MEMORY IN 3-YEAR-OLD CHILDREN WITH AUTISM

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ABSTRACT

DEBRA M. CHILDRESS: Memory in 3-year-old children with autism (Under the direction of J. Steven Reznick, Ph.D.)

Autism is a severe neurodevelopmental disorder characterized by deficits in communication and language skills; social behavior; and a restricted range of activities and interests (DSM-IV; American Psychological Association-APA, 1994). Furthermore, it is well documented that individuals with autism display a pattern of deficits and strengths in cognitive ability. Although considerable effort has been applied to investigating memory in autism, agreement has not been reached with respect to the status of almost any aspect of memory functioning in individuals with autism. Furthermore, we know little about the early development of any component of the memory system in autism due in large measure to methodological limitations. These factors leave the status of the characterization of memory abilities in autism as inadequate. The current study examines working memory, long-term memory and language ability in two groups of children: children with autism and typicallydeveloping children matched to children with autism on receptive verbal mental age and gender. Working memory for this study was assessed using two spatial delayed response and two verbal working memory tasks. Long-term memory was examined using deferred imitation. Children were presented with four four-sequence events: two arbitrary (no clear relationship between the actions in the sequence) and two enabling (actions that needed to be completed in order for the subsequent action to be completed) sequences. Results indicate

that working memory as measured on spatial and verbal tasks is significantly lower in children with autism. Additionally, children with autism showed significantly lower performance on action and action pairs generated as part of the deferred imitation task. However, both groups of children demonstrated memory gains as demonstrated by a significant difference in number of actions and action pairs that occurred during the baseline and recall phases. Potential applications of these working memory tasks and deferred imitation procedures are discussed.

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I. INTRODUCTION

Autism is a severe neurodevelopmental disorder characterized by deficits in three main domains: (1) communication and language skills; (2) social behavior; and (3) a restricted range of activities and interests (DSM-IV; American Psychological Association-APA, 1994). Behavioral heterogeneity in each of the core domains can be found across individuals with autism. For example, 50% of children with autism remain mute throughout their life, while other individuals with autism are affected by more subtle pragmatic language deficits. In the social domain, some children exhibit extreme aloofness or avoidance of others while other children express interest in social interaction but have difficulty learning and applying social rules. Finally, repetitive behaviors may be expressed as part of motor stereotypies, a pronounced preference for sameness or elaborate routines that cause significant distress when interrupted. Furthermore, although cognition per se is not included among the three core diagnostic descriptors of the disorder, it is well documented that individuals with autism display a pattern of deficits and strengths in cognitive ability. For example, 70% of individuals with autism have some degree of mental retardation (Fombonne, 2003). Paradoxically, 10% of individuals with autism are reported to have a "savant" or special skill, defined as an ability that far exceeds expectations based on the person's IQ such as calendrical calculation or exceptional rote memory (Mottron, Belleville, Stip, & Morasse, 1998. See Heaton and Wallace, 2004, for a review of this topic).

Descriptions of human cognition are inherently complex, encompassing a vast array of processes such as memory, attention, perception, problem solving and mental imagery, to identify just a few, and researchers and clinicians have investigated each of these domains to discover the role that it might play in autism symptomatology (Pascualvaca, Fantie, Papageorgiou, & Mirsky, 1998; Garretson, Fein, & Waterhouse, 1990; Leslie & Thaiss, 1992). The status of memory function in autism is unique in that it has been characterized as both a primary source of autism symptomatology, such as in the amnesia theory (DeLong, 1992) and a secondary source, in that it reflects a more generalized cognitive deficit that transcends memory, such as in executive function deficits (Williams, Goldstein, Carpenter, & Minshew, 2005). Although considerable effort has been applied to investigating memory in autism, agreement has not been reached with respect to the status of almost any aspect of memory functioning in individuals with autism. Furthermore, while efforts aimed at early identification of children with autism have been increasingly successful, we know little about the early development of any component of the memory system in autism due in large measure to methodological limitations. These factors leave the characterization of the status of memory abilities in autism as inadequate.

1.1 Memory in Autism

Kanner did not provide expansive descriptions of the cognitive profiles of the eleven children in his original description of autism, but he did emphasize the memory abilities of the children (Kanner, 1943):

Their excellent rote memory, coupled with the inability to use language in any other way, often led the parents to stuff them with more and more verses, zoological and botanic names, titles and composers of victrola record pieces, and the like. Thus from the start, language- which children did not use for the purpose

of communication- was deflected in a considerable measure to a self sufficient, semantically, and conversationally valueless or grossly distorted memory exercise. To a child of 2 or 3 years old, all these words, numbers, and poems (questions and answers to the Presbyterian Catechism; Mendelssohn's violin concerto; a French lullaby; an encyclopedia index page) could hardly have more meaning than sets of nonsense syllables to adults, (p. 230) Clearly, the role of memory has been in question from the beginning of efforts to

understand autistic symptomatology. Although there are numerous accounts of exceptional memory abilities in children with autism (Mottron et al., 1998), the pattern of spared and deficient functions seems to be present across the autism spectrum.

A detailed discussion of memory must start with a definition for the broad construct. Memory is the aspect of cognition that refers to the representation, storage, control, maintenance, retrieval, and use of various kinds of information including, but not limited to, aspects of experience (Pelphrey & Reznick, 2003). Although theories in autism have rarely focused primarily on memory, with the notable exception of the amnesia theory, understanding the development of memory in autism is of critical relevance to understanding the underlying etiology of the disorder, as well as, providing a point for intervention. In fact, from an intervention perspective, the unique pattern of intact and impaired abilities may provide both tools for intervention, through intact abilities, and opportunities for intervention, through impaired abilities.

Because the definition of memory refers to a broad set of interrelated processes with their own distinct properties, it may be helpful to select a model of memory to frame the discussion of memory in autism. Because many information processing models divide memory into short-term and long-term components (e.g., Shiffrin & Atkinson, 1969), this model has been selected. It provides a familiar account of memory from a common information-processing model; therefore, it may have utility for a broader

audience. However, it should be noted that any account of memory development would ultimately have to integrate all of the memory systems regardless of the classification system used.

1.2 Short-term and working memory in autism

Short-term memory refers to the process through which information is temporarily stored before being acted upon, transferred to long-term memory, or discarded from memory. Short-term memory can be contrasted with working memory, in which temporarily stored information is manipulated, thus contributing to the performance of cognitive tasks such as language comprehension, reasoning, decisionmaking, problem solving, and mental arithmetic (Baddeley, 1986, 1992; Baddeley & Hitch, 1974). The examination of short-term and working memory in autism has largely been embedded in the investigation of executive dysfunction , although support for the absence and presence of short term and working memory deficits have been generated by almost all of the major theoretical perspectives (Minshew & Goldstein, 2002; Bennetto, Pennington, & Rogers, 1996; Ozonoff & McEvoy, 1994; Hill and Russell, 2002).

1.2.1 Short-term memory

Similar to working memory, performance on short-term memory tasks can be mediated via verbal (or phonological) and non-verbal (or visual-spatial) paths. Both aspects of short-term memory have been examined in children with autism. Individuals with autism appear to have intact phonological short-term memory. Boucher (1978) found that children with autism were not impaired on echoic memory, relative to typically developing controls. More recently, Russell, Jarrold, and Henry (1996) examined articulatory loop abilities in 9 to 15 year-old children with autism. For both

verbal recall (i.e., repeating high-frequency nouns immediately after presentation) and nonverbal recall (i.e., pointing to pictures of verbally presented words), the performance of children with autism was superior to that of children with moderate learning difficulties. Carpentieri and Morgan (1994) did not find impaired performance on a verbal short-term memory task when the performance from a group of low-functioning children with autism was compared to a group of children with mental retardation. Minshew and Goldstein (2002) did not find significant differences between adults with high-functioning autism and typically developing controls on short-term auditory memory tasks or immediate recall of words presented as part of a paired associate task. Notably, span tests of words and instructions produced significant differences in the autism and comparison groups while span test of letters did not (Minshew and Goldstein, 2002). These results suggest that the complexity of the material influences task performance in individuals with autism more severely than in typically developing individuals. However when considering results that rely on the complexity of material, complexity must always be considered in relation to an individual's knowledge base. That is, what is complex for one individual may not represent the same level of complexity to another. In studies of adults with autism, Rumsey and Hamburger (1988) reported unimpaired immediate and delayed verbal memory functioning. Bennetto et al. (1996) found that high-functioning children and adolescents with autism showed intact performance on short-term verbal memory measures from the California Verbal Learning Test when compared to individuals with learning disabilities. Verbal recall was tested in three groups of children: children with autism, children with PDD-NOS and a group of

typically developing children (Buitelaar, van der Wees, Swaab-Barneveld, & van der Gaag, 1999), and no difference in performance was found among the groups.

Short-term visual recognition memory also appears to be intact in autism. Barth, Fein, and Waterhouse (1995) found that four- to five-year old high- and low-functioning children with autism, subjects with developmental language disorders, and those with mental retardation, all performed similarly on visual recognition memory tasks, when covaried on nonverbal intelligence. Visual memory recall was tested in three groups of children: children with autism, children with PDD-NOS and a group of typically developing children from the previously mentioned study by Buitelaar and colleagues (Buitelaar et al., 1999). No difference in performance was found among the groups. Carpentieri and Morgan (1994) also found unimpaired performance on a visual spatial short-term memory task when comparing performance from a group of low-functioning children with autism to a group of children with mental retardation.

In summary, individuals from childhood to adulthood, and across the autism spectrum, seem to display consistently intact short-term phonological and visual recognition memory abilities, and this pattern of results is consistent regardless of comparison group, (see Burack, 2004, for a review of the implications of comparison group selection).

1.2.2 Working memory

As previously discussed, working memory is considered to be one aspect of executive functioning in that it captures the organizational aspects of memory and the active use of representations in planning behavior (Pennington & Ozonoff, 1996). Specifically, working memory is an active process involving the concurrent storage and

processing of representations, often in the face of distraction and interference, and the use of those representations to plan and constrain goal-directed behavior (Baddeley, 1986). Although the role of working memory has been examined as part of executive dysfunction and information processing models of autism, the results of working memory studies in autism have been inconsistent.

Evidence supporting working memory impairment in autism was initially provided through studies of performance on Tower of Hanoi and London tasks (Ozonoff & McEvoy, 1994; Ozonoff & Strayer, 2001). Because these tasks require the participants to generate and hold potential moves in memory while considering other moves, they have been considered tasks of working memory (Ozonoff, Pennington, & Rogers, 1991). Tower of Hanoi (or London) performance has been found to be significantly impaired in several studies (Ozonoff et al., 1991; Bennetto et al., 1996; Ozonoff & McEvoy, 1994; Hughes, Russell, & Robbins, 1994) and remains one of the most robust and consistent findings in support of executive dysfunction in autism (Ozonoff & Strayer, 2001). Additional evidence for impaired working memory also emerged in a group of highfunctioning adults with autism who completed two verbal working memory tasks: a counting and a sentence span task (Bennetto et al., 1996). These results remained even after verbal short-term memory was co-varied. In contrast, Russell et al. (1996) did not find working memory deficits, as measured by dice counting and sentence completion tasks, in a comparison of children with autism and matched children with mental retardation.

