



PAPER

Does theory of mind performance differ in children with early-onset and regressive autism?

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Abstract

A deficit in theory of mind (ToM), or the ability to infer the mental states of others, has been implicated as one of the major characteristics of Autism Spectrum Disorder (ASD); however, little attention has been devoted to possible differences in ToM ability within ASD. The current study examined ToM performance in children with early-onset autism and regressive autism in comparison to typically developing children. Results indicated that children in the regressive autism group performed significantly better than the early-onset autism group on the non-verbal appearance–reality task. Additionally, Fisher’s exact tests indicated a pattern of lowest scores in the early-onset group and highest scores in the typically developing group, whereas the regressive autism group tended to score in between the early-onset and typically developing groups. The apparent heterogeneity in ToM performance within ASD could account for the lack of universality in ToM ability found in previous studies.

Introduction

Autism spectrum disorders (ASD) are characterized by restricted repetitive behaviors and qualitative impairments in social interaction and communication (Lord, Shulman & DiLavore, 2004). A deficit in theory of mind (ToM), or the ability to infer the mental states of the self and others, has also been implicated as one of the major characteristics of ASD. However, some research has questioned this assertion by demonstrating that a deficit in ToM is not unique to autism (Pellicano, Maybery, Durkin & Maley, 2006) or universal in all cases of ASD (Dahlgren & Trillingsgaard, 1996). Moreover, ToM deficits do not fully account for the impairments that accompany ASD (Peterson, Wellman & Liu, 2005).

Some investigators have also questioned the methodology used to test ToM by indicating that traditional ToM tasks may be inappropriate for young children or children with language impairments. As a result, some children may fail ToM tasks not because they lack a ToM, but because they lack the language skills necessary to complete the tasks (see Colle, Baron-Cohen & Hill, 2007; Happe, 1995; Rice, Koinis, Sullivan, Tager-Flusberg & Winner, 1997; Sapp, Lee & Muir, 2000). Non-verbal versions of common ToM tasks have been

developed in order to address such criticisms. For example, investigators have creatively examined the appearance–reality distinction by asking children to choose between objects by pointing rather than using expressive language to give an answer. Results from these investigations indicate that many typically developing children who failed traditional appearance–reality tasks gave correct responses when asked to respond by choosing an object as opposed to choosing a word (Rice *et al.*, 1997; Sapp *et al.*, 2000). Given the language impairments that usually accompany ASD, one might expect this modification to benefit performance in these children; however, few studies have examined non-verbal ToM task performance in children with ASD.

In addition to controversy over the language demands associated with traditional ToM tasks, findings that suggest that children with ASD show significant developmental change in ToM ability over time (Steele, Joseph & Tager-Flusberg, 2003) and that ToM performance in individuals with ASD can be improved through a social skills training program (Ozonoff & Miller, 1995) further complicate the links between ASD and ToM. Studies such as these indicate that at least some individuals with ASD who did not demonstrate an early ToM are capable of acquiring this ability during childhood, which may

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account in part for the diversity of findings and controversy over whether ToM deficits are necessarily implicated in ASD diagnoses.

Although a considerable amount of research on ToM and ASD has now been amassed, scant attention has been devoted to examining the differences in ToM ability within ASD. The question of whether performance differs as a function of 'subgroup' is an important issue that might help to explain the apparent lack of universality in associations between autism and ToM. Regressive autism, sometimes considered to be a subgroup of ASD, has an estimated prevalence of 10–40% in children diagnosed with ASD (Hansen, Ozonoff, Krakowiak, Angkustsiri, Jones, Deprey, Le, Croen & Hertz-Picciotto, 2008; Luyster, Richler, Risi, Hsu, Dawson, Bernier, Dunn, Hepburn, Hyman, McMahon, Goudie-Nice, Minshew, Rogers, Sigman, Spence, Goldberg, Tager-Flusberg, Volkmar & Lord, 2005).

Some of the observed heterogeneity in ToM abilities within studies of ASD might be due to the inclusion of children with both early-onset and regressive autism in the ASD sample. Early-onset autism is characterized by continuous atypical social and communicative development prior to 18 months of age. In contrast, regressive autism is characterized by the loss of acquired language and/or social skills between approximately 18 and 24 months, and the onset of social and communicative impairments (Goldberg, Osann, Filipek, Laulhere, Jarvis, Modahl, Flodman & Spence, 2003). Although regressive autism can include a loss of skill in numerous forms of socio-communication, loss of language is often the most salient and hence is commonly used by researchers to identify children with regression (Lord, 1995; Shinnar, Rapin, Arnold, Tuchman, Shulman, Ballaban-Gil, Maw, Deuel & Volkmar, 2001).

Despite studies designed to delineate the different developmental trajectories for children with early-onset and regressive autism, the distinction remains poorly understood. Many studies have examined the developmental outcomes of children with regressive autism; however, developmental trajectories prior to and after the onset of regression remain unclear (Lord *et al.*, 2004; Luyster *et al.*, 2005). The mixed state of the science was recently summarized by Baird and colleagues (Baird, Charman, Pickles, Chandler, Loucas, Meldrum, Carcani-Rathwell, Serkana & Simonoff, 2008): some studies suggest that outcomes for children with regressive autism include lower language levels, IQ, and reduced adaptability compared to their early-onset counterparts. In contrast, other studies find no difference between the groups; still others include data to show that sometimes outcomes for children with regressive autism are mixed depending on the abilities under examination. For example, in a sample of 381 children, Richler and colleagues (Richler, Luyster, Risi, Hsu, Dawson, Bernier, Dunn, Hepburn, Hyman, McMahon, Gouldie-Nice, Minshew, Rogers, Sigman, Spence, Goldberg, Tager-Flusberg, Volkmar & Lord, 2006) reported significantly

higher mean verbal IQ scores for the early-onset compared to the regressive autism group. Upon further inspection, bimodal scores on verbal IQ were detected within the subsample of children with regressive autism. One-third of the regressive autism group demonstrated high verbal IQ performance, and a second mode was found for very low verbal IQ performance, prompting the authors to propose a possible continuum of outcome severity within the regressive autism subgroup.

