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Distinctive Role of Symbolic Number Sense in Mediating the Mathematical Abilities of Children with Autism

Alexis Hiniker^{2,6}, Miriam Rosenberg-Lee^{1,3}, and Vinod Menon^{1,3,4,5}

Miriam Rosenberg-Lee: miriamrl@stanford.edu; Vinod Menon: menon@stanford.edu

¹Department of Psychiatry and Behavioral Sciences, Stanford University School of Medicine, 401 Quarry Rd, Stanford, CA 94305-5778, USA

²School of Education, Stanford University, Stanford, CA, USA

³Stanford Neuroscience Institute, Stanford University, Stanford, CA, USA

⁴Department of Neurology and Neurological Sciences, Stanford University, Stanford, CA, USA

⁵Symbolic Systems Program, Stanford University, Stanford, CA, USA

Abstract

Despite reports of mathematical talent in autism spectrum disorders (ASD), little is known about basic number processing abilities in affected children. We investigated number sense, the ability to rapidly assess quantity information, in 36 children with ASD and 61 typically developing controls. Numerical acuity was assessed using symbolic (Arabic numerals) as well as non-symbolic (dot array) formats. We found significant impairments in non-symbolic acuity in children with ASD, but symbolic acuity was intact. Symbolic acuity mediated the relationship between non-symbolic acuity and mathematical abilities only in children with ASD, indicating a distinctive role for symbolic number sense in the acquisition of mathematical proficiency in this group. Our findings suggest that symbolic systems may help children with ASD organize imprecise information.

Keywords

Number sense; Autism spectrum disorders; Math ability

Introduction

In the popular imagination, autism has been associated with superior mathematical ability and incredible feats of rapid numerical estimation. For example, in the 1988 drama, *Rain*

Correspondence to: Miriam Rosenberg-Lee, miriamrl@stanford.edu; Vinod Menon, menon@stanford.edu.

⁶Present Address: Human Centered Design and Engineering, University of Washington, Seattle, WA, USA

Alexis Hiniker and Miriam Rosenberg-Lee have contributed equally to this work.

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Man, Raymond Babbitt can instantaneously enumerate hundreds of toothpicks and compute square roots to eight decimal places (Draaisma 2009). Yet no studies to date have systematically investigated whether basic number sense might be a particular strength in children with autism spectrum disorders (ASD). Non-symbolic number sense—the ability to rapidly apprehend approximate quantities—is an evolutionarily conserved capacity (Cantlon and Brannon 2007; Dehaene et al. 1998), while symbolic number sense refers to the uniquely human ability to automatically access quantity information from numerical symbols (Ansari 2008; Dehaene 2011). Emerging research in typically developing (TD) children has suggested that both forms of number sense are predictive of formal mathematics achievement (De Smedt et al. 2013). Here, we investigate three questions: (1) how do symbolic and non-symbolic estimation abilities in children with ASD compare with those of their TD peers, (2) how are symbolic and non-symbolic number sense related to overall mathematical abilities in children with ASD, and (3) how do these relations differ from those observed in age-, IQ-, and achievement-matched TD children. Characterizing foundational cognitive capacities and their relation to academic achievement can identify relative strengths and weaknesses and thereby potentially inform best practices for training individuals with autism.

Autism spectrum disorders are a set of neurodevelopmental conditions characterized by impairments in social communication, restricted interests and repetitive behaviors (American Psychiatric Association 2013). ASD is most often investigated from the perspective of its core deficits, yet there is growing evidence to suggest that individuals with the disorder can also demonstrate cognitive strengths (Baron-Cohen et al. 2009; Bennett and Heaton 2012). Mathematics represents one of a handful of skills routinely reported as a savant talent in autism, and indeed some individuals with ASD have shown extraordinary powers of calculation and mathematical reasoning (Howlin et al. 2009; Treffert 2009). Beyond such rare cases, mathematics has been documented as an area of relatively spared or even enhanced performance in a large proportion of individuals with ASD (Chiang and Lin 2007; Iuculano et al. 2014; Jones et al. 2009; Wei et al. 2015).

Despite these findings and anecdotal evidence of enhanced math skills in ASD, little is known about basic numerical competencies and their relation to math abilities in children with ASD. Non-symbolic number sense enables humans as well as non-human species to estimate quantity information from a set of discrete items without serial counting. This capacity also allows us to rapidly compare quantities, determining at a glance the more numerous of two sets of items. Like other forms of psychophysical discriminations, the precision of these judgments conforms to the Weber-Fechner law, such that accuracy scales progressively with the ratio between the size of the two sets, rather than the absolute difference between them (Dehaene 2003). For example, it is easier to determine the more numerous quantity when comparing 4 dots to 3 than when comparing 8 dots to 7. Over the course of early mathematics education, children learn to associate non-symbolic quantities first with number words and then with numerical symbols (Von Aster and Shalev 2007). Evidence that symbolic constructs are mapped onto existing non-symbolic conceptions of quantity comes partly from the finding that adults display similar imprecision for both formats (Dehaene 2003; Moyer and Landauer 1967). For example, neurotypical adults can

more quickly determine the larger of the two numerals '4' and '3' than they can determine the larger of '8' and '7.'

Theories of mathematical skill development have posited that non-symbolic number sense forms a foundation for the acquisition of mathematical knowledge (Butterworth 2005; Dehaene 2003). Consistent with this proposal, several studies in TD individuals have linked individual differences in non-symbolic number sense directly to performance on standardized measures of math ability (Feigenson et al. 2013; Halberda et al. 2008; Libertus et al. 2011). However, other studies have failed to find such an association, instead finding that symbolic number sense is a robust predictor of math ability, especially in children (Durand et al. 2005; Holloway and Ansari 2009; Kolkman et al. 2013). Little is known about such relations in children with ASD.