A similar pattern of inconsistent findings is present in studies of non-verbal working memory. Dawson et al. (Dawson, Meltzoff, Osterling, & Rinaldi, 1998)

administered delayed non-match to sample and delayed response tasks to children with autism then compared their performance to children with Down syndrome and typical development. Delayed non-match to sample and delayed response tasks have been used extensively to study nonverbal working memory in typically developing children (for a review see Reznick, 2007). Young children with autism, aged 3 to 4.5 years old, performed significantly worse on both tasks compared to both comparison groups (Dawson et al., 1998). This finding is supported by evidence from non-human primate studies in which delayed non-match to sample tasks were found to be deficient in primates who had received a medial-temporal lesion (Bachelavier, 1994). A different study completed with adolescents and young adults with autism found deficits in spatial working memory as measured by an occulomotor delayed response task (Minshew, Luna, & Sweeney, 1999). This task assesses the capacity for making a saccadic eye movement to the location of the target previously presented in the periphery and has been widely used in primate and human research to define the circuitry of the prefrontal cortex (Minshew et al., 1999). Landa and Goldberg (2005) found that adolescents with high functioning autism were impaired on a spatial working memory task when the working memory load was high (6- and 8- locations that could be searched) but they were not impaired when the load was low (3- and 4- search locations).

In contrast, Ameli, Courchesne, Lincoln, Kaufman, and Grillon (1988) found that adults with autism were not impaired on relatively simple visual memory tasks compared to sex- and age-matched typically developing comparison group. Impairments were observed, however, when information to be encoded was complex or when organizational processes to aid recall were required (Ameli et al., 1988). More recently, negative

evidence emerged in a study of working memory by Ozonoff and Strayer (2001). In this study, three computerized tasks of spatial working memory were presented to groups of children with autism, Tourette syndrome and typical development. Although there were significant effects of memory load and delay, no working memory parameter interacted significantly with the group variable (Ozonoff & Strayer, 2001). The authors suggest that the pattern of inconsistent results from working memory tasks may be accounted for by the computerized format of the tasks. Specifically, tasks administered by humans, which required some degree of social interaction to indicate correct response, amplify a cognitive impairment that may otherwise be borderline (Ozonoff & Strayer, 2001). However, the spatial task used by Landa and Goldberg (2005) was also computerized and this study found evidence for working memory deficits when memory load was greater. Ozonoff and Strayer (2001) also suggest that mixed results in the literature on working memory may be due to sample specific characteristics. That is, older and highfunctioning individuals with autism do not display, or perhaps no longer display, working memory impairment. Although not mentioned by the authors, it is possible that working memory load may have to be increased to cause the task scores of the individuals with autism to diverge from those of mental age matched controls. This would be consistent with the complex information-processing model of Minshew and Goldstein (1998) and the results obtained as part of the Landa and Goldberg (2005) study.

Additional insights can be gained from two recent studies by Williams and colleagues (Williams et al., 2005; Williams, Goldstein, & Minshew, 2006). These studies are unique in that they examined verbal and spatial working memory in the same groups of high functioning individuals with autism and verbal-mental age matched comparison

groups, thus producing results across different dimensions of working memory *in the same individual*. In the first study (Williams et al., 2005), high functioning adolescents and young adults with autism had a spatial working memory deficit when task demands were complex (Williams et al., 2005). Specifically, the adults with autism performed as well as typically developing individuals except when the subtests involved social stimuli (e.g., memory for faces and memory for social scenes) or spatial working memory-stimuli that were also high in information-processing demands (Williams et al., 2005). In the most recent study, Williams et al. (2006) tested the same hypotheses in a group of children with high-functioning autism and a verbal mental-age matched comparison group. Consistent with data from the adults tested in the previous study, children were shown to have poor spatial working memory but intact verbal working memory (Williams et al., 2006). Interestingly, in a discriminant function analysis, spatial working memory was one of the factors that discriminated most accurately between the autism and control groups (Williams et al., 2006).

The combination of intact short-term memory with impaired working memory previously described in individuals with autism (Bennetto et al., 1996) provides an elegant illustration of the importance of preserving the distinction between short-term memory and working memory, a distinction that is frequently overlooked in discussions of memory. Of importance for autism, information addressing the development of distinct but interrelated systems (e.g., short term versus working memory, verbal versus spatial working memory) may provide insights into underlying neural relationships.

In summary, the results from studies examining verbal and non-verbal (spatial) working memory in individuals with autism are equivocal. There is positive and negative

evidence for verbal and non-verbal working memory deficits across age and level of functioning. This pattern of inconsistent findings on verbal and non-verbal working memory tasks is not altogether surprising given the variability in task demands used with individuals in different levels of functioning, age ranges, and comparison groups used in the studies. Variability in task demands is often related to how working memory is defined. That is, how 'work' is instantiated in a working memory task has critical implications for outcomes. Ensuring that the limits of the system are being taxed seems to be critical in at least some of the studies of autism (Minshew & Goldstein, 1998; Landa & Goldberg, 2005). Furthermore, task demands, as illustrated previously by Ozonoff and Strayer (2001), are often more complex than previously recognized, and they vary along dimensions that are not yet under explicit investigation (i.e., language, attention, or social engagement).

Of note, the investigation of working memory performance in autism may be unique among executive processes as something that can be empirically investigated from infancy through adulthood. The importance of working memory as an early emerging executive process has been established in studies of typical development. For example, Reznick and colleagues (Reznick, Morrow, Goldman, & Snyder, 2004) have identified the onset of working memory in typically-developing infants to occur at approximately five to six months. This is long before other executive processes that are impaired in autism come online. With increased emphasis on early identification through screening (Reznick, Baranek, Reavis, Watson, & Crais, 2007; Gillberg, Ehlers, Schaumann, & Jakobsson, 1990) and studies of 'at-risk' infant siblings of children with

autism, we have the opportunity to capture the developmental origins of perhaps the earliest emerging executive function.

1.3 Long-term memory in autism

In contrast to short-term memory, long-term memory has a limitless capacity and in some sense contains everything that a person knows. However for information to be stored in long-term memory, it must be encoded, and for it to be used, it must be retrieved. Over time, memories may decay or become unaccessible. In fact, performance on tasks that tap long-term memory can be adversely affected by problems with encoding, storage and retrieval.

The phenomenal long-term memory ability of certain individuals with autism has fascinated people since Kanner first described the disorder. Although savant memory skills are somewhat rare in autism, the majority of individuals with autism display a unique pattern of impaired and intact long-term memory functions that appears to be unrelated to overall intellectual ability. Specifically, episodic memory, including autobiographical memory, tends to be impaired in autism while procedural and semantic memory remains intact.

1.3.1 Long-term recognition and recall memory

Long-term recognition memory has been shown to be intact in a study in which a group of children with high functioning autism was compared with a verbal mental age matched comparison group for recognition of pictures (Molesworth, Bowler, & Hampton, 2005). Likewise, Salmond, Ashburner, Connelly, Friston, Gadian, and Vargha-Khadem (2005) found that children with autism demonstrated intact recognition memory when compared to mental age matched typically-developing children across a range of

increasingly complex recognition stimuli including word lists, word pairs, and stories. Results from both of these studies are consistent with numerous other reports of intact memory function on simple recognition memory tasks (Hala, Rassmussen, & Henderson, 2005; Bennetto et al., 1996; Griffith, Pennington, Wehner, & Rogers, 1999; Ozonoff & Strayer, 2001; Russell et al., 1996) and may represent one of the most stable findings in the literature on memory in autism.

Salmond et al. (2005) also investigated long-term recall memory and found that children with autism had relatively intact recall memory for material previously presented in the testing session when compared to mental age matched typically-developing children on tests of word list and word pair recall. This result is consistent with numerous other reports of intact memory function on simple recall memory tasks (Minshew, Goldstein, Muenz, & Payton, 1992; Minshew &Goldstein, 1993; Bennetto et al., 1996).

1.3.2 Semantic memory and 'savant' memory

Semantic memory, defined as the memory for facts or world knowledge, (Tulving, 1972) appears to be unimpaired in individuals with autism based on the majority of studies to date. Barth et al. (1995) found both visual and verbal memory impaired in high- and low-functioning children with autism. However, the most severe impairments were noted on verbal tasks that required semantic organization of information (e.g., remembering stories), which suggests that complexity of the semantic material might be a factor in memory performance of children with autism. Here again, it is worth noting that complexity must be related to an individual's knowledge base. For children with autism, their difficulties in the social domain will undoubtedly make tasks involving social stimuli or themes much more complex than they might be for their

typically-developing peers. In contrast, several other recent studies have reported intact recognition and recall for semantic material in individuals with autism. In a study by Minshew and Goldstein (1993), individuals with autism had intact recall and recognition memory. As part of their previously cited study, Salmond et al., (2005) found that children with autism had intact semantic memory, as measured on the Pyramids and Palm-trees Test and Wechsler test Information, Vocabulary and Similarities subtests (WISC-III; Wechsler, 1991) when compared to mental age matched typically-developing children. Ben-Shalom (2003) noted that memory for facts is generally a strength in individuals with high-functioning autism.

Further evidence of intact semantic memory comes from reports of individuals who have extremely strong memory skills for specific types of information. It is a relatively common clinical occurrence to encounter a child or adult who knows countless facts on a highly specific topic (e.g., all of the statistics for the New York Yankees since 1945, or mailing addresses for all of his classmates). However, little is known about the development of these types of memory skills (Heaton & Wallace, 2005). Although prodigious memory abilities are often present from extremely early in development, it is unclear what facilitates these abilities given relative weaknesses in many modes of acquiring and organizing information. It may be notable that these special memory abilities are often related to topics that are either highly organized at the outset or lend themselves to this type of organization, which is consistent with Baron-Cohen's empathizing-systemizing theory (Baron-Cohen & Belmonte, 2005).

1.3.3 Episodic and autobiographical memory

Episodic memory refers to the memory for events and episodes (Tulving, 1972). Autobiographical memory is a type of episodic memory in which the memory is personally relevant (i.e., the rememberer is part of the event). An early study of episodic memory in autism (Boucher, 1981) reported that high-ability children with autism remembered significantly less about recently experienced events than normal agematched and mentally retarded age- and ability-matched comparison groups. She proposed that, as a result, children with autism might encode less information from a complex stimulus such as social interaction or conversation. Millward, et al. (2000) extended this finding and discovered that children with high-functioning autism have a specific deficit in recall of personally experienced events. In fact, children with autism remembered more about an event that happened to another person than they did when the event happened to them without another person present. Therefore, the problem is not with recall per se, but rather, with how memories are formed when the child with autism is the agent of action (Millward, Powell, Messer, & Jordan, 2000). Similar results were found in a study with low-functioning adults with autism (Klein, Chan, & Loftus, 1999). Specifically, there was impaired access to personal experiences but there was intact knowledge of personal traits.

Bowler, Gardiner, and Grice (2000) reported more 'know' (related to semantic memory) and fewer 'remember' (related to episodic memory) responses during recognition tasks in individuals with Asperger's syndrome as compared to typically developing individuals. In additional support for this finding, Salmond et al. (2005) found that children with autism demonstrated significantly impaired performance on a test of episodic memory, the Rivermead Behavioral Memory Test, when compared to

mental age matched typically-developing children. The Rivermead Behavioral Memory Test includes subtests such as remembering a name, an appointment, a belonging, a route around the room; remembering to deliver a message; and orienting to time and place. These results are consistent with earlier evidence of impaired memory for recent events reported by Boucher (1981).