There is also evidence for the idea that symptom onset may be best characterized as a continuum rather than early-onset or regressive categories. Recent prospective examinations of at-risk infant siblings of children with ASD support this possibility (e.g. Ozonoff, Iosif, Baguio, Cook, Hill, Hutman, Rogers, Rozga, Sangha, Sigman, Steinfeld & Young, 2010). Other recent research, however, has indicated support for distinct subgroups of early-onset and regressive autism under the assumption that these behavioral profiles might have different etiologies and outcomes. For example, Rogers and colleagues (2010) recently investigated differences in early object imitation performance in children with early-onset and regressive autism, citing the possibility that early social difficulties faced by children with early-onset autism may cascade into later developmental impairments. In contrast, they proposed that children with regressive autism may be buffered or protected from later impairments because they did not experience early social difficulties. The data reported by Rogers and colleagues did not support this conjunction; instead, children in the early-onset autism group demonstrated better object imitation performance than children in the regressive autism group (Rogers, Young, Cook, Giolzetti & Ozonoff, 2010). However, these findings provide some support for the possibility of distinct subgroups. Further, the cascade mechanism described by Rogers and colleagues could potentially be involved in the acquisition of ToM in children with ASD, as precursors to ToM (e.g. joint attention) are thought to begin developing during the first year of life in typically developing children (Olinek & Poulin-Dubois, 2005, 2007; Wellman, Phillips, Dunphy-Lelii & LaLonde, 2004; Youngblade & Dunn, 1995). We note that the use of the early-onset and regressive autism subgroup distinction within the present study does not negate the possibility of a continuum of autism symptom severity within each subgroup or within broadly defined ASD.

To date, no studies have compared ToM abilities in children with early-onset and regressive autism. The paucity of research on this topic is surprising in light of the large body of literature examining differences in socio-communicative development and outcomes among children within early-onset and regressive autism. Research suggests that socio-communicative development and social interaction during the first years of life may facilitate the acquisition of ToM. For example, a collection of recent articles indicates developmental continuity in typically developing children between ToM

precursors in infancy and ToM performance during the preschool years (Olineck & Poulin-Dubois, 2005, 2007; Wellman *et al.*, 2004; Youngblade & Dunn, 1995). Moreover, meaningful early social interactions such as 'maternal mind-mindedness' during free play at 6 months of age have been shown to be associated with later ToM ability in both typically developing children and in children with ASD (Meins, Fernyhough, Wainwright, Das Gupta, Fradley & Tuckey, 2002). These findings highlight the importance of social interaction and development during infancy for ToM acquisition. To the extent that children with early-onset autism and regressive autism experience different developmental trajectories and social experiences during the infancy period, different outcomes in ToM performance might be expected.

Indeed, successfully acquiring a ToM during childhood is essential for developing the ability to appropriately navigate the social world. As has been indicated previously, children with ASD experience difficulty in this domain, but it may be the case that not all children are similarly affected. Differences in ToM ability between children within the possible subgroups of ASD could account for the lack of universality in ToM performance found in previous research and may have practical implications for intervention programs aimed at improving socio-communicative skills in children with ASD. Thus, the present study examined ToM in ASD by comparing the performance of children with early-onset autism and regressive autism relative to a typically developing control sample on a battery of verbal and non-verbal ToM tasks. After establishing that the two ASD groups differed from the typically developing controls, we examined whether differences in ToM ability differentiated children with early-onset and regressive autism. Specifically, we predicted that children with early-onset autism and regressive autism would differ in ToM performance; however, we did not make a prediction as to the direction of this difference given the mixed state of previous literature.

Method

Participants

Seventy-three children (37 with ASD and 36 typically developing) were recruited from one site of a large federally funded national study of autism and from autism-oriented community events. Most of the typically developing children were recruited from the same project as children with ASD; others were recruited from flyers circulated at and around the campus. The larger autism study included both children with early-onset and regressive autism. Children were mostly Caucasian (76%; 11% Asian; 9% more than one ethnicity, and 4% African American or other) and from middle- to upper-middle-class families with well-educated parents. Descriptive

statistics for children's gender and age at time of ToM testing are reported in Table 1. The gender distribution within this sample is consistent with predictions that ASDs are four to five times more likely to occur in males than females (Rice, 2007). Diagnoses of ASD were made independently as a part of the larger autism study and were based on the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter & Le Couteur, 1994) and the Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord, Risi, Lambrecht, Cook, Leventhal, DiLavore, Pickles & Rutter, 2000). The ADI-R, the Regression Supplement Form (Goldberg *et al.*, 2003) and clinical review informed the classification of regression in the larger study. Eighteen of those children with ASD had no early word loss and 15 children with ASD had experienced possible or definite early word loss (defined as the acquisition and subsequent loss of three or more words in addition to 'mama' and 'dada'). This definition is in accord with what has been published previously, as language regression is typically defined as the acquisition and subsequent loss of three to five words prior to 24 months of age (Baird *et al.*, 2008). Four children with ASD who experienced possible or definite early word loss were recruited after the completion of the larger study. Parental report of ASD diagnosis was confirmed using the Social Communication Questionnaire (Rutter, Bailey, Berument, Lord & Pickles, 2003), and the Regression Supplement Form (Goldberg *et al.*, 2003) determined classification of these four into the regressive autism group. Children in the typically developing group were administered the Childhood Autism Rating Scale (CARS; Schopler, Reichler, DeVellis & Daly, 1980) to rule out the possibility of undiagnosed ASD.

Two study sessions were scheduled approximately 2 weeks apart ($M = 19$ days, $SD = 6.39$). Exclusionary criteria included the following: a delay of more than 5 weeks between study sessions so as to ensure the integrity of the two analogous versions of each ToM assessment (three children with early-onset ASD, two children with regressive ASD, and one typically developing child were excluded); or having a sibling with ASD (two typically developing children). After the exclusion of these eight children, the final sample of 65 participants included 15 children with early-onset autism, 17 children with regressive autism and 33 typically developing children.