An alternative model of math skill development posits that individual differences in non-symbolic number sense first lead to differences in symbolic number sense, which then contribute to individual differences in mathematical ability (Von Aster and Shalev 2007). Several recent studies have provided support for this model by demonstrating that measures of symbolic number knowledge, including number recognition and ordinality principles, can mediate the relationship between non-symbolic number sense and math ability in TD individuals (Lyons and Beilock 2011; Van Marle et al. 2014). Surprisingly, even in TD individuals, few studies have measured both symbolic and non-symbolic capacities and standardized math abilities within the same sample. Consequently, the relative contributions of these skills to mathematics remain unknown. Here, for the first time, we examine both symbolic and non-symbolic number sense in 7-to-12 year-old children with ASD, and a well-matched group of TD controls, and relate these measures to their mathematical abilities.

We examined non-symbolic and symbolic number sense in 36 high-functioning children with ASD and 61 TD children explicitly matched on age and full-scale IQ, and found no differences in reading, working memory, and math ability. Participants performed two magnitude comparison tasks, the first with non-symbolic (dots) stimuli and the second with symbolic (Arabic numbers) stimuli. Following the seminal study by Moyer and Landauer (1967) in adults, which demonstrated sensitivity to numerical distance among single-digit numbers, numerosities in the current study ranged from 2 to 9. In addition to accuracy and reaction time, we also computed a Weber fraction (w) for each stimulus type, a measure of the precision of the numerical representation (Dehaene 2003).

We first examined whether non-symbolic and symbolic number sense are areas of relative strength in children with ASD. If low-level visual acuity and perceptual strengths (Mitchell and Ropar 2004; Rinehart et al. 2000) contribute to number sense abilities, one would predict spared or enhanced non-symbolic acuity in children with ASD, relative to TD children. Although no studies to date have investigated symbolic number comparison skills in individuals with ASD, prior work has documented strengths in reading, another academically relevant symbolic representational system (Grigorenko et al. 2002; Wei et al. 2015). For example, hyperlexia—significantly enhanced single-word reading ability relative to other cognitive capacities—has been reported in 20 % of children with pervasive

developmental disorders, such as ASD (Grigorenko et al. 2002). Therefore, if strengths in symbolic processing are broad or general skills that extend to numerical stimuli, one would predict enhanced or spared symbolic acuity in children with ASD.

Next, we examined whether non-symbolic and symbolic acuity measures predict individual differences in math abilities, as assessed using the Math Composite measure of the Wechsler Individual Achievement Test-second edition (WIAT-II; Wechsler 2002). Meta-analytic studies in TD children have provided evidence for predictive associations with both measures, but few studies have assessed both symbolic and non-symbolic number sense in the same participants (De Smedt et al. 2013). Moreover, even when both are measured, direct comparisons between them can be influenced by high co-linearity. Here, we employ, for the first time in this domain, the Cox test (Davidson and MacKinnon 1981), which enables comparison of the relative explanatory power of non-nested models. Finally, we used a mediator model to investigate the pathways by which symbolic capacities may mediate the relationship between non-symbolic number sense and formal mathematics ability. Understanding the relative contributions of these two measures and any divergence from patterns observed in TD children promises to inform our understanding of the means by which children with ASD acquire skills in an area of great practical relevance.

Methods

Participants

Participants were recruited from schools and clinics in the San Francisco Bay Area. The study was approved by Stanford University's Institutional Review Board. Data were collected from 36 children (32 males) with high-functioning autism (i.e. normal intelligence) between the ages of 7–12 and a matched control group of 61 (54 males) typically developing participants. TD participants were selected from a larger study on the development of mathematical ability. This larger sample consisted of 54 male and 48 female participants. Our matching criteria were age and full-scale IQ. As male TD participants did not differ in these factors from the sample of male participants with ASD (both p s > .669), we included all male TD participants from the original sample ($N = 54$). To match the female participants with ASD, we used a genetic algorithm to select 7 female TD participants that were matched to our female participants with ASD on age and full-scale IQ (both p s > .354). The genetic algorithm works by randomly selecting 10,000 sets of 7 female TD participants and determining if their average age and full-scale IQ matches that of the female participants with ASD. The set most closely matching the ASD values is then selected for further processing by randomly swapping some of the participants producing a new set of potential participants. This process was repeated 5000 times until the groups did not differ on the designated factors.

All children in the ASD group had a clinical diagnosis of autism. In 31 of these participants, we were able to confirm a diagnosis within the autism range, through either the Autism Diagnostic Interview-Revised (ADI-R) or Module 3 of the Autism Diagnostic Observation Schedule (ADOS) (Table 1).

Standardized Measures of Cognitive Abilities and Academic Achievement

All participants were administered the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler 1999) to measure Verbal, Performance, and Full-Scale IQ (FSIQ). All participants had FSIQ scores above 75. No participants were excluded due to low IQ.

The WIAT-II (Wechsler 2002) was used to measure reading and mathematical ability. Reading ability was assessed using the Word Reading and Reading Comprehension subtests. Math ability was assessed using two mathematical subtests, the untimed arithmetic component called Numerical Operations and a Mathematical Reasoning component which assesses contextual understanding of mathematics using, for example, clock arithmetic, monetary questions, and word problems. To create a single measure of mathematics proficiency for our participants, we combined the Number Operations and Mathematical Reasoning subtests, following WIAT-II procedures, to compute a “Math Composite” score for each individual.