To summarize: individuals with autism tend to have impaired performance on episodic memory tasks but relatively preserved abilities on simple memory tasks or tasks that rely on 'rote' or semantic memory. Impaired episodic memory appears to be related to the complexity of the information, available encoding and recall strategies, and difficulties in temporal processing. As with studies of short-term memory, increased emphasis on early identification through screening (Reznick et al. 2007; Gillberg et al., 1990) and studies of 'at-risk' infant siblings of children with autism will provide the opportunity to explore the development of long-term memory. However, finding tasks appropriate for the range of long-term memory components in infants who cannot recount their experiences verbally presents a challenge. The earliest precursor might be found in studies of deferred imitation (Bauer, Werner, Dropik, & Wewerka, 2000; Carver & Bauer, 2001). Though not sufficient to cover all aspects of long-term memory, the deferred imitation paradigm would tap the emerging episodic system. Although imitation has been studied in autism, it is most often explored in relation to social skills or as a precursor for language development (Charman, Baron-Cohen, Swettenham, Baird, Drew, & Cox, 2003; McDuffie, Yoder, & Stone, 2005). However, researchers interested in the development of explicit memory in typically-developing children use deferred imitation procedures to assess memory for an event in children as early as in the first year of life

(Bauer, Wiebe, Carver, Waters, & Nelson., 2003). Tasks tapping this early emerging analogue to verbal reporting will be critical for constructing a developmental account of long-term memory and the underlying neural representation. This task is all the more critical given the emerging evidence suggesting that brain development in autism starts to go off-course before the second birthday (Hazlett, Poe, Gerig, Smith, Provenzale, Ross, et al., 2005; Courchesne, 2002), and thus before any behavioral measure of long-term memory has been applied in autism.

1.4 Assessment of memory in non-verbal children with autism

Because half of the children with autism are mute and the other 50% have language difficulties (as expected given the defining criteria of the disorder), it is imperative to develop tasks that can tap memory abilities in children who do not have strong verbal skills. This same obstacle faces researchers interested in infant and toddler development. For example, there is an active debate surrounding the onset of autobiographical memory in typically developing children that is largely focused on the question of whether children who cannot represent their experiences verbally can have autobiographical memories in the way that verbal children do (Nelson & Fivush, 2004; Howe & Courage, 2004). Furthermore, some accounts of infantile amnesia suggest that it is the lack of verbal encoding that makes it impossible to recall memories before around the age of two (Nelson & Fivush, 2004). Similarly, researchers investigating the ontogeny of working memory have not only had to develop measures of working memory in preverbal infants but also have been forced to defend the application of the construct to preverbal humans (Pelphrey & Reznick, 2003; Pelphrey, Reznick, Davis-Goldman, Sasson, Morrow, Donahoe, et al., 2004; Reznick et al., 2004; Reznick, 2007).

Some modifications will be necessary to make tasks used with typicallydeveloping infants and toddlers applicable for studies of non-verbal children with autism. Researchers must employ tasks and stimuli that engage a child who may not otherwise be interested in the social components of the task, components that would be highly motivating for a child who is developing typically. For example, playing 'peek-a-boo' with a child as part of a windows and curtains procedure, where the examiner makes eye contact with the child as part of a delayed response task, has been shown to activate prefrontal areas used in working memory tasks with typically-developing infants. The examiner provides an extremely salient stimulus for typically-developing children. However, children with autism may find the interaction to be less compelling or even aversive. Therefore, use of these social stimuli as part of a delayed-response paradigm may underestimate the child's ability, not because of memory deficits but because of a lack of interest in the stimuli.

Furthermore, the task characteristics will need to capitalize on the child's motor abilities and attention. Although the young child with autism may be processing information in a capacity similar to a younger toddler or infant, he will likely have age appropriate gross motor skills that will affect the manner in which he may demonstrate the capacity. For example, an infant may sit and watch repeated presentations of a delayed-response task, but a child with autism is unlikely to remain seated for long periods of time, even though this type of task is cognitively appropriate. Of course, it is important to note if a child has difficulty attending to social stimuli, but without excluding these social demands from the task, working memory cannot be separated from the social aspects of the task. These bidirectional relationships reflect the complex ways

that children develop, and they present even greater obstacles when the goal is to understand development in children with atypical development. For evidence, consider not only proposed difficulties in the delayed response task described above but also the conflicting evidence from computer versus human administered tasks. It is likely that even the most carefully designed and seemingly straightforward task will require the integration of multiple behaviors for successful task performance, and sorting out the contributions of each factor will undoubtedly be an enormous challenge.

Because we are becoming able to identify children with autism at younger and younger ages, and thus long before children have verbal fluency, we must construct appropriate tasks for assessing cognitive abilities in order to attain a full account of development. From this perspective, the biggest obstacle in autism research may be finding methods that are appropriate across the relevant age and abilities, and that can be used to describe development longitudinally. Without non-verbal tasks of cognitive development, we cannot construct an ontogeny of memory processes and determine the timing of the onset of memory differences in autism.

1.5 The current study

The current study examines working memory, long-term memory and language ability in children with autism and in typically-developing children matched to children with autism on verbal mental age and gender. These data will be useful in helping us understand the early emerging memory systems in young children with autism. Furthermore, utilizing measures of memory that do not require sophisticated language abilities will allow us to include a broader range of children with autism.

Working memory for this study was assessed using spatial and verbal working memory tasks. This strategy was selected to address the contradictory pattern of findings from older children with autism suggesting that spatial working memory is impaired, but verbal working memory may be intact (Williams et al., 2005). Two spatial working memory measures that utilize a delayed-response design, one verbal working memory measure, and one measure that could be performed using spatial and/or linguistic cues were administered.

Long-term memory was examined using a deferred imitation paradigm. This well-established paradigm, in which children observe a unique sequence of actions and are later asked to repeat the sequence, has been used extensively with typically developing children from infancy through early childhood (Bauer et al., 2000; Cheatham & Bauer, 2005). Additionally, it has been used with infants of diabetic mothers (DeBoer, Wewerka, Bauer, Georgieff, & Nelson, 2005), children who use American Sign Language (West & Bauer, 1999), children with developmental amnesia (Adlam, Vargha-Khadem, Mishkin, & de Haan, 2005), and infants with low birth weight (de Haan, Bauer, Georgieff, & Nelson, 2000). For this study, children were presented with four foursequence events: two arbitrary (no clear relationship between the actions in the sequence) and two enabling (actions that needed to be completed in order for the subsequent action to be completed) sequences. All sequences were designed to maximally engage participants with autism. Imitation is a weakness in children with autism (Charman et al., 2003, McDuffie et al., 2005; Dawson, 1984; Charman, 1994), however most examinations of imitation focus on immediate imitation of single events, gestures, or motor movements that may not be maximally engaging to the child with autism.

Furthermore, imitation has not been adequately examined as a cognitive precursor to memory in studies of autism.

A standardized test of language and cognition (Mullen Scales of Early Learning) and clinical assessments of autism (Autism Diagnostic Observation Schedule and Autism Diagnostic Interview-Revised) were performed to provide descriptive information about the participants.

1.6 Goals and hypotheses

This project addresses two primary goals:

Goal 1: To evaluate spatial and verbal working memory performance in young children with autism as compared to a group of verbal mental aged matched typically-developing children.

Questions and Hypotheses:

1A. Are spatial and verbal working memory impaired in young children with autism when language level is partialed out?

- Children with autism will show significantly lower performance on all working memory tasks when compared to verbal mental age matched children.
- Children with autism will show significantly lower performance on verbal working memory compared to spatial working memory tasks. Typically developing children will not show significant spatial and verbal working memory performance differences.

Goal 2: To describe long term memory performance in young children with

autism as compared to a group of verbal mental aged matched typically-developing children.

Questions and Hypotheses:

2A. Is long-term memory as measured on deferred imitation tasks impaired in young children with autism?

 Children with autism will show significantly lower performance on deferred imitation tasks involving arbitrary sequences but not on enabling sequences when compared to verbal mental age matched typically developing children.

II. METHODS

2.1 Participants

2.1.1 Children with autism

Thirty-one children (24 males/ 7 females) with a known diagnosis of autism between the chronological ages of 23 and 52 months (2.0 to 4.5 years) were screened for possible inclusion in this study. Ten children (9 males/ 1 female) from this group were excluded from participation: 5 did not meet diagnostic cutoff for autism on the ADI-R or ADOS and 5 could not complete the testing battery due to noncompliance or falling below a verbal mental age of 18 months. Twenty-one children (15 males/ 6 females) with a known diagnosis of autism between the chronological ages of 32 and 52 months (2.5 to 4.5 years) were included in this study.

Participants were recruited from parent support groups throughout North Carolina and local service providers. Informational flyers were distributed to parents of potential participants. Interested parents contacted us through phone or email. Following a brief telephone conversation describing the study and assessing the tentative diagnosis of autism (i.e., a clinical diagnosis by a clinician/physician), participants were invited to join the study. Potential participants with autism were screened for additional exclusionary diagnoses (i.e., Fragile X Syndrome, Down Syndrome, neurofibromatosis, tuberous sclerosis, cerebral palsy, severe head trauma, or a known genetic syndrome), but no potential participant had any condition that would preclude participation. The diagnosis of autism was verified with the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000), the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter,& LeCouteur, 1994), and according to DSM-IV diagnostic criterion (APA, 1994). Specifically, children with autism had to meet the diagnostic algorithm cut-offs on the ADI-R and ADOS.

2.1.2 Children with typical development

Twenty-one children with no known diagnosis of autism, or family history of first-degree relatives with a diagnosis of autism, were matched to participants with autism based on verbal mental age and gender. Typically-developing children's chronological age ranged from 23-36 months. In addition to the twenty-one children included in the study, fifteen children (12 males/ 3 females) were screened but excluded: 9 children had scores greater than 1.5 SD on the Mullen Expressive Language subscale and 6 children could not be matched to any of the participants with autism.

Participants were recruited through a registry of infants/children maintained by Dr. Steven Reznick. The registry was established by mailing an initial recruitment letter to parents identified from birth records obtained from the North Carolina Department of Vital Statistics. This letter describes a wide range of research activities and invites parents interested in participating in developmental research to return a prepaid postcard with information about their infant and other children in the household. Parents of children who were estimated to be appropriate verbal mental aged matched comparisons for children with autism were contacted by telephone to determine if they might be interested in the current study. All children who participated were from full-term pregnancies and did not have any major health problems.

Typically-developing children were matched to children with autism based on the Mullen Scales of Early Learning Expressive Language subscale. Typically-developing children with scores ± 1.5 SD were not considered for matches.

The ADOS was administered to the typically-developing children following standard administration protocol. Children who had scores in any diagnostic algorithm domain falling in the autism spectrum or autism range would not be included in the study. No children were excluded due to elevated ADOS scores.