Procedure

At the first session, each child was administered the Peabody Picture Vocabulary Test III (PPVT; Dunn & Dunn, 1997). This untimed, verbally administered standardized measure assesses single-word receptive language in individuals over the age of 2 years. The PPVT is often used as a proxy for verbal ability (Hinton, De Vivo, Nereo, Goldstein & Stern, 2000) and is commonly included as a covariate in studies of ToM (Pellicano, 2007, 2010). Table 1 includes descriptive statistics by group for receptive language as measured by the PPVT.

Table 1 Descriptive statistics by study group

Demographic variable	Children with ASD		
	Early-onset (<i>n</i> = 15)	Regressive (<i>n</i> = 17)	Typical children (<i>n</i> = 33)
Age (years) ^a			
Mean	9.47 (2.85)	8.88 (3.33)	5.76 (1.42)
Range	6–14	4–16	4–9
Verbal intelligence (PPVT) ^b			
Mean	76.20 (25.28)	76.80 (20.72) (<i>n</i> = 15)	114.93 (13.42) (<i>n</i> = 31)
Range	40–114	42–109	87–145
Non-verbal intelligence ^c			
Mean	84.06 (16.67) (<i>n</i> = 14)	85.00 (15.83) (<i>n</i> = 13)	
Range	54–106	62–114	
Gender (percent of males)	93%	80%	83%

^a Age was significantly different between the typically developing group and the ASD groups, Brown-Forsythe (2, 33.92) = 12.33, $p < .001$. The typically developing group was significantly younger than the early-onset and regressive autism groups (Games-Howell, $p < .001$).

^b Verbal intelligence was significantly different between the typically developing and ASD groups, Brown-Forsythe (2, 32.86) = 25.192, $p < .001$. Verbal intelligence scores were significantly higher for the typically developing group than the early-onset and regressive autism groups (Games-Howell, $p < .001$).

^c No significant difference was found in non-verbal intelligence between the early-onset and regressive autism groups, ($t = -0.05$, $p = .96$).

Non-verbal intelligence scores (Stanford-Binet $n = 24$; Mullen $n = 3$) were obtained for children with ASD recruited from the larger study.

Children were also presented with four established ToM tasks at each study session: the change of location task (i.e. Sally-Anne; Baron-Cohen, Leslie & Frith, 1985); the change of contents task (i.e. Smarties; Wimmer & Hartl, 1991); and verbal and non-verbal versions of the appearance–reality task (Flavell, Flavell & Green, 1983; Sapp *et al.*, 2000). At the second session, the names of the characters and the critical objects were changed to create different but analogous versions of each task. The order of presentation of the ToM tasks and developmental assessments was counterbalanced to reduce order effects.

The change of location and change of contents tasks were employed to measure false-belief reasoning (i.e. whether children understand their own or another person's direct belief about reality). In the change of location task, dolls were used to act out a story in which one character has a false belief about the location of an object. As in Baron-Cohen *et al.* (1985), children passed this task if they reported that the character with the false belief would look for the object in the location where she left it (as opposed to the location where the second character had moved it) in addition to correctly answering two control questions about where the ball is now and where it was in the beginning.

In the change of contents task, the child was presented with a tube of M&Ms (or Play-Doh), and was then shown that it contained an unexpected object (e.g. crayons). The child was asked what a puppet that did not see the unexpected contents would think was in the tube. Children passed the test if they responded with the expected object (i.e. M&Ms or Play-Doh) and correctly answered the control question on the first or second try (Wimmer & Hartl, 1991).

The verbal and non-verbal appearance–reality tasks were included to assess children's ability to infer their

own mental states (Flavell *et al.*, 1983; Sapp *et al.*, 2000) and distinguish objects that superficially appear to be something different from what they are in reality. In the verbal task, each child was shown a deceptive object (i.e. a sponge that looked like a rock or a candle that looked like a crayon) and was then asked what the object looked like. After the child answered the questions, the true property of the object was revealed, and the child was asked what the object really was.

The non-verbal appearance–reality task was intended to be similar to the verbal task, but did not require the child to use expressive language to formulate a response. It is important to note that although the term 'non-verbal' is used here and elsewhere (Abelev & Markman, 2006) and was the term used by the developers of the task (Sapp *et al.*, 2000), the task requires that the researcher use expressive language. That is, the researcher describes the task verbally, but the response requirement is non-verbal in that the children's behavior is used as the indication of their response. Thus, the task requires only receptive language ability on the part of the child.

Once children demonstrated understanding of the verbal instructions in a pre-test, the children were each presented with six objects: four of the five following non-critical objects (i.e. tissue, flower, baby cup, apple, and a ball) and two critical objects (i.e. either a rock and a sponge or a crayon and a candle, depending on the version of the task). After a functional property of each object was demonstrated to the child, each child was asked to show the function of each object. The child was given two chances to imitate the demonstrated action with the object. Next, each child was presented with five new objects: four non-critical objects that differed from the earlier objects in color only and one critical object. The critical object was deceptive; either a sponge that looked like a rock, or a candle that looked like a crayon. Each child was asked to show the function of each of the four non-critical objects and then the critical deceptive

object. Following Sapp *et al.* (2000), tasks were discontinued if the child was unable to show the proper function of the object after two demonstrations and two attempts. After the demonstrations and imitation were complete, children were introduced to a teddy bear and asked to help the researcher take a picture of the teddy bear with a non-deceptive object (e.g. a ball). A similar question including the deceptive object was then asked. These questions assessed the child's understanding of appearance. Next, the child was introduced to a doll and asked to help find objects for the doll (e.g. non-critical: a ball; critical: a candle for a cake, a sponge to clean up a spill). These requests assessed the child's understanding of reality. The child passed the task by giving the correct object for each of the four questions (Sapp *et al.*, 2000).