The Working Memory Test Battery-Children (WMTBC; Pickering and Gathercole 2001) was used to assess working memory capacity. The Digit Recall subtest measured phonological working memory, while Block Recall tapped visuo-spatial working memory. Two tests were used to assess central executive working memory: Counting Recall and Backwards Digit Recall. We computed a Working Memory Composite (WMC) score by first Z-normalizing the raw working memory scores across the full data set and then averaging the four measures.

Experimental Procedures

Participants were tested individually. The two quantitative judgment tasks described below, along with a third task not reported in this paper, were performed in a single session and were run on a Dell laptop computer using the E-Prime 2.0 suite. The order of the three tasks was counterbalanced among participants. Participants sat at a distance of approximately 50 cm from the computer screen.

Number Sense Tasks

Non-symbolic Number Sense Task—Participants completed a non-symbolic quantitative comparison task, modeled on a task previously used by Halberda and Feigenson (2008). In each trial, participants were presented with two sets of green dots occupying adjacent left and right panels of the computer screen, and were asked to judge which set contained the larger number of dots (Fig. 1a). Responses were collected on a Cedrus RB-530 button box (Cedrus Corporation, San Pedro, California) and participants indicated whether the left or right panel contained more dots using either a left- or right-handed button-press.

Each trial displayed a visual fixation for 500 ms, followed by the dot stimulus for 1500 ms, and finally a blank screen for 1000 ms (Fig. 1a). If the participant did not make a response within this time, the task automatically advanced to the next trial. Stimuli consisted of two sets of side-by-side dots, presented within rounded-corner boxes. All possible combinations of 2–9 dots that fell into the following ratio bins were included: 1:2, 2:3, 3:4, 4:5, 5:6, 6:7, 7:8, and 8:9. For example, a trial with a ratio of 1:2 might show a set of 4 green dots on the

left and 8 green dots on the right, or 6 dots on the right and 3 dots on the left. There were 13 possible combinations, and each pair was presented four times, twice with the greater number on the left and twice with it on the right, for a total of 52 trials in a block. To prevent participants from using cumulative area as an indicator of quantity, the total area of the left set of dots was approximately equal to the total cumulative area of the right set of dots in half the trials. These “Area-controlled” trials were counterbalanced with “Size-controlled” trials, in which the average dot-size in the left set was approximately equal to the average dot-size in the right set preventing participants from using dot-size as an indicator of quantity. In order to further reduce the correlation between area, size, and numerosity, the cumulative area and the average dot size of the left and right panels could differ by as much as 35 %, following Halberda et al. (2008).

Symbolic Number Sense Task—Participants also completed a symbolic version of this task, which used the exact same set of stimuli but presented as Arabic numerals (Fig. 1a). As in the non-symbolic (dots) task, participants were asked to indicate whether the left or right panel contained a larger quantity using a left- or right-handed button-press. The trial procedure for the numbers task was identical to that of the dots task, using numerals rather than dots as stimuli (Fig. 1a).

Weber Fraction (w) Calculation

Separately, for both the non-symbolic (dots) and symbolic (numbers) tasks, we calculated the Weber fraction (w) which best characterized each participant's pattern of responses. Conceptually, for any value n , a participant's perception of numerosity can be represented as a Gaussian distribution with a mean of n and a standard deviation of $n \times w$. The overlap between the Gaussian representations of a larger n_1 and smaller n_2 corresponds to the proportion of cases in which the participant incorrectly judges n_2 to be the larger quantity. Thus an individual's raw accuracy, measured by percentage correct, will be 1 minus this error rate. It follows that we can calculate expected percentage correct for any two unique quantities n_1 and n_2 as:

$$1 - \frac{1}{2} \operatorname{erfc} \left(\frac{(n_1 - n_2)}{\sqrt{2}w \sqrt{(n_1^2 + n_2^2)}} \right)$$

where erfc is the complementary error function.

For each task, we used the method developed by Price and colleagues to compute w (Price et al. 2012). For each participant, we fitted the model above with each possible value of w from 0 to 1 in increments of 0.01. We then calculated the error between the participant's accuracy and the model and selected the value of w which minimized the difference in sum of squares between the observed accuracy (percent correct) and expected accuracy (the value of the model with the given w). This method enabled us to estimate a value of w even for participants with noisy data that did not converge in accordance with Weber's law, a limitation of other calculation methods (Halberda et al. 2008). Repeating this process for

each task type yielded two values of acuity (w) for each participant: one computed for the non-symbolic (dots) task and one for the symbolic (numbers) one.

Regression and Mediation Analyses

We examined the relationship between math ability and number sense using regression models of the effects of IQ, age, working memory and symbolic or non-symbolic w on math ability. Because these models are not nested, we used the Cox test, implemented in the R library *lmtest* (Hothorn et al. 2014) to compare between them. Essentially, this statistic compares the fit of two models by assuming that if one model's parameters are complete, then the other model's parameters should not add any further explanatory value. We compared the beta-values produced by the Cox test between children with ASD and TD children using pooled two-sample t-statistics.

Further, we conducted mediation analyses to examine the contributions of symbolic and non-symbolic number sense to formal mathematics performance. Motivated by the proposal that non-symbolic number sense forms a foundation for symbolic number sense, which in turn leads to the acquisition of formal mathematics (van Marle et al. 2014), we evaluated whether symbolic number sense is the source of the relationship between non-symbolic number sense and mathematics performance and whether these models differ between our participant groups. This allowed us to understand not only the strength of these relationships but also the extent to which our data support the theorized relations between them. To do so, we modified the Sobel test function from the R library *multilevel* (Bliese 2013) to allow for mediation with common covariates across all models (here IQ, WMC, and age). We specifically tested whether symbolic w mediated the relationship between non-symbolic w and math ability in each group. We compared the betas for the indirect effects in children with ASD and TD children using pooled two-sample t-statistics.

