indirect effects, standard errors and confidence intervals. The estimate of indirect effect would be considered to be statistically significant if 95% confidence intervals did not include zero, which would support the hypothesis of mediation. Model 1 tested the direct and indirect effects of T1 language on T3 adaptive communication and Model 2 tested the direct and indirect effects of T1 language on T3 adaptive socialization. Nonverbal IQ was included as a covariate of all paths in both models. Calculations were performed using the INDIRECT plug-in by A. F. Hayes in SPSS 20.0 (<http://www.afhayes.com/spss-sas-and-mplus-macros-and-code.html>).

Results

Within the cohort of 39 children, one individual enrolled but was unavailable for Time 1 data collection (family moved), however returned at T2. Three children were lost to follow-up at T3 (moved or declined follow-up). Boys comprised 90% of the available sample of 35 participants ($n=31$). Table 1 provides further descriptive statistics. Tests of normality indicate that the variable distributions were within normal limits.

Mediation analyses

Adaptive Communication Outcome. Figure 1 depicts the mediation model, partitioned into its component regression associations. The overall model accounted for 70% of the variance explained in the VABS Communication Subscale at Time 3 ($R^2=0.70$, $p<0.01$). As illustrated by path a, 41%

Table 1. Descriptive statistics of sample (n = 35)

	Mean (SD)	Minimum	Maximum
Age T1 (mo.)	89.00 (11.56)	73.00	112.00
Age T2 (mo.)	182.30 (14.27)	158.00	211.00
Age T3 (mo.)	208.54 (17.76)	175.00	254.00
TOLD-2 T1	5.21 (3.34)	1.50	13.50
Leiter NV IQ T1 **	86.48 (17.34)	36.00	127.00
“Eyes” Test T2 †	12.03 (3.45)	4.00	19.00
VABS COMM T3 ††	57.06 (21.21)	19.00	89.00
VABS SOC T3 ‡	49.78 (18.86)	19.00	91.00
* Test of Language Development-2, standard score points			
** Leiter nonverbal intelligence quotient score, standard score points			
† “Seeing the Mind in the Eyes Test”, modified for current study, maximum attainable score = 30.			
†† Vineland Adaptive Communication Score, standard score points			
‡ Vineland Adaptive Socialization Score, standard score points			

of the variance in the mediator variable - T2 ToM as measured by the Eyes Test - was predicted by individual language abilities at T1 ($B=0.41(0.13)$, $\beta = 0.65$, $p < 0.001$). In Path b_j , T2 ToM was also significantly associated with T3 VABS Communication, controlling for language at T1 ($B=2.09(1.00)$, $p=0.05$). There was a significant indirect effect of T1 language on T3 adaptive communication through T2 ToM, as depicted by the product term of the regression coefficients for paths a_i and b_j ($a_i b_j = 0.87(0.54)$, $CI=0.01, 2.17$). Language competency at T1 also demonstrated a significant direct association with T3 Adaptive Communication (Path c_1 : $B=3.21(0.86)$, $p < 0.001$). The analyses controlled for nonverbal IQ, which demonstrated a non-significant effect on the outcome variable ($B=0.11(0.17)$, $p=0.49$). In summary, in more cognitively able youth with ASD, structural language as measured by the TOLD-2 at age 6-8 was directly associated with adolescent adaptive communication as measured by the Vineland at age 12-14 years and indirectly through the effects of ToM as measured using the Eyes Test.

Adaptive socialization outcome. In contrast, language ability was not a statistically significant predictor of variance in the Vineland Socialization domain scores at Time 3, through either a direct path (path c_2 : $B=1.57(1.14)$, $p=0.18$) or indirect path through ToM ($a_2 b_2 = 0.043(0.56)$; $CI = -1.18, 1.13$), when controlling for nonverbal IQ (see Figure 2). Overall the entire model predicted a much smaller amount of variance in adaptive socialization ($R^2=0.29$, $p=0.01$) compared to the results for adaptive communication. ToM as measured using the Eyes test at T2 was not significantly associated with adaptive social skills as measured by the Vineland.