2.2 Design and procedure

Multiple tasks were used to examine working memory and long-term memory. Parents were invited to bring their child to our laboratory at UNC Family and Community Research Center in Chapel Hill, North Carolina or, if they preferred, testing could take place in their home. Eighty of participants with autism and 65% of children with typical development were seen in their homes. We encouraged parents to select the environment that they felt would be best for their child. The UNC research space is child friendly, however, some children with autism and typically-developing children, are uncomfortable in unfamiliar settings. We wanted the child to be an environment that would provide him with the opportunity for giving his best performance. The tasks were developed to be portable and compatible for use in participant homes. If home testing was selected by the parents, a quiet room with minimal distractions (i.e., a room without toys) was selected. In both settings, a booster seat was placed in a chair for the child's use during the elicited imitation procedures; all other testing was completed on the floor or on a child sized work surface. The testing occurred in three one-hour sessions on three different days. The Mullen Scales of Early Learning and the Autism Diagnostic

Observation Schedule (ADOS) were administered in the first visit. The second one-hour visit was scheduled to occur within 3 weeks of the first visit. During the second visit children were presented with two of the short/term working memory tasks (Egg Hunt and Animal Houses) and two of the deferred imitation sequences (Ramp and Train). The third one-hour visit was scheduled to occur 1-7 days after the second one-hour visit. During the second visit, children were presented with the remaining tasks: two short-term working memory tasks (Cotton Wells and Nonsense Words) and two of the deferred imitation sequences (Funnel and Boat). If the child could not complete the tasks scheduled for any visit due to fussiness or fatigue, those tasks were completed in the next visit. A fourth visit was required for 15 children (10 with autism and 6 with typicaldevelopment). All visits were scheduled to accommodate the participant's schedule, and the child's parent was present for all testing, usually sitting in a chair in the room away from the activities but available to the child, if needed. At the beginning of the first testing session, the parent read and signed the informed consent document. The parent was encouraged to ask questions about the study. Parents were informed that they could discontinue their child's participation at any time, without any penalty. No parent took advantage of this opportunity.

During testing, children were given parentally approved food snacks, praise, and small toys as a reward for successful completion of the various activities described below. The children were presented with as many of the planned games as could be enjoyably completed within the given testing time. The task structure did not require the child to remain focused on any single activity for long periods.

Each child's actions and responses were recorded by video camera. Videotapes were used for scoring responses and verifying live codes. The experimenter coded the child's responses on the short term/working memory activities as the trials progressed; the videotape of the session was referenced to resolve any discrepancies and for scoring the elicited imitation activities.

2.3 Measures

2.3.1 Autism diagnostic measures

Autism Diagnostic Observation Schedule (ADOS; Lord, Risi, Lambrecht, Cook, Leventhal, DiLavore, et al. (2000). The ADOS is a semi-structured direct assessment designed to evaluate individuals suspected of having autism. It is appropriate for use with individuals from a broad age spectrum (toddlers to adults) and with a range of language ability (no speech through verbal fluency). The ADOS consists of various activities and associated toy materials that allow the assessor to create situations to observe social and communication behaviors in an enjoyable and fun environment. These activities provide interesting, standard contexts in which interaction can occur. Administration of the ADOS requires that the assessor follow a hierarchy of behavioral presses aimed at allowing the child to make and follow social and communication overtures.

The ADOS consists of four modules each requiring 35-40 minutes to administer. The individual being evaluated is given just one module, depending on his/her expressive language level, which can be determined in a brief interaction prior to starting the ADOS, and chronological age.

Administration of the ADOS occurred during the first visit. Due to the age and language levels of the children only two of the modules were used: Module 1 for children with no speech through simple phrases and Module 2 for children with simple phrases through verbal fluency. Following administration, behavioral codes were recorded and the diagnostic algorithm scores computed. All administration and scoring of the ADOS was completed by an individual who had completed reliability training with the instrument developer (80% reliability on the complete protocol and algorithm from three administrations). Parents of children with autism who meet algorithm cutoffs for autism were administered the Autism Diagnostic Interview-Revised (ADI-R). Four children with a clinical diagnosis of autism did not meet ADOS algorithm cut-offs for autism, so they were excluded from the study. No typically-developing children were excluded due to meeting autism spectrum or autism cut-offs on the ADOS.

Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, &, LeCouteur, 1994). The ADI-R is a semi-structured interview in which an experienced interviewer questions a parent or caregiver who is familiar with the developmental history and current behavior of the individual being evaluated for the presence of autism. The interviewer follows up scripted questions with interviewer-generated questions aimed at obtaining a detailed description of the behavior of the child. The descriptions are then used by the interviewer to make behavioral ratings. Administration and scoring required from 1 1/2 to 2 1/2 hours. Results were scored and interpreted using a diagnostic algorithm that provides minimum score cutoffs needed to support an autism diagnosis for communication, social and ritualistic/repetitive behaviors.

The ADI-R was performed with a parent of participants who met or exceeded the diagnostic algorithm cutoffs for autism on the ADOS. Because of the length of time required to complete the ADI-R, the parent was interviewed in a session separate from the child visits. Parents were given the option of completing the interview in our laboratory or in their home. Eighty percent selected to have the interview done in their home. Following administration, behavioral codes were recorded and the diagnostic algorithm scores (i.e., communication, social and ritualistic/repetitive behavior) were computed. All administration and scoring of the ADI-R was completed by an individual who had completed reliability training with the instrument developer and who demonstrated 90% reliability on the complete protocol and algorithm from three administrations.

2.3.2 Standardized measure of development and language

Mullen Scales of Early Learning (Mullen Scales). The Mullen Scales consist of five scales aimed at providing a broad assessment of early cognitive and motor development by tapping fundamental abilities such as attending to a picture, matching objects, the ability to attend to sounds and/or simple questions, and the ability to produce sounds and/or words. The five scales measure gross motor, fine motor, visual reception, expressive language and receptive language skills in children from infancy through 5 years and 8 months of age. Due to time constraints, children were not administered the gross or fine motor scales. For each subscale, a T-score, percentile rank, and age equivalence was generated. Each child was given the Mullen Scales in the first visit following standard testing procedures for the instrument.

2.3.3 Working memory measures

Sample score sheets for all working memory games can be found in Appendix A.

Cotton Wells Game. This task is a modification of a task that Newcombe and colleagues used to examine children's ability to represent the spatial location of a hidden object (Newcombe, Huttenlocher, Drummery, & Wiley, 1998; Huttenlocher, Newcombe, & Sandberg., 1994). We altered the task to increase the demand placed on working memory.

A rectangular plastic box (18" x 30" x 6") filled with cotton balls was placed in front of the child. The box was covered with a black, felt board that has either three or five 5" diameter holes in it. The cover prevents the child from accessing multiple locations simultaneously. The child was positioned equal distance from either end of the box. An item that interests the child was selected from a collection of toys. That item was buried in the cotton balls by the examiner through one of the holes as the child watched. The child was then encouraged to find the object by digging in the cotton with his/her hand after a brief delay. During the delay the child was distracted from the apparatus to keep him from focusing his attention on the location where the toy was hidden.

This task was developed through a prior study of 27 month-olds to have increasing levels of difficulty based on the number of locations (3 and 5) and length of delay to search (7, 14 and 21s). The experimenter first gave the participant teaching trials to make sure that he understood the game. The teaching trials were repeated as necessary to teach the rules of the game, which is to find the hidden object. Most children did not require more than two presentations to demonstrate an understanding of the task. The teaching trials were not used in scoring. In the rare event that the child did not complete

the teaching trials, then the game was discontinued. Following the teaching trials, a starting level was selected. It was up to the experimenter's judgment to select the most appropriate level based on the specific child; for typically developing children age-related starting levels were suggested. For example, a 3 year-old child with autism would not generally start the task at the same level of difficulty as a child with typical development. The structure for this task is similar to many tests of cognitive development and allows performance to be assessed with less child fatigue and performance frustration. Each level of the task has three trials. If the child gets at least two trials correct in a level then he advances to the next level. When the child reaches a level where he is no longer getting at least two trials correct, the game is ended. If he gets at least one trial correct in any level, he is given partial credit for that level (reflected as .5 on his score). If the level selected for a child is too difficult (i.e., the child gets fewer than 2 correct) then the experimenter goes back two levels or to the easiest level, which ever is appropriate. By going back two levels, the experimenter has a greater chance of selecting a level in which the child will be proficient and will reduce the likelihood of the child disengaging from the game because of frustration. This procedure can be repeated as needed (i.e., if the child fails the newly selected level).

The hiding location of the toy is predetermined for each trial. Each participant's reach responses were coded for all administered trials and a maximum level of performance was determined (ranging from 0.0 to 6.0).

Animal Houses. A tray covered with green felt, on which small wooden houses with hinged roofs can be placed, was positioned in front of the child. The child was presented with two wooden houses that wooden animal tokens fit in. Each house was

introduced to the child, "This is where the _____ lives," as the child watched the examiner place each of the animal tokens in the appropriate houses; the matching tokens were reserved by the examiner. The houses were then closed (i.e., the roof was placed on top of the 'house' so that the child cannot see the animal inside). Following a brief delay, the child was presented with one of the tokens and asked to identify the house that the particular animal lived in: "Where does the _____ live?" The child could respond by pointing to the house or lifting the roof from one of the houses. After the child responded, the houses were rearranged and the procedure was repeated.

As in the other spatial working memory tasks, the task was structured to have levels of difficulty based on number of locations (2, 3, or 4) and length of delay (3 or 6s). The order of the animal houses, as well as, the particular animal that the child was asked to find was predetermined for each trial. Before beginning the teaching trials, the experimenter determined that the child was familiar with the names of the animals used in the game by asking the child to point to the various animals: "Show me the _____." If the child was unfamiliar with one of the animals, it was replaced with an animal known by the child. Eight animals were available for selection: cat, turtle, pig, whale, alligator, elephant, bird, and dog. If the child could not indicate knowledge of common animal names, the game was discontinued. Following the introduction of the animals, the experimenter gave the participant teaching trials to make sure that the child understood the game. The teaching trials could be repeated as necessary to teach the rule of the game, which is to find the specific animal that the examiner requests. After investigating the apparatus, most children do not require more than two presentations to demonstrate an understanding of the task. The teaching trials were not used in scoring. In the rare

event that the child did not complete the teaching trials, then the game was discontinued. Following the teaching trials, a starting level was selected based on the experimenter's judgment. The experimenter was conservative in selecting a starting level as children can easily lose interest if the game demands exceed their skill level too rapidly. Each level of difficulty had three trials, and to advance to the next level the child must get two trials correct. Children who were correct on at least one trial in any level were given partial credit for that level (reflected as a score of .5). Scores on the game ranged from 0.0 to 6.0.

Egg Hunt Game. The child was asked to sit on an appliqué that was placed on the floor and that pictured a flower, racecar or train. The examiner then placed a row of opaque plastic containers approximately 24 inches in front of the child such that the containers were out of easy arm's reach but the entire array was visible. The containers were placed approximately 8 inches apart to discourage a child from selecting two containers simultaneously. A transparent egg containing a highly motivating treat or an interesting toy was placed under one of the containers as the child watched. The child was then encouraged to find the object by lifting the container following a brief delay. During the delay, the child was distracted from the array to preclude the possibility of attending to the location where the food/toy was hidden.