Results

Neuropsychological Measures

Participants were explicitly matched on age and FSIQ; however, the ASD and TD groups did not differ on standardized measures of academic achievement (Word Reading, Reading Comprehension, Numerical Operations, and Mathematical Reasoning subcomponents of the WIAT-II, Table 1, all $ps > .40$) or working memory (Digit, Counting, Block and Backward Digit Recall measures of the WMTB-C, Table 1, all $ps > .22$). There was also no difference between groups on the Math Composite measure, obtained by combining Numerical Operations and Mathematical Reasoning scores ($t = -.263$, $p = .793$) nor on the Working Memory Composite (WMC; $t = -.029$, $p = .977$). Removing participants without a research diagnosis of autism did not change these results (all $ps > .10$).

Behavioral Performance on the Number Sense Tasks

Non-symbolic (Dots) Task—We computed accuracy, mean response time on correct trials, and a Weber fraction (w) for each participant on the dot array comparison task. Data were analyzed using non-parametric statistical tests as some of the measures were not normally distributed. A Mann–Whitney U test revealed significantly lower accuracy in

participants with ASD than in TD participants on the dots task ($Z = 2.313$, $p = .021$, Fig. 1b). Accordingly, participants with ASD also showed higher values of w than TD participants on this task ($Z = -2.895$, $p = .004$, Fig. 1d), indicating significantly worse acuity. There was no significant difference between groups in response time ($Z = 0.523$, $p = .601$, Fig. 1c). Removing participants without a research diagnosis of autism did not change this pattern (accuracy: $Z = 2.208$, $p = .027$; w : $Z = -2.955$, $p = .003$; response time: $Z = .268$, $p = .788$).

We next examined performance on the two subconditions of the dots task: Area-controlled trials, which equate total surface area across the two displays, and Size-controlled trials, which equate average dot-size across the two displays. For all participants, performance was slightly lower on the Area-controlled than Size-controlled trials (82.37 vs. 83.95 %; Figure S1), and this difference was marginally significant (Wilcoxon signed ranks test, $Z = 1.811$, $p = .07$). Participants in the ASD group were consistently worse than the TD group on both trials types (Area: $Z = 2.141$, $p = .032$; Size: $Z = 2.378$, $p = .017$). However, there was no interaction between trial type and group, as assessed by a Mann–Whitney U test on the difference score between the measures ($Z = 0.282$, $p = .778$). Group $\times$ trial type interaction in response time and w were also nonsignificant (all p s $> .59$), thus we used the combined data from both types of trials for all subsequent analyses.

Symbolic (Numbers) Task—We computed accuracy, mean response time on correct trials, and a Weber fraction (w) for each participant on the Arabic numbers comparison. Data were again analyzed using non-parametric statistical tests as some of the measures were not normally distributed. The ASD and TD groups did not differ on accuracy, response time, or w (all p s $> .53$) (Fig. 1b–d). Removing participants without a research diagnosis of autism did not change this pattern (all p s $> .41$).

Removing Participants with Erratic Response Patterns—Consistent with previous reports (Halberda et al. 2008), a small number of children (5 participants with ASD and 3 TD participants) displayed an erratic pattern of responses that did not fit the Weber function well (i.e. w values of .99 for one or both tasks). Removing these participants from the w comparisons did not change the pattern of results; participants with ASD still performed significantly worse on the dots task, ($p = .007$), but not the numbers task ($p = .990$).

Comparison of Behavioral Performance on Non-symbolic and Symbolic Tasks

We next used the Wilcoxon signed ranks test to examine performance differences between the tasks. Overall, performance was significantly worse on the dots task relative to the numbers task (accuracy: $Z = -3.353$, $p = .001$; w : $Z = -3.597$, $p < .001$). In order to formally test for an interaction between Group (ASD, TD) and Task (Symbolic, Non-symbolic) using non-parametric statistics, we computed a difference score between performance on the dots and numbers task for each individual. A Mann–Whitney U test revealed greater performance differences between the dots versus numbers comparison tasks in participants with ASD than TDs (accuracy: $Z = -1.967$, $p = .049$, w : $Z = -2.423$, $p = .015$). Follow-up analyses revealed that participants with ASD showed significantly lower accuracy and acuity (w) on the dots compared to numbers task (accuracy: $Z = -3.234$, $p = .001$; w : $Z = -3.490$, $p < .001$). These differences were non-significant in the TD participants for both accuracy ($Z =$

$-1.664, p = .096$) and w ($Z = -1.615, p = .106$). Participants' reaction times were slower on dot comparisons relative to number comparisons ($Z = -4.521, p < .001$), but there was no interaction between Group and Task ($Z = -1.232, p = .218$).

In order to confirm that the ratio manipulation affected behavior, we computed a 2 Type (dots, numbers) by 8 Ratio (1:2, 2:3, 3:4, 4:5, 5:6, 6:7, 7:8, 8:9) repeated measures ANOVA with Group (ASD, TD) as a between-subjects factor. As expected, there was an extremely robust significant main effect of Ratio ($F = 102.06, p < .001$) driven by lower accuracy on smaller ratios (i.e. 8:9) than larger ratios (i.e. 1:2) (see Fig. 2). There was also a main effect of Type with higher accuracy on the numbers task than the dots task ($F = 21.04, p < .001$) and a Type by Ratio interaction ($F = 5.69, p < .001$) driven by lower performance on dots than numbers in the middle range of ratios but not the ends. Consistent with the analyses of overall accuracy and w , there was no main effect of Group ($F = 2.54, p = .114$), but there was a Group by Type interaction ($F = 4.60, p = .034$) driven by children with ASD being less accurate than TD children on the dots task ($F = 4.99, p = .028$) but not the numbers tasks ($F = .60, p = .442$, see Fig. 2). There were no other interactions (all $ps > .10$).