Scoring of theory of mind tasks

For each version of each task, a score of zero was given when a child provided the incorrect answer to a critical question and a score of one was given when the child gave the correct answer. The scores from the two versions of each task were summed to create a total score for each individual task. Thus, a child could receive a minimum score of zero and a maximum score of two for any particular task. Because of zero cell counts for the 'no pass' score in the regressive autism and typically developing groups on the non-verbal appearance–reality tasks, the no pass (0) and partial pass (1) scores were combined in order to create a dichotomous outcome variable (i.e. no pass versus full pass) for all of the tasks. A score of two was classified as a full pass, whereas a score of zero or one was classified as a no pass for the purpose of fitting logistic regressions. Because the 'pass' group had to succeed on both trials, the possibility of passing by guessing or by possessing an emerging, but not fully developed, ToM was minimized. Children only received a passing score if all control questions were answered correctly. Some of the tasks were discontinued if certain control questions were not answered correctly (e.g. appearance–reality tasks).

Plan of analysis

Preliminary analyses included 3 (group: early-onset autism, regressive autism, typically developing) \times 2 (pass, no pass) Fisher's exact tests to examine whether children with early-onset autism, regressive autism and typically developing children differed on the four ToM tasks described above. Exact logistic regression analyses followed to test the main objectives of the study, and are described below. STATA software (Version 10) was used for statistical analysis.

Age and verbal intelligence ranged widely in the sample. Because the ANOVA assumption of homogeneity of variances was not met, separate Brown-Forsythe tests were conducted to determine whether it was necessary to

include age and verbal intelligence as covariates. Significant group differences warranted inclusion of these two variables as covariates in the regression analyses (the mean age and PPVT scores for the typically developing group were significantly different – lower and higher, respectively – than both ASD groups, $p < .001$; see Table 1). A Mann-Whitney test was also conducted to compare non-verbal intelligence between the early-onset and regressive autism groups; no significant difference was found ($z = -0.05$, $p = .961$). As is common in ASD samples, there were disproportionately more males than females. Using a 2×2 Fisher's exact test for each ToM task, no gender differences in performance were found within the ASD sample.

Exact logistic regression was used to test for differences among children with early-onset autism, regressive autism, and typically developing controls on the four ToM tasks, controlling for age and verbal intelligence. Separate exact logistic regression models were used to predict performance for each of the four ToM tasks. The independent variables included in the model were group (the early-onset and regressive autism groups were coded as two indicator variables with the typically developing control group as the reference), age, and verbal intelligence. Age and verbal intelligence were controlled in the exact logistic regression models when testing the effect of the group variable on the individual ToM tasks. Performance of the early-onset and regressive autism groups was directly compared by obtaining p -values from a second set of exact logistic regression models for each ToM task. The current sample was underpowered to detect small to moderate differences. When present, missing data were not systematic and were managed by assigning the subgroup mean for each specific missing data point (Tabachnick & Fidell, 2001).

Results

Fisher's exact tests

Figure 1 shows the percentage of full passes, partial passes, and no passes for each study group for each task. The Fisher's exact tests revealed a significant difference in performance between the early-onset autism, regressive autism and typically developing groups on all four ToM tasks (see Table 2). Following up on the significant group difference, visual inspection of the table indicates that the percentage of full pass scores for the typically developing group appeared to be substantially higher than that of the two autism groups. Examination of the data also indicated that the percentage of full pass scores for the early-onset autism group was consistently lower than the regressive autism group on three of the four tasks, although the extent of difference varied. The largest apparent differences between the ASD groups were observed in the verbal and non-verbal appearance–reality tasks, with the percentage of full pass scores lower

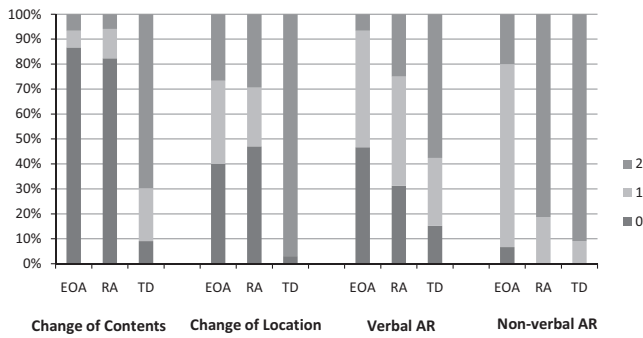


Figure 1 Percentage of no pass, partial pass and full pass for study groups on ToM tasks Note: EOA = Early-onset autism. RA = Regressive autism. TD = Typically developing.

Table 2 Fisher’s exact tests: theory of mind task performance by ASD group

Study variable	Children with ASD		Typical children % Pass	p-values for overall comparison
	Early-onset % Pass	Regressive % Pass		
Change of location	26.67	29.41	96.97	<0.001
Change of contents	6.67	5.88	69.70	<0.001
Appearance–reality				
Verbal	6.67	25.00	57.58	<0.010
Non-verbal	20.00	81.25	90.91	<0.001

Note: ASD = Autism Spectrum Disorder.

for the early-onset group than for the regressive group for both tasks. An exception to this pattern was observed in the change of contents task such that the early-onset and regressive autism groups performed almost identically.

Exact logistic regressions

The exact logistic regression models are shown in Table 3. These analyses indicate that group was a significant predictor of ToM performance for each of the four ToM tasks when age and verbal intelligence were controlled. Children with early-onset autism were sig-

nificantly less likely than the typically developing group to pass three of the four ToM tasks (change of location OR = .013, *p* < .001; change of contents OR = .034, *p* < .01, and non-verbal appearance reality OR = .062, *p* < .05), and were 92% less likely than the typically developing group to pass the verbal appearance–reality task, although the odds ratio fell short of significance (OR = .081, *p* = .06).

Children with regressive autism demonstrated a different pattern of performance. Whereas these children were significantly less likely than the typically developing group to pass the change of location (OR = .014, *p* < .001) and the change of contents tasks (OR = .027, *p* < .05), they did not perform differently from the typically developing group on the verbal and non-verbal appearance–reality tasks (verbal OR = .383, *ns*; non-verbal OR = .956, *ns*).