The administration of this task was identical to that described previously for the Cotton Wells Game. Briefly, the task has increasing levels of difficulty based on the number of locations (3 and 5) and length of delay to search (7, 14 and 21s). The experimenter first gave the participant teaching trials to insure understanding of the game. Following the teaching trials, a starting level was selected based on the

experimenter's judgment for each child. Each level of the task had three trials. Children who got at least two trials correct in a level then advanced to the next level. When the child reached a level where errors predominated, the game ended. Children who had one correct trial in any level were given partial credit for that level, reflected in a scored of .5. If the level selected for a child was too hard (i.e., the child had fewer than 2 correct trials) then the experimenter went back two levels or to the easiest level, whichever was appropriate. By going back two levels, the experimenter had a greater chance of selecting a level in which the child was proficient and reduced the likelihood of the child losing interest in the game because of frustration. This procedure could be repeated as needed (i.e., if the child failed the newly selected level).

The hiding location of the food/toy was predetermined for each trial. Each participant's reach responses were coded for all administered trials and a maximum level of performance was determined ranging from 0.0 to 6.0.

Nonsense Word Repetition. Each child was asked to repeat a series of nonsense words (i.e., a word with no known meaning but that follows English linguistic rules). Strings of nonsense words contained 1 to 6 words (e.g., tur ug; sah poe voor; tonk voo pur ug). Each word or word string was presented two times, and if the child did not respond then the next item was presented. The words were presented with the aid of puppets or child-safe objects to enhance the child's attention and interest in the game. The child was asked to repeat several common words before the nonsense words began to establish general understanding of the task.

There are five levels in the game: level one consisted of four single word trials; level two contained four two-word trials; level three contained four three-word trials;

level four contained four four-word trials; and level five contains four five-word trials. There was no delay between presentation of words and repetition. If the child missed two or more word strings in a level, the game was concluded. Each child started at the same level of the game and was presented trials until errors occurred in more than two trials in any level. Responses were recorded on a data sheet. Each child was given an overall score based on the complexity of the word strings repeated (i.e., one- to five-word strings), on a scale of 0.00 to 5.00 with scores increasing by 0.25 increments.

2.3.4 Long-term memory

Deferred Imitation. This task began with introductory phase in which the child was given the opportunity to observe and imitate a simple sequence such as putting a ball in a cup and pressing a leaver to make it pop up. When the child demonstrated understanding of the task, the four-step arbitrary and enabling sequences were presented. The sequences were selected to contain events that would be highly motivating for children with autism. Appendix B lists detailed descriptions of each event sequence. For each of the four events, the child was presented with a tray of objects to examine as long as the child remained interested or until 5 minutes had passed. This initial exploration served as a baseline measure for object exploration. The examiner then replaced the items on the tray. Following a brief introduction, the examiner used the items to present a sequence of arbitrary or enabling actions to the child. For this study, minimal verbal descriptions of actions were given to reduce any advantage that these verbal cues may give the typically-developing children. Instead, the actions were accompanied by statements such as: "Watch this."; "This goes here."; "Look now." Each sequence was presented twice. The tray was removed for approximately 10 minutes. During that time,

the child was quietly engaged with a toy or other object of interest. Following the delay, the tray was re-introduced to the child. The child was encouraged to use the items again: "Remember what we did?" The child was then given encouragement such as "good job" or "what else can you do" during the imitation period regardless of whether the child was performing the target actions. The examiner did not label specific activities or actions. The task was concluded when the child completed the sequence or was no longer interested in the material. Videotapes were used to record the sequences. The videotapes were reviewed to code each of the four actions or three action pairs in each sequence at both baseline and imitation.

III. RESULTS

3.1 Data verification and reduction

3.1.1 Working memory measures

Verification. Measures of working memory were coding live during the assessment sessions. Following the visits, 20% of the testing sessions for each group were reviewed and rescored for reliability by an undergraduate research assistant who did not participate in the initial visit. The rescored results were consistent with the original scoring in all cases (ranging from 92-100% agreement).

3.1.2 Deferred imitation.

Verification. Video tapes were reviewed to assess two types of behavior in these tasks: actions and action pairs. First, the number of individual actions was recorded for the baseline and recall phases. In each of the four four-element sequences, four actions could be recorded. Second, the number of action pairs was recorded for the baseline and recall phases. In each of the four four-element sequences, three action pairs could be recorded. Only the first occurrence of an action was considered for an action pair. This helps to reduce the likelihood that the action pairs score will become inflated due to chance or trial and error instead of memory. For example, if a participant produced a string of actions 4, 1, 2, 3, 4; they would be given credit for 4 actions but only two action pairs (i.e., 1-2 and 2-3). Actions and action pairs for each sequence are presented in Appendix C. There were two reviewers for this task: a primary rater (the author) and a secondary rater (an undergraduate research assistant). Both reviewers were trained on a

existing set of deferred imitation demonstrations and established reliability at greater than 90% on a set of six sequences that were similar to those used in this study. The primary reviewer scored 100% of the assessments and the secondary reviewer scored 20% of the assessments. There was acceptable agreement between the reviewers: actions (ranging from 80 to 98%, averaging 91%) and action pairs (ranging 83-95%, averaging 92.5%).

Reduction. Summary scores were calculated for actions and action pairs. First, the actions from each of the four sequences were summed independently for the baseline and recall phases. The sum for baseline and recall actions have possible ranges of 0-16. This sum was used in all analyses. Second, the action pairs from each of the four sequences were summed independently for the baseline and recall phases. The sum for each phase had a possible range of 0-12.

3.2 Data analysis

3.2.1 Sample characteristics and matching

Twenty-one children with autism and 21 children with typical development received complete evaluations and were included in analyses. Fifteen children in each group were male and six were female (2.5:1). There were not enough females to conduct independent analyses on gender. Nineteen of the 21 participants were Caucasian in both groups. Independent sample *t*-tests were performed on chronological age, Mullen Receptive Language age equivalent, Mullen Expressive Language age equivalent, and Mullen Visual Reception age equivalent to examine group differences. Chronological age differed significantly for the two groups, t (40) = -8.92, p < .001, as would be expected given the matching strategy. Children with autism are less likely to have receptive and expressive language scores that are equivalent to their typical age matched

peers. Because matching was based on verbal age equivalents, the children with autism would be expected to be older. The independent samples *t*-test for the matching variable, Mullen Receptive Language age equivalent, was not significant, t(40) = .064, p = .94, indicating that the participants were well matched. Results from the Mullen Expressive Language and Visual Reception age equivalents showed significant group differences, t(40) = 3.87, p < .001 and t(40) = 3.70, p < .001, respectively. Significant difference in the Expressive Language age equivalents was not unexpected; expressive language is often more significantly delayed in children with autism than is receptive language. Results are summarized in Table 1.

The ADOS scores for the groups are summarized in Table 2. Two modules were used for this study: Module 1 (for children without speech or only producing simple phrases) and Module 2 (for children with simple phrases or fluent speech). As anticipated, the significant differences in the Mullen Expressive Language age equivalents were reflected in the number of children receiving the two language based modules: Module 1 was administered to 19 children with autism and 16 children with typical development. Module 2 was administered to 2 children with autism and 5 children with typical development. Independent sample *t*-tests were computed for each diagnostic sub-domain, but they should be interpreted with caution because the ADOS is not designed to capture the range of typical behavior that might be seen in each of these domains. Significant floor effects in the typically developing group were present. However, it should be noted that the two-tailed independent t-test is robust despite a non-normal distribution. As expected, the groups differed significantly on ADOS scores

(Communication: t (40) = 17.76, p < .001; Social: t (40) = 17.07, p < .001; and Communication/Social Sum: t (40) = 22.51, p < .001).

Descriptive statistics for ADI-R sub-domain scores for the autism group only are reported in Table 3. The scores for the communication domain ranged from 7 to 11 (M = 8.52, SD = 1.08); social domain ranged from 12 to 26 (M = 19.1, SD = 3.6); and ritualistic/repetitive behavior domain ranged from 3 to 8 (M = 4.3, SD = 1.2). Parents of children with typical development were not administered the ADI-R.

3.2.2 Is working memory impaired in young children with autism?

Data on working memory measures was examined for normality, outliers and unequal variances. No significant outliers were observed on any measure, and the data were normally distributed in both groups on the Cotton Wells, Egg Hunt and Nonsense Words tasks as indicated by the Kolmogorov-Smirnov test. The Animal House task was not normally distributed in the autism group. Levene's Test for Equality of Variances indicated that the group variances were significantly different. Because data from Animal Houses violated two assumptions for parametric tests, a non-parametric test was used to examine these data. Parametric tests were to examine the remaining data.

Spatial working memory. Independent-sample *t*-tests were conducted on each of the four working memory measures to evaluate the hypothesis that children with autism would perform significantly worse than children with typical development. The Animal House task was also examined using non-parametric tests due to violations of normality and equal variances. Non-parametric tests for this task will provide the most conservative and accurate comparison. Results for all measures are summarized in Table 4. The test for the Egg Hunt was significant, t(40) = 5.53, p < .001. Levene's Test for

the Equality of Variances was significant for the Cotton Wells, F(1,41) = 8.76, p = .005; the *t* statistic result based on the unequal variances is significant, t(40) = 5.63, p < .001. The autism group had significantly lower performance on both the Cotton Wells and Egg Hunt.

Verbal working memory. Only 16 children with autism were able to complete the Animal Houses. Levene's Test for the Equality of Variances was significant for the Animal Houses, F(1,41) = 7.77, p = .005; the *t* statistic result based on the unequal variances is significant, t(31.4) = 9.79, p < .001. Again, the group of children with autism had significantly lower performance than the group with typical development. The Mann-Whitney test was also computed for the Animal House task. The non-parametric test transforms the data into ranks and finds group differences in the constructed ranking variable. The results, consistent with the *t*-test, indicate a significant group difference, U = 2.0, p < .001. The Nonsense Words task had the lowest rate of completion; 11 children with autism and 19 children with typical development. Despite the small sample size, the independent samples *t*-test on the Nonsense Words task was significant, t(28) = 7.93, p < .001, suggesting a notably robust effect. As with the previous tasks, the autism group performed at a level lower than the children with typical development.

Relationships among working memory measures. Given the magnitude of group differences, Pearson correlations among the four working memory measures with the groups combined will be artificially inflated. An alternative strategy for tapping meaningful relations among working memory measures is to calculate correlations separately for the autism and typical groups. This strategy causes a notable reduction in

power, but supports a more straightforward interpretation. Table 5a contains the correlation matrix for the autistic group and Table 5b has the comparable correlations for the typical group. It should be noted that the Pearson correlation makes the same distributional assumptions as the parametric tests. Relationships between the Animal Houses and other measures should be interpreted with caution. However, the Pearson correlation is not recommended for use with data that contains fewer than five levels on a given variable. Therefore, the Spearman correlation was computed for the autism group. (Note: Spearman correlations on the data from the group with typical development reflected the same pattern of results as those reported here.) The relationships between the hypothesized spatial and verbal working memory tasks were of specific interest.

In the group with typical development, performance on the Cotton Wells was significantly related to each of the other measures: Egg Hunt, r = .78, p < .000; Animal Houses, r = .43, p < .05; and Nonsense Words, r = .66, p < .002. Performance on the Egg Hunt was significantly correlated with the Animal Houses, r = .46, p < .04 and Nonsense Words, r = .65, p < .002. Notably, Nonsense Word performance was not significantly correlated with Animal Houses, r = .24, p = .32, possibly indicating that verbal working memory is not being measured on Animal Houses. Additionally, the significant relationships between the Animal Houses and the two spatial working memory tasks suggest that children may not be using verbal working memory for performing this task. All results with the Animal Houses should be viewed with caution due to the violations of parametric test rules in the data. In the group with autism, performance on the Cotton Wells was significantly related to the Egg Hunt, r = .48, p =

.026 and marginally related to Nonsense Words, r = .59, p = .055. All other relationships were not statistically significant.