Relation Between Number Sense and Math Ability

Correlations Between Non-symbolic and Symbolic Acuity and Math Ability—

To test the hypothesis that numerical acuity—whether symbolic or non-symbolic—predicts math ability (see Halberda et al. 2008), we systematically assessed the relationship between standardized measures of math ability and number sense acuity on the non-symbolic and symbolic comparison tasks. Due to the non-normal distribution of w and to facilitate regression analyses, we first log-transformed the number sense measures. We used Pearson correlations to examine their relation with FSIQ, WMC, age, and Math Composite scores of the WIAT-II scale (Table 2), which were comparable to Spearman correlations on the raw values (see Table S1). Both symbolic and non-symbolic w predicted math ability in TD children (dots: $r = -.29, p = .023$; numbers: $r = -.38, p = .003$). The same pattern held in children with ASD (dots: $r = -.36, p = .045$; numbers: $r = -.50, p = .002$).

Relation Between Non-symbolic and Symbolic Acuity and Math Ability,

Controlling for IQ, Working Memory and Age—A standing concern in studies of number sense and math ability is whether domain-general cognitive capacities drive the relationships between these measures. Additionally, number sense acuity is not a static capacity but improves with age (Halberda and Feigenson 2008). To address these issues, we performed multiple linear regressions to assess the effects of symbolic and non-symbolic acuity on math ability, controlling for FSIQ, WMC and age. In these regression models, for TD participants, neither non-symbolic nor symbolic w were significant predictors of math ability (dots: $p = .274$; numbers: $p = .328$, Table 3; Fig. 1e). In contrast, for participants with ASD, non-symbolic w still marginally predicted math ability ($p = .066$), while symbolic w was highly significant ($p = .002$, Table 4; Fig. 1f).

Comparison of Symbolic Versus Non-symbolic Number Sense Models—

Because symbolic and non-symbolic acuity were significantly correlated ($rs > .590, ps < .001$), we next examined whether models including symbolic or non-symbolic acuity

differentially predict math ability. To compare the fits of the symbolic and non-symbolic models to each other, we used the Cox test (Davidson and MacKinnon 1981), implemented in *R* library *lmtest* (Hothorn et al. 2014), which assesses whether the fit of one model (e.g. symbolic acuity model) adds explanatory value over and above another model (e.g. non-symbolic acuity model). We found that for ASD children, the symbolic acuity model added significant explanatory value over and above the non-symbolic acuity model ($\beta = -4.93$, $SE = .90$, $p < .001$). But the reverse is not true; that is, the non-symbolic acuity model did not add significant explanatory value beyond the symbolic acuity model ($\beta = -.27$, $SE = 1.39$, $p = .843$). In contrast, for the TD group, neither measure added explanatory value over and above the other model (all $ps > .25$). This difference in predictability of symbolic versus non-symbolic number sense models was significantly stronger in the ASD, compared to the TD group ($t = -4.79$, $p < .001$). These results indicate that symbolic number sense ability holds significantly more explanatory power, over and above non-symbolic number sense, in the ASD group than for their TD peers.

Mediating Influence of Symbolic Number Sense on the Relation Between Non-symbolic Number Sense and Math Ability

To further assess the differential effects of symbolic and non-symbolic number sense on math ability, we used mediation analyses to test the hypothesis that the relationship between non-symbolic number sense and math ability is mediated by symbolic number sense. We conducted a formal mediation analysis using the *R* library *multilevel* (Bliese 2013) to assess the effect of symbolic acuity on the relationship between non-symbolic acuity and math ability while controlling for FSIQ, WMC and age. In the ASD group, there was a significant indirect effect of symbolic w in mediating the relationship between non-symbolic w and math ability ($Z = -2.17$, $p = .030$, Fig. 3, top panel). In the TD group, there was no indirect effect of symbolic w in mediating the relationship between non-symbolic w and math ability ($Z = -0.48$, $p = .633$, Fig. 3, bottom panel). Comparing the mediation effects in the two groups revealed that this effect was significantly stronger ($t = -2.22$, $p = .029$) in the participants with ASD ($ab = -18.76$, $SE = 8.64$) compared to the TD participants ($ab = -1.50$, $SE = 3.14$). These results indicate that symbolic number sense mediates the relationship between non-symbolic number sense and math ability in the ASD, but not the TD, group.

Relation Between Symbolic and Non-symbolic Acuity and ASD Clinical Measures

There were no significant relations between any of the subscales in the ADI-R or ADOS and any of accuracy, w , or response time for either the non-symbolic or the symbolic tasks (all $ps > .05$).

Discussion

In this investigation, we addressed three questions: (1) do children with ASD differ from their TD peers in non-symbolic and symbolic number sense, (2) are these capacities associated with mathematics ability in the ASD population, and if so, (3) is this pattern of association differently mediated in ASD compared to the TD group? We found that children with ASD performed at similar levels to their TD peers when comparing symbolic stimuli

but were significantly worse when comparing non-symbolic stimuli. In children with ASD, symbolic acuity was a strong predictor of math ability even after accounting for IQ, working memory and age. In contrast, the same analysis did not reveal any such relation in TD children. Importantly, symbolic numerical acuity mediated the relationship between non-symbolic acuity and math ability in children with ASD, but not TD children. Together, these results suggest that symbolic quantity representations play a more crucial and distinctive role in the development of math skills in children with ASD than in their TD peers.