In order to examine the primary objective, each model was rerun with regressive autism as the comparison group to directly compare the performance between the early-onset and regressive autism groups. There was no significant difference in performance between these two groups on the change of location, change of contents, and verbal appearance–reality tasks [change of location (OR = 1.066, *ns*); change of contents (OR = 1.126, *ns*); verbal appearance–reality (OR = .202, *ns*)]. In contrast, the early-onset group was 93% less likely than the regressive autism group to pass the non-verbal appearance–reality task (OR = .064, *p* < .01).

Discussion

The importance of early experience for ToM development has been well documented in reference to typically developing children (Olineck & Poulin-Dubois, 2005, 2007; Meins *et al.*, 2002; Wellman *et al.*, 2004; Youngblade & Dunn, 1995); however, less is known about whether early experience is related to ToM in children with ASD. The current study examined the possibility that children with early-onset and regressive autism differ in ToM performance, an important omission in the burgeoning literature on ToM and autism. Some support

Table 3 Exact logistic regression odds ratios: theory of mind task performance by ASD group

Study variable	Theory of mind tasks							
	Change of location		Change of contents		Verbal appearance–reality		Non-verbal appearance–reality	
	OR	95% CI	OR	95% CI	OR	95% CI	OR	95% CI
Group (typically developing as reference, <i>n</i> = 31)								
Early-onset autism (<i>n</i> = 15)	0.013***	0.000, 0.220	0.034**	0.000, 0.561	0.082†	0.001, 1.051	0.062*	0.003, 0.674
Regressive autism (<i>n</i> = 15)	0.014***	0.000, 0.218	0.027*	0.000, 0.639	0.383	0.026, 4.022	0.956	0.062, 19.878
Group (regressive autism as reference)								
Early-onset autism	1.066	0.135, 9.101	1.126	0.013, 96.54	0.202	0.004, 2.604	0.065**	0.004, 0.531

Note: OR = Odds Ratio; CI = Confidence Interval. †*p* < .10; **p* < .05; ***p* < .01; ****p* < .001. Missing data on the PPVT reduced the sample size for the regressive autism group by two and the typically developing group by two.

was found for the hypothesis that ToM abilities would differ between children with early-onset and regressive autism. When controlling for age and verbal ability, children with early-onset autism were significantly less likely than their regressive counterparts to pass the non-verbal appearance–reality task, and the regressive autism group did not differ significantly from the typically developing children on this task. In contrast, the early-onset and regressive autism groups did not differ significantly on the three other theory of mind tasks: change of location, change of contents, and verbal appearance–reality. However, the early-onset and regressive autism groups differed in their performance relative to the typically developing group. Whereas the early-onset autism group was significantly less likely than the typically developing group to pass three of the four ToM tasks and was marginally less likely to pass the fourth task, the regressive autism group did not differ significantly from the typically developing group on two of the four ToM tasks: the verbal and non-verbal appearance reality task. This set of findings supports our hypothesis that some of the controversy in whether or not ToM is a defining feature of autism might reflect the inclusion of children with and without regression in the same ASD sample. The additional heterogeneity inherent in ASD samples may have obscured the chances of detecting true distinctions between early-onset and regressive autism groups on the change of location, change of contents and verbal appearance–reality tasks.

Findings from this study also inform the controversy about whether children who once had early language skills but later lost language (i.e. the regression group) retain some advantage over children whose symptoms of autism, including language delays, began early in life (i.e. the early-onset group). Of the studies that indicate differences between these two possible subgroups, most, but not all, suggest that children with regressive autism tend to have less favorable outcomes (Baird *et al.*, 2008; Hoshino, Kanako, Yashima, Kumashiro, Volkmar & Cohen, 1987; Kobayashi & Murata, 1998; Kurita, 1985; Rogers & DiLalla, 1990, Rogers *et al.*, 2010).

Our finding that children who once had language and then regressed performed better on the non-verbal appearance–reality task was somewhat surprising, given the direction of the difference. Possible explanations come from developmental cascade theories of autism. These theories predict less severe impairment for children with regressive autism in areas of development that are affected by early exposure to social processes. From this perspective, children who experienced relatively typical development up until 18–24 months of age may be protected from early deficits in social processes that cascade into the later impairments observed in children with ASD (see Rogers *et al.*, 2010). In other words, children with regressive autism may have been afforded developmental benefits from early developing self-understanding, other early ToM precursors and social interaction prior to autism symptom onset. If this were indeed the

case, children with regressive autism would be expected to outperform children with early-onset autism on ToM tasks. Thus, children with access to language early in life may have been able to learn about certain aspects of the social world that then facilitated the development of an understanding of the appearance–reality distinction. Developmental cascade theories of ASD have heretofore gone mostly unsupported, but with replication the current findings may be construed as empirical support for this theoretical position.

Also somewhat surprising was the finding that the early-onset and regressive autism groups differed on the non-verbal appearance–reality task in particular. Further, the regressive autism group did not differ from the typically developing group on either the verbal or the non-verbal appearance–reality tasks. Appearance–reality tasks are thought to assess children’s understanding of their own mental states (Flavell *et al.*, 1983; Sapp *et al.*, 2000), and recent work indicates the role of self-understanding, a skill that undergoes rapid development during the first years of life, in mentalizing abilities (Lombardo & Baron-Cohen, 2011). Children who experienced somewhat typical development up until 18 to 24 months of age prior to experiencing language loss may have developed the prerequisites necessary for achieving an understanding of the appearance–reality distinction. In contrast, children with persistent early deficits in language from infancy may not have had the opportunity to acquire these early social cognitive skills. Further, the removal of expressive language constraints in the non-verbal appearance–reality task may have made these abilities salient. An alternative interpretation of the present findings could be that deficits in regressive autism may be primarily in the language domain. This interpretation may account for the enhanced performance observed from children with regressive autism on the ToM task that did not require expressive language. Additionally, it lends further support to previous findings of poorer communicative abilities (Brown & Prelock, 1995) and lower language level in children with regressive autism in comparison to children with early-onset autism (Bernabei, Cerquiligni, Cortesi & D’Ardua, 2007).