3.2.3 Is long-term memory as measured using deferred imitation tasks impaired in young children with autism?

The deferred imitation data were examined for normality, outliers and unequal variances. Summary statistics are presented in Table 6. Significant outliers were not observed in any category: Baseline Actions, Baseline Action Pairs, Recall Actions or Recall Action Pairs; enabling or arbitrary. There was a notable floor effect in both groups during the baseline condition as well as for the autism group in the recall conditions. That is, the majority of children produced very few target actions. Based on this fact, it is not surprising that the data failed the Kolmogorov-Smirnov test of normality. Levene's Test for Equality of Variances was only significant for the Baseline Arbitrary Pairs condition, F = 9.85, p < .003. That is, the variances for the groups were not generally significantly different. As discussed earlier, the general linear model is robust to nonnormal distributions. Therefore, the planned 2 (group) X 2 (condition) X 2 (sequence) mixed-design analysis of variance controlling for Mullen Expressive Language age equivalent was conducted for the actions and action pairs. The between-subjects factor was group (autism or typically-developing). The two within-subjects factors were condition (baseline or recall) and sequence (enabling or arbitrary). The Huynh-Feldt correction was applied to the data due to sphericity using the procedure recommended in Girden (1992). The corrected degrees of freedom are reported.

Actions. The analysis revealed a significant main effect of group, F(1, 39) = 45.11, p < .0001. The children with autism had significantly lower performance scores

than the children with typical-development. There was not a significant main effect of condition, F(1, 39) = .144, p = .706 or sequence, F(1, 39) = .008, p < .923. However, there was a significant 2-way interaction between group and condition, F(1,39) = 44.24, p < .0001.

Follow-up analyses revealed that the groups did not differ significantly at baseline performance, but that the children with typical development performed significantly better than the children with autism at recall, t (39) = 10.42, p < .0001. Further follow-up analyses revealed that both groups produced significantly more actions at recall than at baseline: autism; t (20) = 6.65, p < .0001, and typically-developing; t (20) = 21.1, p < .0001, indicating memory for action pairs.

An analysis of action pairs revealed a significant main effect of group, F(1,39) = 12.27, p = .001. The children with autism had significantly lower performance than the children with typical development. There was not a significant main effect of condition, F(1,39) = .016, p = .89 or sequence, F(1,39) = .139, p = .711. However, there was a significant 2-way interaction between group and condition, F(1,39) = 9.25, p = .004.

Follow-up analyses revealed that the groups did not differ significantly at baseline performance, but that the children with typical development performed significantly better than the children with autism at recall, t (39) = 10.42, p < .0001. Further follow-up analyses revealed that both groups produced significantly more actions at recall than at baseline: autism; t (20) = 3.99, p = .001, and typically developing; t (20) = 8.20, p < .0001, indicating memory for action pairs.

IV. DISCUSSION

The goal of this study was to examine working memory and long-term memory differences in a well-characterized group of children with autism, as early in development as possible. To accomplish this goal, a unique battery of working memory tasks and deferred imitation tasks were employed. Children with autism were matched to a group of typically-developing children based on verbal mental age and gender. The characterization and matching procedure were important components of this study because the pattern of results from previous studies of memory in autism contain a broad range of participant ages, diagnostic categorizations, functioning levels and comparison samples, resulting in a conflicting account of memory in autism. In fact, when unexpected results in previously reported studies were found, be they positive or negative, the issue of appropriateness of the comparison groups was often raised. Indeed, finding an appropriate comparison group is challenging. For example, if a 15 year old child with autism who has a verbal mental age of 7 is matched with a typically developing child of age 7 and verbal mental age 7, it is difficult to know what effect 8 additional years of life experience exerts on the development of memory and related neural structures in the child with autism. Matching based on mental age and chronological age across two syndromes (i.e., autism, Down syndrome, learning disabled) carries the assumption that the reason for the retardation in the two cases is comparable (Johnson, Halit, Grice, & Karmiloff-Smith, 2002), but this is often not the case. Unfortunately, there are no well-established rules that researchers can use to select

comparison groups (i.e., learning disability, typically-developing, Down Syndrome) or matching strategies (i.e., chronological age, verbal mental age, non-verbal mental age, IQ). The lack of widely accepted matching rules likely contributes to the pattern of inconsistent results and almost certainly impedes progress in the field (Dawson, 1996).

Overall, sample characterization and matching procedures were successful. The group with autism met all diagnostic standards for the study; the comparison sample was matched closely on verbal mental age and gender. Unfortunately, the strict characterization of the group of children with autism (i.e., meeting autism criteria on the ADI-R, ADOS, and DSM-IV) resulted in the loss of a large number of potential participants. This loss of participants could affect generalizability of the results on both the lower and upper levels of functioning in autism. The exclusion of children who are non-compliant with testing procedures is a limitation for any study employing measures where participants must actively participate. It is impossible to predict how these children might have performed, but it is reasonable to think that their difficulties with the testing procedures, in-spite of the efforts of an experienced examiner, may reflect a greater degree of cognitive impairment. On the other end of the spectrum, the children who were excluded due to diagnostic test scores that did not meet required cut-offs may provide an interesting comparison group. Is there a specific memory profile that can be seen across the entire spectrum of children with autism? If there is not, the pattern of intact and impaired abilities could be used to develop meaningful sub-groups; which could aid in our understanding and treatment of this complex disorder.

Given the expected ratio of males to females in autism (4:1), this study had a relatively large proportion of females (the ratio here was 2.5:1). The group of females

was not large enough to sustain independent examination. No special effort was given to recruiting a large sample of females, so it is unclear whether this discrepancy reflects an artifact of recruitment or ashift in the rate of autism in females. Females with autism have historically had higher rates of mental retardation. However, the females in this group did not have any systematic features to distinguish them from the males. Because little is known about females with autism, future studies should focus recruitment efforts on this group.

4.1 Working memory

Working memory may be unique among executive processes because it is a cognitive process that can be empirically investigated from infancy through adulthood. The importance of working memory as an early emerging executive process has been established in studies of typical development. The onset of working memory in typicallydeveloping infants occurs at approximately five to six months (Reznick et al., 2004), long before the emergence of other executive processes that have been found to be impaired in autism. Notably, working memory is necessary for completion of many executive function tasks that have been used to identify executive function deficits in autism. For example, in flexibility tasks such as the Wisconsin Card Sorting Test (WCST) the cognitive set (e.g., the current sorting rule) is held in working memory to guide behavior. The contents of working memory must then be updated as the sorting rule changes. Similarly, inhibitory tasks require maintenance of the inhibitory rule in an activated state in working memory to guide behavior. Because the components of executive function are interrelated and interdependent, with working memory playing a central role (Pennington & Ozonoff, 1996), the examination of working memory in autism early in

development may provide a useful basis for identifying children who may develop problems with other executive processes.

One may ask if the 'work' of working memory was fully evoked by the tasks used in this study. Task complexity is a critical component of studies that have shown significant differences in working memory performance (Williams et al., 2005; Landa & Goldberg, 2005). However, there are many ways to increase complexity (e.g., amount, type of information, type of task, length of time). The manner in which these dimensions are varied may have important effects on outcomes and may contribute to the pattern of inconsistent findings that characterize research on working memory in autism. For this study, the inclusion of multiple locations, delay, and repeated trials was based on previous studies in which these dimensions have been consistently shown to be effective (Reznick et al., 2004; Pelphrey et al., 2004). Perhaps the best indicator that the tasks used here were sufficiently complex is the robust group differences found across the four working memory tasks. Although Landa and Goldberg (2005) found that the memory load in a working memory task needs to be high (e.g., 6 to 8 locations) in order to evoke group differences in adolescents with high functioning autism, the present study suggests that tasks using 3 to 5 search locations is quite adequate to detect group differences in a younger and lower-functioning group of children.

The significant results from the two spatial working memory tasks are consistent with numerous previous studies. Dawson et al. (1998) administered delayed non-match to sample and delayed response tasks to young children with autism and children with Down syndrome. In that study children with autism performed significantly worse, indicating not only working memory deficits but also that performance is not solely

related to global cognitive impairment. In another study, a computer based version of a delayed response task used eye movements to examine performance (Minshew et al., 1999). This study too found significant differences in a group of high-functioning adults. Interestingly, the same procedure has been used to define the circuitry of the prefrontal cortex. The previously mentioned study by Landa and Goldberg (2005) found that adolescents with high-functioning autism were impaired on a spatial working memory task when the working memory load was high but not low, indicating that working memory is not completely eliminated in autism. Finally, two studies by Williams et al. (2005 & 2006) also showed spatial working memory deficits in adults and children with autism. These studies suggest that working memory deficits are present across the autism spectrum and throughout development.

The results from the two verbal working memory used here are consistent with previous work by Bennetto et al. (1996) and Williams et al. (2005) that revealed verbal working memory differences in adolescents with high-functioning autism. Although the group of children seen in this study would not be considered high-functioning, the children who completed the verbal working memory measures may be on a different developmental trajectory than the other children in the study, representing a higher functioning group relative to the rest of the sample.

One might contend that the Nonsense Word task is not a working memory task but a short term memory task because it does not require the manipulation or maintenance of the words. However, we propose that in young children working memory will be activated by the maintenance of increasingly long strings of 'words'. Interestingly, and consistent with our hypothesis that this is a working memory task; the

significant results for the task in this study are inconsistent with studies that have examined short-term/echoic memory in autism. In those studies, children with autism did not show significant deficits in short term/echoic memory (Boucher, 1978; Rumsey & Hamburger, 1998; Russell et al, 1996). Therefore, the pattern of results here suggests that the Nonsense Words task is activating working memory.

Previous versions of the tasks used in this study have employed a procedure in which participants all received the same number of trials. Composite scores were computed by calculating the proportion of correct responses. This procedure is problematic for several reasons. First, children sometimes get bored or frustrated prior to the completion of all of the trials. It is impossible to determine if the child has stopped participating because the game is too hard or because the child is bored. If enough trials are omitted then the participant's information is usually omitted. This presents a particular problem for studies of children with autism who often have short attention spans and can be difficult to engage. In fact, a rule of thumb in working with children who are preschool aged and younger is to get the maximal amount of accurate information before something more interesting comes along. Prior to the start of this study, considerable effort was aimed at developing scales that do not rely on proportions but on a scale of difficulty (i.e., based on number of locations and length of delay). These scores reflect the maximum level that a child can perform rather than a proportion of correct performance that for this group may have become a measure of endurance more than of memory. Unfortunately, because the scales incorporate increases in the number of locations and length of delay with a variable rate of presentation (i.e., children receive only the number of trials needed to reach their maximal level of performance

based on multiple correct responses in a given level), it is not possible to determine which dimension of the scale, locations or time, create greater obstacles for children with autism. A direction for future work would be to determine the minimum number of search locations required to detect differences across a broad age range of children. Because this type of task can be used with individuals of all ages, and verbal and intelligence levels, this information could be used to construct a measure appropriate for longitudinal studies. As is always true with measurement development, the procedures for these working memory tasks will continue to be modified in future studies.