Symbolic and Non-symbolic Number Sense in Children with ASD

Based on previous findings reporting superior visuo-spatial processing skills (Mitchell and Ropar 2004), attention to local features (Rinehart et al. 2000), and savant skills in non-symbolic estimation (Scripture 1981), we hypothesized that non-symbolic number sense might be enhanced in individuals with ASD. Instead, we found that children with ASD were significantly impaired on a non-symbolic comparison task relative to their TD peers. This pattern is broadly consistent with previous studies on non-symbolic number sense in ASD which also failed to find superior quantity estimation in individuals with ASD (Meaux et al. 2014; Titeca et al. 2014; Turi et al. 2015). While attention to local features may be beneficial in tasks requiring identification of individual items, such as the Navon Task (Muth et al. 2014; Navon 1977), our results suggest that this skill does not translate to enhanced non-symbolic quantity processing in children with ASD.

The reduced performance on non-symbolic comparison reported here contrasts with a previous study in pre-school children that found no differences in non-symbolic processing (Titeca et al. 2014); however, children with ASD were significantly older than TD children. Given that performance on this task improves rapidly in the 6–7 age range studied (Halberda and Feigenson 2008), we might have expected the older ASD group to perform better than their younger TD peers. Our findings align with the recent study by Turi et al. (2015) who found reduced adaptation for non-symbolic quantity in children with ASD. Although non-symbolic number sense itself could not be adequately determined from their experimental paradigm, their findings nevertheless suggest weakness in integration of complex visuo-spatial quantity information.

Previous studies in TD children (Szucs et al. 2013) have suggested that Area-controlled trials (which equate stimuli based on total surface area) are harder than Size-controlled trials (which equate stimuli based on average surface of each dots). This effect is likely driven by Area-controlled trials requiring participants to resolve a potential conflict between the surface area and the number of items in the display, a process thought to depend on executive function capacities (Fuhs and McNeil 2013; Gilmore et al. 2013). In our study, there were no differences between the groups in executive function, as measured by a comprehensive working memory assessment (Table 1). Nevertheless, based on reports that executive function is impaired in children with ASD (Christ et al. 2007), we examined the possibility that poor performance on the non-symbolic (dots) trials in the ASD group may be driven by difficulties in processing the Area-controlled trials. However, we did not find a significant group by trial-type interaction (Figure S1). Together, these results suggest that

difficulties children with ASD have with non-symbolic stimuli are not related to executive dysfunction.

Based on reports of precocious reading abilities and hyperlexia in children with ASD (Grigorenko et al. 2002; Wei et al. 2015), we considered the alternate hypothesis that these children might have spared or enhanced symbolic number sense capacities. Indeed, despite their poor performance on the non-symbolic (dots) comparison task, children with ASD did not differ from their TD peers on the symbolic (numbers) comparison task. Although this capacity was not enhanced relative to TD participants, it was significantly better than the performance of participants with ASD on the non-symbolic (dots) task. This pattern of results suggests that children with ASD may use alternative cognitive and brain mechanisms to achieve normal symbolic numerical acuity even in the presence of impaired non-symbolic number sense.

Predictors of Math Ability in Children with ASD

There has been considerable debate regarding the relative contributions of symbolic and non-symbolic number sense to overall math abilities in neurotypical children and adults (De Smedt et al. 2013). While some studies have pointed to the primacy of non-symbolic capacities (Halberda et al. 2008), others have suggested a stronger relation between symbolic number sense and math abilities (Durand et al. 2005; Holloway and Ansari 2009; Kolkman et al. 2013). The emerging consensus based on a recent review of studies in TD children is that symbolic number sense may be a more robust predictor of math skills (De Smedt et al. 2013). Consistent with this analysis, we found that in children with ASD, after accounting for the effects of IQ, working memory and age, only symbolic number sense significantly predicted math ability. This result is consistent with the one previous study examining non-symbolic number sense predicting math skills, which found comparable predictability between children with ASD and TD, but no residual relationship after accounting for IQ (Titeca et al. 2014).

To further compare the explanatory power of symbolic and non-symbolic acuity in predicting math abilities, we next computed the Cox test (Davidson and MacKinnon 1981). Uniquely in children with ASD, symbolic acuity added significant explanatory value to the model with non-symbolic acuity while the reverse was not true. Our findings help resolve some of the controversies regarding the differential role of symbolic and non-symbolic number sense in predicting math abilities. In TD children, previous studies that have investigated both measures have tended to find symbolic, but not non-symbolic, acuity to be a significant predictor (Holloway and Ansari 2009; Kolkman et al. 2013; Lonnemann et al. 2011; Mundy and Gilmore 2009; Vanbinst et al. 2012). A direct comparison between the predictive power of these constructs is complicated by the high correlation between non-symbolic and symbolic acuity (in our sample all r s > .590, p s < .001). Traditional step-wise regression analyses can identify the strongest predictor, but they cannot differentiate between the explanatory powers of these highly collinear measures. We addressed this directly by employing, for the first time, the Cox test (Davidson and MacKinnon 1981), which we suggest is a powerful tool for comparing models with highly correlated regressors.

Together, our results demonstrate for the first time that symbolic number sense is a stronger and unique predictor of math ability in children with ASD.

The Differential and Critical Role of Symbolic Number Sense in Mediating Math Ability in Children with ASD

We found that although children with ASD displayed weaker non-symbolic acuity, they did not differ in overall math ability from their TD peers. In TD children, it has been hypothesized that non-symbolic number sense provides a foundation for symbolic capacities, which in turn contributes to formal math ability (Von Aster and Shalev 2007). However, as described in the previous section, our data demonstrate that even after accounting for non-symbolic acuity, individual differences in symbolic acuity are significantly more predictive of math ability in children with ASD. This result suggests that symbolic number sense is not entirely attributable to non-symbolic abilities in children with ASD. The tight relationship between symbolic number sense and math performance in our participants with ASD—stronger than in their TD peers—suggests that children in this population may be more dependent on symbolic number sense capacities for math problem solving.