The significant difference between children with early-onset and regressive autism on the non-verbal appearance–reality task, along with the similar probability of passing for the regressive autism and typically developing groups, indicates the importance of accounting for symptom onset in ASD samples. The present research highlights the fact that although children with autism often perform worse on ToM tasks relative to typically developing children (Baron-Cohen, 1989), children with regressive autism may not have a delay in this particular facet of ToM. Future studies might include a measure of executive functioning in order to examine the possibility that a difference in executive functioning between children within different ASD subgroups may underlie differences in ToM ability.

Although only one of the exact logistic regression models comparing performance of the early-onset and regressive autism groups met conventional levels of statistical significance (i.e. the non-verbal appearance–reality task), the percentage distributions of full pass and no pass scores from the Fisher’s exact tests for the contrasts on two of the other ToM tasks were in the same direction. In all but one instance, the regressive autism group scored higher than the early-onset group and lower or about the same as the typically developing group. This consistent pattern of performance could be indicative of an overall advantage for the regressive autism group over the early-onset group across three of the four ToM tasks reported in this study. Although our analyses were underpowered to detect small and moderate between-group differences, our sample size is similar to those included in other published reports examining ToM in children with ASD examined as a whole (i.e. not separated by early-onset and regressive autism; e.g. Hamilton, Brindley & Frith, 2009; Sally & Hill, 2006; Sobel, Capps & Gopnik, 2005). The significant between-group difference on the non-verbal ToM task may have been more easily detected because of the reduced constraints on expressive language capacity provided by the design of the task. The present findings only indicate a difference between early-onset and regressive autism on the non-verbal appearance–reality task; however, future examinations utilizing larger samples may reveal statistically significant differences between these two groups in other facets of ToM.

Another interpretation is that the discrepancies in performance across ToM tasks observed in all three study groups were due to inherent higher order constraints associated with each specific task. For example, although both the change of contents and change of location tasks measure false-belief reasoning, the linguistic and cognitive demands of the change of contents task are thought to differ from those of the change of location task given the types of questions asked (Lind & Bowler, 2009). Notably, percentage of passing on the change of contents task was nearly identical for the children with and without regression, suggesting that children with regressive autism perform similarly on the change of contents task relative to their early-onset counterparts. Additionally, the performance of all three groups on the change of contents task was considerably low in comparison to the other tasks in this study. Thus, the heightened linguistic demands of the change of contents task may be one explanation for the observed variation in performance across ToM tasks for all study groups. Differential linguistic and cognitive demands may also be responsible for the relatively low percentage of pass observed from the typically developing group on the verbal appearance–reality task.

The observed and possible differences in ToM performance reported in this study should also be discussed in the context of recent prospective examinations of at-risk infant siblings of children with ASD that indicate

that the distinction between early-onset and regressive autism may not be as dichotomous as was once thought. In particular, Ozonoff and colleagues (2010) propose a continuum of autism symptom onset rather than the traditional categorical distinction. Although microgenetic investigations of the onset of autism symptoms may indicate that a continuous view is indeed appropriate, the current sample was well defined with regard to classifications of early-onset and regressive autism, such that participants were classified by parental interview, questionnaire, and clinical review. This multi-source method reduced the likelihood of misclassifying children with regressive ASD. Additionally, researchers have continued to examine possible differences between children with early-onset and regressive ASD, citing the need to understand differences between possible subgroups that may not have shared etiologies (Rogers *et al.*, 2010). We caution that the findings from the current study were not intended to determine whether or not categorization or continuum views are preferred, but rather to indicate the importance of accounting for heterogeneity in performance on ToM tasks in ASD samples and to address whether ToM represents a single set or range of abilities.

In this study, the regressive autism group was defined by language loss. One might argue that the differences found in performance on the non-verbal task could be due to communicative abilities; however, all of the tasks required the use of receptive language on the part of the children, and the group difference on the non-verbal appearance–reality task withstood control for receptive language. Further, because the pattern of results was mostly consistent throughout the verbal and non-verbal tasks (although not statistically significant), we suspect that these differences may not be limited to the non-verbal task. Replication with larger samples is needed, however, to support this conjecture.

Some previous research has suggested that a deficit in ToM is one of the primary attributes of ASD. However, the results of the current study lend support to the growing body of literature that indicates that ToM in ASD might be better characterized in terms of a range of abilities rather than a deficit. Specifically, the differing patterns of performance between the early-onset and regressive autism groups indicate that the reported lack of universality in ToM ability among children with ASD may be due in part to the inclusion of multiple subgroups (or a wide continuum of abilities) in ASD samples. These findings suggest that the heterogeneity of ASD symptoms and symptom onset should be considered when examining ToM, and possibly other cognitive and behavioral deficits associated with the disorder.

In conclusion, it is important to consider that ToM is likely a multi-faceted ability (rather than an ‘all or none process’) such that individuals with ASD may demonstrate adeptness in some areas while experiencing deficits in others. This possibility is confirmed by the differing patterns of performance observed in the current study, and by prior research using more advanced ToM tasks

(e.g. the Eyes task, Baron-Cohen, Wheelright, Hill, Raste & Plumb, 2001; the Strange Stories task, Happe, 1994), which has revealed subtle ToM deficits into adolescence and adulthood in high-functioning ASD samples (Kaland, Callesen, Moller-Nielsen, Mortensen & Smith, 2008). Thus, the findings of the present study bear on the lack of universality, or the multi-faceted nature, of ToM found in previous research with ASD samples. The present findings also serve to create a more complete picture of regressive autism and its distinctive place within ASD.