4.2 Long-term memory and deferred imitation

Individuals with autism tend to have impaired performance on episodic memory tasks but relatively preserved abilities on simple memory tasks or tasks that rely on 'rote' or semantic memory. Impaired episodic memory seems to be related to the complexity of the information, available encoding and recall strategies, and difficulties in temporal processing. Furthermore, source monitoring seems to be impaired, which undoubtedly contributes to disruptions in memory performance.

A major obstacle to the examination of early emerging long-term memory in children with autism is the lack of measures that do not rely on relatively sophisticated verbal skills, both receptive and expressive. In fact, it is not easy to conceptualize measures of long-term episodic memory that do not require verbal measurement. Obviously, this threshold excludes over half of the children with autism from being represented in studies of most aspects of long-term memory. The deferred imitation procedure used in the present study provides an exciting first look into episodic memory in children with autism who do not have sophisticated verbal skills.

The primary aim of this part of the study was to investigate whether children with autism showed memory impairments on tasks involving the imitation of action sequences when compared to a typically-developing verbal mental age matched comparison group. Additionally, the memory results were examined in relation to the structure of the to-beremembered sequence. Specifically, sequences with initial actions that enable subsequent actions may facilitate temporal recall. Sequences with initial actions that do not enable subsequent actions, or that occur in an arbitrary manner, will pose greater difficulty at recall. It was hypothesized that children with autism would not be significantly different from typically developing children on the enabling sequences.

Children with autism had significantly lower recall performance on all tasks, regardless of sequence structure. This could reflect that the sequence types are not processed differently in autism, which is inconsistent with other studies of nonverbal imitation (Bauer, 1996; Adlam et al., 2005). However, the children with typical development also did not differ significantly on their production of enabling versus arbitrary actions and action pairs. Therefore, results regarding the sequence type must be viewed with caution. Examination of the means for each of the conditions and sequences reflect trends in the expected direction so it is possible that with a larger group or shorter sequences these differences would be observed. Another possibility is that the sequences themselves are problematic. The sequences used here were modifications of published sequences, designed to increase the salience of the event for children with autism. It is possible that these changes also served as a distracter for children with typical development.

The results suggesting significantly poorer deferred imitation in autism should not come as a surprise based on at least two factors that underscore the neural substrates of this disorder. First, the brain regions that are used to perform this task (specifically, the medial temporal lobe), were the focus of a theory that proposed that autism could be characterized as a specific type of amnesiac disorder resulting from damage to the hippocampus and amygdala. The theory was developed on the basis of neuropsychological and neuroanatomical studies in which performance by individuals with autism was similar to performance by individuals with medial temporal lobe amnesia (DeLong, 1992). Also, Boucher found that individuals with low-functioning autism had poor free recall and recognition of pictures, written words, and spoken words compared to verbal and non-verbal ability in matched controls (Boucher, 1981; Boucher & Warrington, 1976). Neuroanatomical evidence was found in studies of humans with medial temporal amnesia when compared to those with autism (DeLong, 1992). The relationship between autism and amnesiac disorder has been supported by post-mortem neuroanatomical research showing abnormalities in the hippocampus and related structures in both clinical groups (Bauman & Kemper, 2005; Bauman, 1996). The amnesia theory of autism has been discounted because the memory impairments seen in autism are not as severe as those seen in medial temporal lobe amnesia, and memory results from individuals across the autism spectrum have been inconsistent (Minshew, et al., 1992; Minshew & Goldstein, 1993; Renner, Klinger, & Klinger, 2000; Rumsey & Hamburger, 1988). However, the role of the hippocampus, amygdala and frontal regions support performance on this task are still under investigation in autism.

Second, a previous study using the deferred imitation paradigm with individuals diagnosed with developmental amnesia associated with bilateral hippocampal volume reduction, found a similar pattern of results (Adlam et al., 2005). Notably, individuals with developmental amnesia displayed impaired episodic memory despite relatively preserved semantic memory (Adlam et al., 2005). This is similar to patterns seen at least on a group level in autism. Interestingly, Adlam used the deferred imitation paradigm with individuals in late childhood through early adulthood, which suggests that this paradigm has the flexibility to cover a broad age range suitable for longitudinal study.

An additional concern with the deferred imitation paradigm is the fact that children with autism are known to have difficulty with imitation (Charman et al., 2003 and McDuffie et al., 2005). Therefore, the results in this paradigm may not reflect memory but be a result of the child's inability to imitate. This is a reasonable hypothesis that future studies should examine specifically. However, the results here suggest that an inability to imitate cannot be solely responsible for the differences in the children with autism and typical-development because the children with autism were able to produce more actions and action pairs after observing the examiner demonstrate the task, thus indicating memory for the event and imitation ability. Also, the motivation to imitate may be enhanced when imitation causes an interesting event to occur.

The verbal mental age matching procedure was important in this study because it reduced any potential advantage that children who had more sophisticated language skills might have at encoding and subsequently at recall. Specifically, we eliminated the descriptions of task actions that are often used in the elicited imitation procedure. This was an extremely conservative decision. It is possible that labeling actions during the task

may have facilitated memory in both groups and not just the group with typical development. It will be helpful in future studies to examine the role that labeling actions has on encoding and subsequent recall.

Another factor that should be considered in future studies is sequence length. The length of sequence used in this study was selected based on work with typically developing children of approximately equivalent verbal mental age. However, future studies should examine shorter sequences to determine if sequence length was significantly related to memory performance. Additionally, it would also be interesting to see if children with autism exhibited increases in memory scores if the sequences were presented on previous recorded video tapes. This would remove the social pressure of having to interact with the examiner and might allow the child to focus more easily on the task.

Clearly, the deferred imitation paradigm offers many lines for inquiry into the factors that contribute to impaired episodic memory in autism. The flexibility of the paradigm makes it particularly well suited for longitudinal studies.

4.3 Brain development and memory

Although direct structural and functional measures of the brain were not included in this study, it is worth considering how the memory results from this study may be supported by studies on the brain in autism. Because autism is a neurodevelopmental disorder, understanding these brain-behavior connections is critical. Although a complete review of the literature supporting the neurobiological basis of autism is beyond the scope of this paper (for a review see Cody, Pelphrey, & Piven, 2002 and Akshoomoff, Pierce, & Courchesne, 2002), a few results are of particular relevance for a discussion of

memory. Unfortunately, the neurological findings are filled with the same pattern of inconsistencies that are found in the behavioral studies. For example, studies examining the integrity of the hippocampal formation contain reports of reduced volume (Aylward, Minshew, Goldstein, Honeycutt, Augustine, Yates, et al., 1999; Saitoh, Karns, & Courchesne, 2001), while others have found no abnormalities (Piven, Bailey, Ranson, & Arndt, 1998; Haznedar, Buchsbaum, Wei, Hof, Cartwright, Bienstock, et al. 2000). Furthermore, postmortem studies have confirmed increased cell density and abnormally small cells in the hippocampal formation (Raymond, Bauman, & Kemper, 1996). A recent study by Salmond et al. (2005) showed increased grey matter density in the hippocampal formation and peri-hippocampal cortex, as well as the fusiform gyrus, dorsolateral prefrontal cortex and cerebellum. This study also reported behavioral data consist with impairments in episodic memory that is consistent with the results from the elicited imitation task in our study.

Results from an MRI study of adolescents and adults with high-functioning autism indicate increased volumes of the parietal, temporal, occipital lobes and total brain volume but no increases in frontal lobes, compared to controls (Piven, Arndt, & Bailey, 1996). Thus, relative to the rest of the brain, the frontal lobes may be the most abnormal in volume (Eigsti and Shapiro, 2003). Frontal regions are responsible for executive function (including working memory) although other brain regions may be recruited as well depending on the task. Recent functional imaging studies show differences in the dorsolateral prefrontal cortex in adolescents with autism as compared to typically developing children (Silk, Rinehart, Bradshaw, Tonge, Egan, O'Boyle, et al., 2006). This

is the same area that is activated in delayed non-match to sample tasks and delayed response tasks similar to those used in this study.

4.4 Relevance

The increasing emphasis on early identification through screening (Gillberg et al., 1990; Reznick et al., 2007) and studies of 'at-risk' infant siblings of children with autism, presents an opportunity and challenge. Measures that can be used with preverbal infants and older children can reveal the developmental origins of executive function and different components of long-term memory. These measures can then be paired with data that reflect the timing of abnormal neural development in autism that appears to be going awry long before the point in time that most current behavioral studies can assess. The integration of biological and behavioral factors provides the greatest likelihood of elucidating developmental mechanisms in autism. Moreover, understanding these mechanisms can provide points for critical intervention.

4.5 Toward a developmental model of autism

Increasingly it has been acknowledged that the investigation of developmental processes, whether typical or atypical, must involve investigators from every discipline interested in human function (i.e., psychology, biology, neurology, chemistry, genetics and so on). To that end, Gottlieb (2002) described a model in which genetic activity, neural activity, behavior and environment influence each other bi-directionally over time to produce psychobiological development. In this model, all levels of an organism are of equal importance with no a priori deterministic assumptions. The need for models such as this seems self-evident when one considers the study of autism. In fact, we now have evidence that supports behavioral, genetic, neural and environmental contributions to the

development of autism. However, it should be noted that psychobiological models do not focus only on the cross-level (i.e., genes-brain-behavior) interactions but within-level interactions. In the case of memory, how are different behavioral traits working together to produce memory outcomes? We must also consider how different memory subtypes are interacting to create memory (i.e., executive processes for integrating new and old memories).

For a simple example, consider the role of social function. A child who fails to engage in social behavior will receive less stimulation, support for learning, and reduced opportunities to communicate. All of these constructs are interacting to facilitate development, and the absence of any construct will alter the system. Consider once again Kanner's quote about the remarkable memory ability of the children with autism. He later discusses how the parents encourage the rote memory behavior in these children almost to the exclusion of other activities. Kanner does not discuss the possibility that the parent's desire to promote the unusual memory abilities in the child may be driven by the parent's inability to connect with the child in a more conventional manner due to social and language deficits in the child. In this way, parents capitalize on an intact ability as an opportunity for some type of social interaction with their child. Of course, as Kanner points out, simply promoting rote memory skills will not in and of itself encourage the development of meaningful language or social skills that these children lack. Moreover, development of memory is a complex process influenced by and having an influence on the development of language, knowledge and social skills. Although few empirical studies of memory have been conducted in a manner that fully incorporates multiple levels, conceptualizing memory as being 'socialized' is becoming more widely

adopted (Ornstein, Haden, & Hedrick, 2004) and might provide new sights into the unique pattern shown in autism.

Unfortunately, the corpus of information on the development of autism is limited, representing snapshots of memory abilities and dysfunction across a broad range of ages, language levels and global cognitive abilities. The present study provides another marker along the road, although it is clearly a marker closer to the beginning of the trail. Nevertheless, there is room for considerable optimism that we may be able to elucidate the ontogeny of memory in autism given improvements in early diagnosis, technology, cross discipline collaborations, and funding.