To examine this hypothesis, we employed a mediation analysis to systematically test whether the relationship between non-symbolic acuity and math ability is mediated by symbolic acuity. We found a significant mediation effect in the ASD group such that including symbolic w in the model rendered the indirect path (c') non-significant. Furthermore, direct comparison of the size of the indirect effects (ASD: $ab = -18.76$, $p = .030$ vs. TD: $ab = -1.59$, $p = .619$) revealed that the mediation effects were stronger in the ASD than the TD group. The source of this between-groups difference was the predictive influence of symbolic w on math ability in ASD ($b = -27.00$, $p = .015$) but not in TD ($b = -4.66$, $p = .619$) participants. Previous studies in TD samples have identified a role for symbolic number knowledge in mediating the relationship between non-symbolic number sense and math ability (Lyons and Beilock 2011; van Marle et al. 2014). Critically, these studies used multiple measures of symbolic knowledge (e.g. symbolic acuity during numeral comparison, number recognition, ordinality principles) and failed to find a significant mediation effect of symbolic numerical acuity. These results provide support for the view that symbolic number sense specifically contributes to spared or sometimes enhanced math ability in children with ASD, potentially allowing them to overcome impairments in their non-symbolic number sense capacities.

Links to Other Cognitive Strengths in Autism

Previous research has linked mathematics ability in ASD to a propensity for systematization and creation of deterministic input-to-output rules (Baron-Cohen 2009), and investigations of other savant talents have proposed that facility with rule-based systems might enable individuals with ASD to, for example, act as calendar calculators, or produce masterful musical compositions (Cowan and Frith 2009; Heavey et al. 1999). One common thread across these areas of strength is that they leverage abstract, symbolic notations to organize continuous measures such as quantity, time, and pitch. This trend may suggest a more general principle for helping individuals with ASD organize and understand complex, fuzzy

systems such as social communication and language (Qian and Lipkin 2011), and is a topic that warrants further investigation.

Limitations and Future Work

Our study has focused on a group of high-functioning children with ASD (IQ ≥ 75 , age 7–12 years). Characterizing symbolic and non-symbolic number sense in low-functioning children with ASD remains a high priority for future research as these children may benefit the most from targeted interventions. Further studies examining developmental changes between childhood and adolescence in ASD are also needed. Symbolic number sense is only one of many competencies that contribute to the acquisition of more complex mathematical skills. Crucially, early in development, children acquire mathematical knowledge by building on their intuitive sense of non-symbolic quantity to understand number words and symbols and ultimately arithmetic concepts (Von Aster and Shalev 2007). In the present study, we found that despite deficits in processing non-symbolic quantities, children with ASD were still able to acquire basic mathematical knowledge. This finding suggests that symbolic skills may provide alternative pathways to math skill acquisition in this population. More work is needed to investigate how children with ASD can leverage their preserved skills to further develop their mathematical skills. In addition, the role of other foundational skills, such as ordinality (Lyons and Beilock 2011) and number line estimation (Fazio et al. 2014) should also be investigated. Finally, further work is also needed to investigate how difficulties in non-symbolic number sense can be remediated in children with ASD. Such work should use training paradigms to probe the causal role of precursor capacities on formal math abilities and to provide insights into more effective methods for enhancing mathematical problem solving skills in this population.

Conclusions

Our study provides a detailed investigation of number sense and its relation to mathematics ability in children with ASD. We found that non-symbolic number sense was impaired in children with ASD, while symbolic performance was comparable to that of TD children. Surprisingly, the influence of symbolic number sense on math ability was stronger in children with ASD than in TD children, and symbolic numerical acuity mediated the relation between non-symbolic acuity and math ability in the ASD group only. These results point to a more critical and distinctive role for symbolic knowledge in the development of mathematical skills in children with ASD. Our findings indicate that symbolic skills are a preserved strength in children with ASD and suggest that leveraging symbolic number knowledge may help them develop mathematical proficiencies.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Fig. 1.

Experimental task, behavioral performance and relation with math ability. **a** In the non-symbolic Dots task participants saw a fixation for 500 ms followed by the two dot arrays presented side-by-side for 1500 ms, followed by a blank screen for 1000 ms. In the symbolic Numbers task, participants saw Arabic numerals rather than dot arrays. **b** Children with autism spectrum disorders (ASD) had lower accuracy on the Dots task than typically developing (TD) children ($*p < .05$), but did not differ on the Numbers task. **c** There were no differences between the groups in reaction time. **d** Children with ASD had larger Weber fractions (w) than TD children on the Dots task ($*p < .05$) but did not differ on the Numbers task. **e** In TD children, after accounting for IQ, Working Memory Composite and age, performance on both the Dots and Numbers tasks was not correlated with math ability (all p s $> .34$). NB: two participants with extremely low w s are not included for visualization purposes, but are included in computation. **f** In children with ASD, after accounting for IQ,

Working Memory Composite and age, performance on the Dots task was not correlated with math ability performance, but performance on the Numbers task was correlated with math ability (** $p < .001$)

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Fig. 2.

Ratio effect. The ratio of numerical quantity significantly affected accuracy on both the Dots and Numbers tasks ($p < .001$), for both children with autism spectrum disorders (ASD) and typically developing (TD) children. Children with ASD were significantly less accurate on the Dots task than their TD peers ($p = .028$), but did not differ on the Numbers task ($p = .442$).

Fig. 3.