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References

- Abelev, M., & Markman, E. (2006). Young children's understanding of multiple object identity: appearance, pretense and function. *Developmental Science*, *9*, 590–596.
- Baird, G., Charman, T., Pickles, A., Chandler, S., Loucas, T., Meldrum, D., Carcani-Rathwell, I., Serkana, D., & Simonoff, E. (2008). Regression, developmental trajectory, and associated problems in disorders in autism spectrum: the SNAP study. *Journal of Autism and Developmental Disorders*, *38*, 1827–1836.
- Baron-Cohen, S. (1989). Are autistic children 'behaviorists'? An examination of their mental-physical and appearance-reality distinctions. *Journal of Autism and Developmental Disorders*, *19*, 579–600.
- Baron-Cohen, S., Leslie, A.M., & Frith, U. (1985). Does the autistic child have a 'theory of mind'? *Cognition*, *21*, 37–46.
- Baron-Cohen, S., Wheelright, S., Hill, J., Raste, Y., & Plumb, I. (2001). The 'Reading the mind in the eyes' test revised version: a study with normal adults and adults with Asperger's syndrome or high-functioning autism. *Journal of Child Psychology and Psychiatry*, *38*, 813–822.
- Bernabei, P., Cerquilignini, A., Cortesi, F., & D'Ardia, C. (2007). Regression versus no regression in the autistic disorder: developmental trajectories. *Journal of Autism and Developmental Disorders*, *25*, 305–309.
- Brown, J., & Prelock, P.A. (1995). Brief report: The impact of regression on language development in autism. *Journal of Autism and Developmental Disorders*, *25*, 305–309.
- Colle, L., Baron-Cohen, S., & Hill, J. (2007). Do children with autism have a ToM? A non-verbal test of autism vs. specific language impairment. *Journal of Autism and Developmental Disorders*, *37*, 716–723.
- Dahlgren, S.O., & Trillingsgaard, A. (1996). Theory of mind in non-retarded children with autism and Asperger's syndrome: a research note. *Journal of Child and Psychiatry and Allied Disciplines*, *37*, 759–763.
- Dunn, L.M., & Dunn, L.M. (1997). *PPVT-III Peabody picture vocabulary test*. San Antonio, TX: Pearson & PsychCorp.
- Flavell, J.H., Flavell, E.R., & Green, F.L. (1983). Development of the appearance-reality distinction. *Cognitive Psychology*, *15*, 95–120.
- Goldberg, W.A., Osann, K., Filipek, P.A., Lulhere, T., Jarvis, K., Modahl, C., Flodman, P., & Spence, M.A. (2003). Language and other regression: assessment and timing. *Journal of Autism and Developmental Disorders*, *33*, 607–616.
- Hamilton, A.F.C., Brindley, R., & Frith, U. (2009). Visual perspective taking impairment in children with autism spectrum disorder. *Cognition*, *113*, 37–44.
- Hansen, R.L., Ozonoff, S., Krakowiak, P., Angkustsiri, K., Jones, C., Deprey, L.J., Le, D.N., Croen, L.A., & Hertz-Picciotto, I. (2008). Regression in autism: prevalence and associated factors in the CHARGE study. *Ambulatory Pediatrics*, *8*, 1530–1567.
- Happe, F. (1994). An advanced test of theory of mind: understanding of story characters' thoughts and feelings by able autistic, mentally handicapped and normal children and adults. *Journal of Autism and Developmental Disorders*, *30*, 225–236.
- Happe, F.G.E. (1995). The role of age and verbal ability in the theory of mind task performance of subjects with autism. *Child Development*, *66*, 843–855.
- Hinton, V.J., De Vivo, D.C., Nereo, N.E., Goldstein, E., & Stern, Y. (2000). Poor verbal working memory across intellectual level in boys with Duchenne dystrophy. *Neurology*, *54*, 2127–2132.
- Hoshino, Y., Kanako, M., Yashima, Y., Kumashiro, H., Volkmar, F., & Cohen, D. (1987). Clinical features of autistic children with setback course in their infancy. *Japanese Journal of Psychiatry and Neurology*, *41*, 237–245.
- Kaland, N., Callesen, K., Moller-Nielsen, A., Mortensen, E.L., & Smith, L. (2008). Performance of children and adolescents with Asperger syndrome or high-functioning autism on advanced theory of mind tasks. *Journal of Autism and Developmental Disorders*, *38*, 1112–1123.
- Kobayashi, R., & Murata, T. (1998). Setback phenomenon in autism and long-term prognosis. *Acta Psychiatrica Scandinavica*, *98*, 296–303.
- Kurita, H. (1985). Infantile-autism with speech loss before the age of 30 months. *Journal of the American Academy of Child and Adolescent Psychiatry*, *24*, 191–196.
- Lind, S.E., & Bowler, D.M. (2009). Language and theory of mind in autism spectrum disorder: the relationship between complement syntax and false belief task performance. *Journal of Autism and Developmental Disorders*, *39*, 929–937.
- Lombardo, M.V., & Baron-Cohen, S. (2011). The role of the self in mindblindness in autism. *Consciousness and Cognition*, *20*, 130–140.
- Lord, C. (1995). Follow-up of two-year-olds referred for possible autism. *Journal of Child Psychology and Psychiatry*, *36*, 1365–1382.
- Lord, C., Risi, S., Lambrecht, L., Cook, E.H. Jr, Leventhal, B.L., DiLavore, P.C., Pickles, A., & Rutter, M. (2000). The Autism Diagnostic Observation Schedule-Generic: a stan-