In summary, memory function in autism remains a compelling topic replete with possibilities for advancing our understanding of the etiology of this complex disorder. The pace of our progress on this frontier will be set by our willingness to incorporate developmental models and methods. There is reason to believe that when design and methodology are aimed at early development, data will emerge that provide a meaningful anchor for understanding the development of memory in autism. It is through development that autism begins. Therefore, it is through development that we will resolve the mysteries of the disorder.

Measure	Aut	Autism Typically-developing				
	M (SD)	Range	M (SD)	Range	<i>t</i> (df)	Р
Chronological age in	41.76 (5.77)	32.0-52.0	27.95 (4.12)	22.0-36.0	-8.92 (40)	.001
months						
Mullen Receptive Language	30 (4.86)	23.0-39.0	30.1 (4.73)	23.0-39.0	.064 (40)	.949
age equivalent						
Mullen Expressive	25.9 (4.18)	20.0-33.0	31.48 (5.09)	24.0-42.00	3.87 (40)	.001
Language age equivalent						
Mullen Expressive	30.52 (4.95)	20.0-33.0	25.28 (4.1)	24.0-41.0	3.70 (40)	.001
Language age equivalent						

Table 1. Comparison by groups on chronological age and Mullen sub-scales

Table 2. Comparison by group on ADOS domains

	Autism		Typic	ally-		
			develo	oping		
Domain	M (SD)	Range	M (SD)	Range	<i>t</i> (df)	Р
Communication	5.57 (1.29)	4 - 8	.29 (.46)	0 - 1	17.76 (40)	<.001
Social	9.57 (2.18)	6 - 14	.57 (.81)	0 - 2	17.07 (40)	<.001
Communication and	15.14 (2.74)	11 - 19	.86 (.96)	0 - 3	22.51 (40)	<.001
Social						

Sub-scale	M (SD)	Range
Communication	8.52 (1.08)	7 - 11
Social	19.10 (3.59)	12 - 26
Ritualistic/Repetitive Behavior	4.33(1.15)	3 - 8

Table 3. Descriptive statistics for the ADI-R domains for children with autism

	Auti	sm	Typically-o	developing		
Measure	M (SD)	Range	M (SD)	Range	<i>t</i> (df)	Р
Cotton Wells	1.14 (.70)	.5 - 3.0	3 (1.33)	.5 - 5.0	5.63 (40)	<.001
Egg Hunt	1.45 (.86)	.5 – 3.0	3.33 (1.29)	1.0 - 5.5	5.53 (40)	<.001
Animal Houses**	1 (.48)	.5 – 2.0	3.33 (.92)	2.0 - 5.0	9.97 (31.4)*	<.001
Nonsense Words	.82 (.78)	.25 – 2.75	3.35 (.87)	1.25 - 5.0	7.93 (28)	<.001

Table 4. Comparison by group on working memory tasks

*Degrees of freedom corrected for unequal variances **Non-parametric Mann-Whitney test also computed

		Cotton Wells	Egg Hunt	Animal Houses	Nonsense Words
	r	1.000			
Cotton Wells	р		-	-	-
	Ν	21			
	r	.484	1.000		
Egg Hunt	р	.026	-	-	-
	Ν	21	21		
	r	.420	.335	1.000	
Animal Houses	р	.106	.205		-
	Ν	16	16	16	
Nonsense Words	r	.591	.073	.135	1.000
	р	.055	.832	.729	
	Ν	11	11	9	11

Table 5a. Correlation matrix of working memory tasks for the autism group*

* Spearman correlation, two tailed test

		Cotton Wells	Egg Hunt	Animal Houses	Nonsense Words
	r	1.000			
Cotton Wells	р	•	-	-	-
	Ν	21			
	r	.781	1.000		
Egg Hunt	р	.000	-	-	-
	Ν	21	21		
	r	.435	.460	1.000	
Animal Houses	р	.049	.036		-
	Ν	21	21	21	
Nonsense Words	r	.662	.655	.241	1.000
	р	.002	.002	.320	
	N	19	19	19	19

Table 5b. Correlation matrix of working memory tasks for the typically-developing group*

* Pearson correlation, two tailed test

			ine	Recall	
Measure	Group	M (SD)	Range	M (SD)	Range
Enabling Action	1				
	Autism	.86 (.73)	0-2	2.48 (1.03)	1-4
	Typical-Development	1.33 (.86)	0 - 3	4.86 (1.06)	3 - 7
Arbitrary Action	1				
	Autism	.81 (.68)	0-2	1.90 (1.14)	0-4
	Typical-Development	1.10 (.77)	0 - 3	5.10 (1.09)	3 - 7
Enabling Pair					
	Autism	.10 (.30)	0 – 1	.76 (.83)	0-2
	Typical-Development	.19 (.40)	0 - 1	2.00 (1.18)	0 - 4
Arbitrary Pair					
	Autism	.05 (.22)	0 – 1	.43 (.68)	0-2
	Typical-Development	.19 (.40)	0 - 1	1.48 (.93)	0 - 3

Table 6. Descriptive statistics by group on elicited imitation tasks

UL

UL

UL

Level-					
T	s	Loca	tion		
test	0	L	R		
test	0	L	R	Level- T	s
1-1	7	L	R	3-1	14
1-2	7	ر ۲	R	3-2	14
1-3	7	L		3-3	14
2-3	7	L	R	4-1	14
		T UL			
2-3	7	L	R	4-2	14
		UL	UR		
2-3	7	ر ۲	R -	4-3	14
		UL	UR		

Cotton Wells

	_			
Location		Level- T	s	Location
L R T		5-1	21	L R
ι R τ		5-2	21	L R T
L R T		5-3	21	L R T
L R T		6-1	21	L R T
UL UR				UL UR
L R T		6-2	21	L R T
UL UR				UL UR
LR		6-3	21	LR
т				Т
UL UR				UL UR

Egg	Hunt

Level- Trial	s	Location
test	0	L R
test	0	L R
1-1	7	LTR
1-2	7	L T R
1-3	7	L T R
2-3	7	UL L T R UR
2-3	7	ullt R ur
2-3	7	ull T rur

Level-Trial	s	Location	
3-1	14	L T R	
3-2	14	L Т R	
3-3	14	L TR	
4-1	14	ULLT R UR	
4-2	14	ULLTR UR	
4-3	14	ulltr UR	

Level- Trial	s	Location	
5-1	21	L Т R	
5-2	21	L TR	
5-3	21	L T R	
6-1	21	ULL T R UR	
6-2	21	ULLTR UR	
6-3	21	UL L T R UR	

I			Annua nous	-	
Test	0			PIG	CAT
Test	0			PIG	CAT
1-1	3	CAT	PIG		
1-2	3	CAT	PIG		
1-3	3	PIG	CAT		
2-1	3		CAT	TURTLE	PIG
2-2	3		TURTLE	PIG	CAT
2-3	3		PIG	CAT	TURTLE
3-1	3	CAT	TURTLE	WHALE	PIG
3-2	3	WHALE	PIG	TURTLE	CAT
3-3	3	TURTLE	CAT	PIG	WHALE
4-1	6	CAT	PIG		
4-2	6	CAT	PIG		
4-3	6	PIG	CAT		
5-1	6		CAT	TURTLE	PIG
5-2	6		TURTLE	PIG	CAT
5-3	6		PIG	CAT	TURTLE
6-1	6	CAT	TURTLE	WHALE	PIG
6-2	6	WHALE	PIG	TURTLE	CAT
6-3	6	TURTLE	CAT	PIG	WHALE

Animal House

NONSENSE WORD REPETITION

- Complete all trials in any level that you begin.
- Do not move to a new level after 2 errors in any level.
- You may repeat the trial no more than 3 times.

Level 1 TOE EGG TUR UG Level 2 DAL LER SER PEM TUR UG Level 3 SAH POE VOOR NA NA BA GAZ A HEEN Level 4 TONK VOO PER UG LE DEESE LEH BOON SHA HEEN COMP DUR Level 5 RIG A SEG JOM IC LOD I VAY FLA ZEE PEM SER MO SAH VOOR Level 6 NA GAZ POE UG DUR TONK BOON SHA PEM VOOR I FLA SER DAL VO DEESE HEEN COMP

APPENDIX B. Description of deferred imitation sequences

Ramp

Description: A hinged wooden ramp is opened (open). When open the ramp has a peg on the side that should be placed facing the ceiling on the elevated section of the ramp (base). The examiner may help the child orient the ramp in this fashion without penalty to the child. A car is then placed on the ramp behind the peg (car). The peg holds the car in place until it is removed in the final action (peg).

Coding guidelines:

-Open: In baseline, any attempt to open the ramp is scored. During recall, the child must open the ramp past 90 degrees to receive credit.

-Base: Any attempt to orient the base to the ramp is scored.

-Car: In baseline, any attempt to place the car on the ramp is scored. During recall the car must be placed behind the peg to receive credit.

-Peg: Any attempt to remove the peg receives credit. Pushing the car down the ramp without placing it on the platform will not receive credit.

<u>Funnel</u>

Description: A stand is created by placing a rod with an attachment on the top into a base (holder). A funnel is then placed into the attachment (funnel). The end of the funnel is closed with a cork. Balls are placed in the funnel (balls). The cork is removed so the balls can fall (stopper).

Coding guidelines:

-Holder: In baseline and recall, attempts to place the rod into the base receive credit. The examiner may help the child insert the rod. Watch for intention.

-Funnel: If the funnel is placed in the circular holder, credit is given.

-Balls: Placing the balls in the cup of the funnel receives credit.

-Stopper: Removing the stopper from the funnel receives credit. The examiner may assist the child in removing the stopper. Watch for intention.

<u>Train</u>

Description: The removable door on the car is placed in the door opening (door). Wooden sticks are displayed in front of the train (tracks). Blocks are placed in the train as cargo (load). A pivoting stick attached to the train is raised by pressing the stick (lift).

Coding guidelines

-Door: Attempts to put the door in the door opening receive credit.

-Tracks: Any attempt to orient the track to the train is given credit. Watch for intention. -Load: In baseline, any attempt to put the cargo in the train is scored. In recall, it must be loaded through the top of the car to receive credit.

-Lift: In baseline, any attempt to raise the brake will be coded. In recall, the child must push the top of the break to receive credit.

<u>Boat</u>

Description: A flat narrow piece of wood is put into a slot on the boat (plank). A flag is placed into a holder on the boat (flag). Plastic fish are placed onto the back of the boat (fish). An anchor attached to the boat is raised by pulling the string (anchor).

Coding guidelines

-Plank: In baseline, any attempt to put the plank on the boat is scored. In recall, the plank must be placed in the slot to receive credit. The examiner may assist. Watch for intention. -Flag: Any attempt to place the flag into the holder is scored. The examiner may assist. Watch for intention.

-Fish: Any attempt to put the fish in the boat is coded.

-Anchor: In baseline, any attempt to raise the anchor is scored. Watch for intention. In recall, the child must pull the rope from the end to lift the anchor to receive credit.

APPENDIX C. List of actions and action pairs for each deferred imitation task

Enabling		Arbitrary	
	Ramp		Train
1	Open	1	Door
2	Base	2	Tracks
3	Car	3	Load
4	Peg	4	Lift
	Funnel		Boat
1	Holder	1	Plank
2	Funnel	2	Flag
3	Balls	3	Fish
4	Stopper	4	Anchor

Action pairs for all sequences:

1	-	2
2	-	3
3	-	4
	2	1 - 2 - 3 -

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