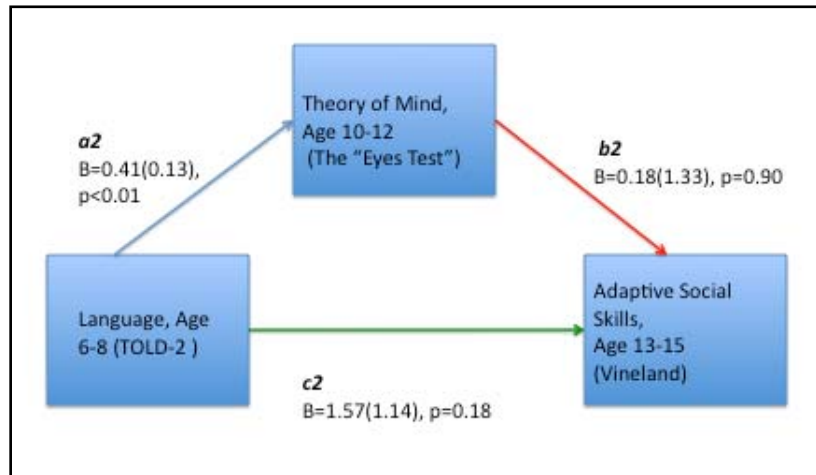
Discussion

Results of these analyses indicate that ToM mediated an association between early structural language and later adaptive communications skills as measured in early adolescence (12-14 years). Neither language ability nor ToM uniquely predicted a significant amount of variance in later adaptive socialization however. Each of these results from the mediation analysis will now be discussed, in turn.

Language and the development of ToM

Strong associations were found between language ability as measured by the TOLD-2 at age 6-8 and the Eyes Task performed approximately four years later in higher-IQ individuals with ASD. The results of the current study reiterate earlier findings of longitudinal associations between language ability and later ToM skills as indexed by narrative or false-belief tasks (Pellicano, 2010; Tager-Flusberg & Joseph, 2005). However they diverge from results obtained by Kaland et al. (Kaland, Callesen, Moller-Nielsen, Moretensen, & Smith, 2008), who failed to find cross-sectional associations between language ability and the Eyes Test in individuals with Asperger Syndrome.

Our results may demonstrate the developmental nature of associations between language and ToM. For example, the Eyes test has been described as an index of “social-perceptual” ToM skills, which involve more immediate decoding of interpersonal information (e.g. deciphering facial expressions) (Tager-Flusberg & Sullivan, 2000). False-belief and similar tasks, meanwhile, may involve more developmentally advanced “social-cognitive” ToM abilities and sophisticated linguistic skills (Tager-Flusberg & Sullivan, 2000). However, the strength of the associations between different components or “building blocks” of ToM and language may depend on the developmental stage of the individual

Figure 2. Mediation model 2

β =regression coefficient (with standard error)

with ASD. Early language advantages in younger children with ASD may enable them to engage more fully in social situations, leading over time to advantages in socio-perceptual ToM. For older verbal individuals with ASD, superior language ability may instead be a stronger requirement for understanding and reasoning through more complex social-cognitive ToM tasks (Tager-Flusberg, 2005).

Adaptive Communication Outcome. ToM mediated the association between language ability at age 6-8 (T1) and adaptive communication measured six years later. This suggests that structural language (grammar and vocabulary), ToM and later adaptive communication are related over the course of development in children with ASD. These longitudinal mediation mechanisms provide empirical support for developmental processes such as bootstrapping, in which early abilities such as language aptitude scaffold later-emerging skills such as ToM, which may then in turn facilitate growth in other domains such as adaptive functioning. As in typically developing children, linguistic understanding and awareness of others' emotions and intentions appear to build on each other over time, leading to more successful adaptive communication functioning in more cognitively able children and adolescents with ASD.

Adaptive Socialization Outcome. Together, language, non-verbal IQ and ToM predicted a relatively small but significant amount of variance in adaptive functioning on the Vineland socialization domain six years later. However, neither language at age 6-8 nor ToM as indexed at age 10-12 were uniquely predictive of variance in adaptive socialization in early adolescence, once accounting for IQ. This diverges from the results of previous studies (Hale & Tager-Flusberg, 2005; Tager-Flusberg, 2003). The present results are consistent with those of certain earlier studies that failed to find an association between false-belief ToM tasks and the Vineland Socialization domain (Frith, Happé, & Siddons, 1994; Fombonne, Siddons, Achard, Frith, &

Happé, 1994). They diverge, however, from those of Tager-Flusberg (2003), who found that ToM was significantly associated with the VABS Socialization scale (measured in the same year) in 69 verbal autistic children aged 4-14. This may reflect a difference in sample size, follow-up time or measurement issues.