- standard measure of social and communicative deficits associated with the spectrum of autism. *Journal of Autism and Developmental Disorders*, **30**, 205–223.
- Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview-Revised: a revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders*, **24**, 659–685.
- Lord, C., Shulman, C., & DiLavore, P. (2004). Regression and word loss in autistic spectrum disorders. *Journal of Child Psychology and Psychiatry*, **45**, 936–955.
- Luyster, R., Richler, J., Risi, S., Hsu, W.L., Dawson, G., Bernier, R., Dunn, M., Hepburn, S., Hyman, S.L., McMahon, W.M., Goudie-Nice, J., Minshew, M., Rogers, S., Sigman, M., Spence, M.A., Goldberg, W.A., Tager-Flusberg, H., Volkmar, F.R., & Lord, C. (2005). Early regression in social communication in autism spectrum disorders: a CPEA Study. *Developmental Neuropsychology*, **27**, 311–336.
- Meins, E., Fernyhough, C., Wainwright, R., Das Gupta, M., Fradley, E., & Tuckey, M. (2002). Maternal mind-mindedness and attachment security as predictors of theory of mind understanding. *Child Development*, **73** (6), 1715–1726.
- Olineck, K.M., & Poulin-Dubois, D. (2005). Infants' ability to distinguish between intentional and accidental actions and its relation to internal state language. *Infancy*, **8**, 91–100.
- Olineck, K.M., & Poulin-Dubois, D. (2007). Imitation of intentional actions and internal state language in infancy predict preschool theory of mind skills. *European Journal of Developmental Psychology*, **4**, 14–30.
- Ozonoff, S., Iosif, A.M., Baguio, F., Cook, I. C., Hill, M.M., Hutman, T., Rogers, S.J., Rozga, A., Sangha, S., Sigman, M., Steinfeld, M.B., & Young, G.S. (2010). A prospective study of the emergence of early behavioral signs of autism. *Journal of the American Academy of Child and Adolescent Psychiatry*, **49**, 256–266.
- Ozonoff, S., & Miller, J.N. (1995). Teaching theory of mind: a new approach to social skills training for individuals with autism. *Journal of Autism and Developmental Disorders*, **25**, 415–433.
- Pellicano, E. (2007). Links between theory of mind and executive function in young children with autism: clues to developmental primacy. *Developmental Psychology*, **43** (4), 974–990.
- Pellicano, E. (2010). Individual differences in executive function and central coherence predict developmental changes in theory of mind in autism. *Developmental Psychology*, **46** (2), 530–544.
- Pellicano, E., Maybery, M., Durkin, K., & Maley, A. (2006). Multiple cognitive capabilities/deficits in children with an autism spectrum disorder: 'weak' central coherence and its relationship to theory of mind and executive control. *Development and Psychopathology*, **18**, 77–98.
- Peterson, C.C., Wellman, H.M., & Liu, D. (2005). Steps in theory-of-mind development for children with deafness or autism. *Child Development*, **76**, 502–517.
- Rice, C. (2007). Prevalence of autism spectrum disorders. *Morbidity and Mortality Weekly Report*, **56** (S S01), 1–11 Retrieved from <http://www.cdc.gov/mmwr/preview/mmwrhtml/ss5601a1.htm>.
- Rice, C., Koinis, D., Sullivan, K., Tager-Flusberg, H., & Winner, E. (1997). When 3-year-olds pass the appearance–reality test. *Developmental Psychology*, **33**, 54–61.
- Richler, J., Luyster, R., Risi, S., Hsu, W.L., Dawson, G., Bernier, R., Dunn, M., Hepburn, S., Hyman, S.L., McMahon, W.M., Gouldie-Nice, J., Minshew, N., Rogers, S., Sigman, M., Spence, M.A., Goldberg, W.A., Tager-Flusberg, H., Volkmar, F.R., & Lord, C. (2006). Is there a 'regressive phenotype' of autism spectrum disorder associated with the Measles-Mumps-Rubella vaccine? A CPEA study. *Journal of Autism and Developmental Disorders*, **36**, 299–316.
- Rogers, S.J., & DiLalla, D. (1990). Age of symptom onset in young children with pervasive developmental disorders. *Journal of the American Academy of Child and Adolescent Psychiatry*, **29**, 863–872.
- Rogers, S.J., Young, G.S., Cook, I., Giolzetti, A.G., & Ozonoff, S. (2010). Imitating actions on objects in early-onset and regressive autism: effects and implications of task characteristics on performance. *Development and Psychopathology*, **22**, 71–85.
- Rutter, M., Bailey, A., Berument, S.K., Lord, C., & Pickles, A. (2003). *Social Communication Questionnaire (SCQ)*. Los Angeles, CA: Western Psychological Services.
- Sally, D., & Hill, E. (2006). The development of interpersonal strategy: autism, theory-of-mind, cooperation and fairness. *Journal of Economic Psychology*, **27**, 73–97.
- Sapp, F., Lee, K., & Muir, D. (2000). Three-year-olds' difficulty with the appearance–reality distinction: is it real or is it apparent? *Developmental Psychology*, **36**, 547–560.
- Shinnar, S., Rapin, I., Arnold, S., Tuchman, R.F., Shulman, L., Ballaban-Gil, K., Maw, M., Deuel, R.K., & Volkmar, F.R. (2001). Language regression in childhood. *Pediatric Neurology*, **24**, 183–189.
- Schopler, E., Reichler, J.R., De Villis, R.F., & Daly, K. (1980). Toward objective classification of childhood autism: Childhood Autism Rating Scale (CARS). *Journal of Autism and Developmental Disorders*, **10**, 91–103.
- Sobel, D.M., Capps, L.M., & Gopnik, A. (2005). Ambiguous figure perception and theory of mind understanding in children with autistic spectrum disorders. *British Journal of Developmental Psychology*, **23**, 159–174.
- Steele, S., Joseph, R.M., & Tager-Flusberg, H. (2003). Developmental change in ToM abilities in children with autism. *Journal of Autism and Developmental Disorders*, **33**, 461–467.
- Tabachnick, B., & Fidell, L. (2001). *Using multivariate statistics* (4th edn.). New York: Harper & Row.
- Wellman, H.M., Phillips, A.T., Dunphy-Lelii, S., & LaLonde, N. (2004). Infant and social attention predicts preschool social cognition. *Developmental Science*, **7**, 283–288.
- Wimmer, H., & Hartl, M. (1991). Against the Cartesian view on mind: young children's difficulty with own false belief. *British Journal of Developmental Psychology*, **9**, 125–138.
- Youngblade, L.M., & Dunn, J. (1995). Individual differences in young children's pretend play with mother and sibling: links to relationships and understanding of other people's feelings and beliefs. *Child Development*, **66**, 1472–1492.

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