Mediation analysis. The mediating effect of symbolic acuity w (Numbers task) on the relationship between non-symbolic acuity w (Dots task) and math ability (WIAT-II Math Composite measure) was significant ($p = .030$) in (*top*) children with autism spectrum disorder (ASD) and but not (*bottom*) typically developing (TD) children ($p = .633$)

Table 1

Participant demographics

Measure	Group		<i>t</i>		<i>p</i>
	ASD (n = 36)		TD (n = 61)		
	M	SD	M	SD	
Males/females	32/4		54/7		
Age (years)	9.66	1.60	9.60	1.49	.192 .848
WASI					
Verbal IQ	104.92	17.84	110.84	16.93	-1.631 .106
Performance IQ	111.64	23.30	108.54	15.20	.793 .430
Full-Scale IQ	109.19	20.45	110.90	15.58	-.463 .645
WIAT-II					
Word reading	110.33	12.92	108.43	9.44	.836 .405
Reading comprehension	106.11	15.17	107.15	10.54	-.396 .693
Numerical operations	105.67	23.57	108.25	17.56	-.614 .541
Mathematical reasoning	109.94	22.36	110.69	14.52	-.151 .881
<i>Math Composite</i>	109.19	25.39	110.33	16.98	-.263 .793
WMTB-C (Raw)					
Digit recall	28.61	6.38	27.13	5.31	1.230 .222
Counting recall	17.75	7.33	19.28	5.73	-1.142 .256
Block recall	23.14	5.28	24.20	4.85	-1.004 .318
Backward digit recall	12.72	5.59	11.74	3.97	1.012 .314
<i>Working Memory Composite</i>	.01	.92	-.01	.71	.029 .977
ADOS <i>I</i>					
Social	8.07	2.27			
Communication	3.52	1.68			
ADI-R ²					
Social	19.30	5.71			
Communication	15.57	5.14			
Repetitive behaviors	5.57	2.66			

Measure	Group		<i>t</i>	<i>p</i>
	ASD (n = 36)	TD (n = 61)		
Developmental abnormalities	M	SD	M	SD
	3.17	1.58		

ASD samples:

1 n = 29,

2 n = 30

WASI/Wechsler Abbreviate Scales Of Intelligence, *WIAS-II* Wechsler Individual Achievement Test-second edition, *WMTB-C* Working Memory Test Battery-Children, *ADOS* Autism Diagnostic Observation Schedule, *ADI-R* Autism Diagnostic Interview-Revised

Table 2
Pearson correlation between cognitive measures, numerical acuity measures, and age

Variable	FSIQ	WMC	Age	Log (w Dots)	Log (w Numbers)
TD					
Math composite	.59 ***	.61 ***	.24 *	-.29 *	-.38 **
FSIQ		.48 ***	.06	-.15	-.19
WMC			.50 ***	-.32 *	-.54 ***
Age				-.30 *	-.35 **
Log (w Dots)					.59 ***
ASD					
Math composite	.63 ***	.67 ***	-.01	-.36 *	-.50 **
FSIQ		.57 ***	-.02	-.33 *	-.21
WMC			.45 **	-.46 **	-.42 *
Age				-.57 ***	-.37 *
Log (w Dots)					.67 ***

FSIQ Full Scale Intelligence Quotient, *WMC* Working Memory Composite

* $p < .05$;

** $p < .01$;

*** $p < .001$

Table 3
Regression analysis of numerical acuity (*w*) predicting math ability, after controlling for Full-Scale IQ (FSIQ), Working Memory Composite (WMC), and age, in TD children

Coefficients	β	$\hat{\beta}$	SE	t	p
TD (Dots, adjusted $R^2 = .463$)					
Intercept	60.84		20.28	3.00	.004**
FSIQ	.42	.39	.12	3.48	.001**
WMC	9.55	.40	3.10	3.08	.003**
Age	-.19	-.02	1.30	-.15	.885
Log (<i>w</i> Dots)	-5.60	-.11	5.07	-1.11	.274
TD (Numbers, adjusted $R^2 = .461$)					
Intercept	55.98		21.36	2.62	.011*
FSIQ	.43	.40	.12	3.58	.001**
WMC	8.66	.36	3.39	2.55	.013**
Age	-.06	-.01	1.29	-.04	.966
Log (<i>w</i> Numbers)	-7.66	-.11	7.76	-.99	.328

Separate analyses were conducted for dots and numbers
FSIQ Full-Scale Intelligence Quotient, *WMC* Working Memory Composite
* $p < .05$;
** $p < .01$

Table 4

Regression analysis of numerical acuity (*w*) predicting math ability, after controlling for Full-Scale IQ (FSIQ), Working Memory Composite (WMC), and age, in children with ASD

Coefficients	β	$\hat{\beta}$	SE	t	p
ASD (Dots, adjusted $R^2 = .606$)					
Intercept	143.20	32.57	4.396		***
FSIQ	.22	.18	.18	1.25	.221
WMC	18.07	.65	4.23	4.28	<.001***
Age	-7.40	-.47	2.34	-3.16	.004**
Log (<i>w</i> Dots)	-20.91	-.27	11.00	-1.91	.066
ASD (Numbers, adjusted $R^2 = .677$)					
Intercept	116.28	28.04	4.15		***
FSIQ	.30	.24	.15	1.96	.059
WMC	15.53	.56	3.92	3.97	<.001***
Age	-6.50	-.41	1.86	-3.49	.001**
Log (<i>w</i> Numbers)	-28.03	-.37	8.34	-3.36	.002**

Separate analyses were conducted for dots and numbers

FSIQ Full-Scale Intelligence Quotient, WMC Working Memory Composite

**
 $p < .01$;

 $p < .001$