This lack of association between ToM and adaptive social functioning reflects important methodological and conceptual issues with respect to measuring social development in individuals with ASD. First, while the communication domain of the Vineland generally depicts individual competencies (e.g. "Reads books", "Writes advanced letters"), the socialization domain, particularly approaching adolescence, describes functioning in a more ecologically valid way (e.g. "Going on double dates"; "Playing on school teams"). Social adaptive functioning may therefore measure the inclusiveness of the environment of an individual with ASD as much as his or her own individual social competencies. Second, the ToM skills measured by the Eyes test may be necessary but insufficient in navigating the complex social world of adolescents (Tager-Flusberg, 2003; Tager-Flusberg & Sullivan, 2000); higher scores on the Vineland may also reflect the development of a wide range of cognitive and other abilities (Frith et al., 1994; Fombonne, Siddons, Achard, Frith & Happé 1994). For example, the battery of false-belief and narrative-based ToM measures in Tager-Flusberg's (2005) study may have tapped more advanced ToM abilities as well as competencies such as executive function and language. From a neuroconstructivist perspective, the successful acquisition of much earlier building blocks of ToM – such as social attunement and joint attention in infancy and toddlerhood – or other early-emerging domain-general skills such as executive function may act as more primary constraints on developmental pathways (Dawson, Meltzoff, Osterling, Rinaldi, & Brown, 1998; Dawson et al., 2004; Mundy, Sigman, & Kasari, 1990; Pellicano, 2010) than socioperceptual ToM

skills in early adolescence. Longitudinal studies that follow younger children into adolescence and that chart interacting developmental pathways such as language, social cognition (including ToM), IQ and executive functioning will be key to clarifying such developmental questions.

Limitations. The mediation model proposed in this study is preliminary and correlational, with several important limitations. First, power issues due to the relatively small sample size may have affected the results, specifically the lack of significant associations with socialization functioning. A larger sample size would also have enabled the use of more sophisticated mediation analyses using structural equation models. Multiple methods and informants and a range of developmentally sensitive ToM tests (ideally including tests requiring minimal language ability) would further strengthen the findings. Furthermore, because the “Eyes” test was first published after the initiation of the study, it was only available at T2, so we were unable to determine the association between language at age T1 and ToM at T2 controlling for baseline ToM. Ideally, all tests (including outcome measures) should be measured at all time points. Finally, more complex models would enable one to test the growth of multiple interacting competencies, such as social skills, IQ, language, executive function and ToM - as well as contextual factors - in children with ASD.

Controlled intervention trials that include mediating and moderating variables can more definitively clarify how and under what circumstances interventions may shape developmental trajectories in children with ASDs. The present data support the idea that among children with ASD and higher cognitive functioning, interventions aimed at improving ToM in late childhood would have an impact on communication but not necessarily on socialization. However, barriers in translating gains made in ToM skills to real-world outcomes may exist. Multiple intervention studies have shown that learning conceptual ToM skills frequently fails to translate to improvements in daily social interactions among individuals with ASD (Begeer et al., 2011; Ozonoff, & Miller, 1995; Williams, Gray, & Tonge, 2012). Promising, albeit preliminary, studies suggest that social cognition-based interventions may overcome such barriers if they are developmentally sensitive (e.g. assessing a child’s readiness to learn; focusing on emerging social cognition in younger children) and ecologically valid (incorporating naturalistic settings; regular coaching in spontaneous situations; engaging parents, teachers, peers as facilitators or co-participants) (Bauminger, 2007; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010). ToM may be included as one part of a battery of interventions to improve social communication along multiple dimensions among children with ASD. However we need to target multiple skills during developmentally sensitive periods in order to address multiple outcomes.

Acknowledgements / Conflicts of Interest

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Child and Adolescent Mental Health Service Management Strategies that may Influence Wait Times

Isabelle Vallerand et al.

Commentary: Letters from Ainsworth: Contesting the 'Organization' of Attachment

Landa et al.