DISSERTATION

THE UNDERSTANDING OF INTENTIONALITY IN CHILDREN WITH WILLIAMS SYNDROME AND DOWN SYNDROME

Submitted by

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In partial fulfillment of the requirements

For the Degree of Doctor of Philosophy

Colorado State University

Fort Collins, Colorado

Summer 2012

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ABSTRACT

UNDERSTANDING OF INTENTIONALITY IN CHILDREN WITH WILLIAMS SYNDROME AND DOWN SYNDROME

This dissertation examined the development of the understanding of intentionality in two different neurogenetic disorders, Williams syndrome (WS) and Down syndrome (DS). The study of intentionality focuses on how children come to understand the intentions of others. Meltzoff’s (1995) behavioral reenactment paradigm is a nonverbal procedure wherein a child is presented with a series of objects. Prior to each presentation, the examiner either performs a successful action (e.g. the target action) or an unsuccessful action (e.g. the failed intentional action). A child’s understanding of intentionality is assessed by their ability to interpret the experimenter’s intention during failed attempt trials, and their subsequent completion of the task. This examination of intentionality was divided into two studies.

Study 1 was designed to test Tager-Flusberg and Sullivan’s (2000) hypothesis that there is a dissociation between social-perceptual abilities and social-cognitive abilities in individuals with Williams syndrome. In order to explore this dissociation, the behavioral reenactment procedure was administered with and without experimenter affective cues. Participants were 25 children with a confirmed diagnosis of WS. There were two groups of WS, one that received affective cues (N=13) and one that did not (N=12). Also, children with WS in the no affect group were compared to 12 mental-age matched children with developmental disabilities. The findings of this study indicates that the understanding of intentionality improves with developmental status in children with WS. Also, this study indicates that there may be a
dissociation between social-perceptual and social-cognitive skills in this population during early social-emotional development. Specifically, it seems that the presence of emotional cues during intersubjective tasks leads to an emotional response instead of a response based on social cognition.

Study 2 was motivated by past research suggesting that children with DS demonstrate deficits in some aspects of social cognition, even though many children with DS have strengths in other aspects of social-emotional functioning. Therefore, it is likely that the understanding of intentionality in children with Down syndrome may be influenced by other foundational cognitive abilities (i.e. joint attention and affect sharing in early childhood and executive functioning in middle childhood). Participants were 40 children with a confirmed diagnosis of Down syndrome, 16 young children with DS and 24 older children with DS. In addition, the 16 young children with DS were compared to 16 mental-age matched children with other developmental disabilities. The results of this study suggests that the understanding of intentionality improves with developmental status for young children with DS. This study also suggest that difficulties in joint attention and EF lead children with DS to miss the target relevant information during the behavioral reenactment procedure leading them to perform more “other actions”.

This dissertation is the first study to examine the development of intentionality in WS and DS. From these studies, it may be possible to begin to characterize how the understanding of intentionality develops in children with WS and DS. Characterizing social cognition in WS and DS will help to identify areas for targeted intervention to prevent the possible cascading effects of difficulties in social cognition on other aspects of development.
I would like to thank Deborah Fidler for all of the guidance and support she has given me over the last four years. Her mentoring has provided me with the skills and knowledge needed to successfully complete this project and prepare me for my future career. I would like to thank David Most for his methodological expertise and his willingness to discuss methodology with me. I would also like to thank Erika Lunkenhiemer and Meena Balgopal for their thoughtful and helpful contributions to this project.

Finally, I would like to thank Bill and Wilma Hahn for their support throughout my academic endeavors and for always believing in me. I would not have achieved this great accomplishment without them.
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CHAPTER 1 – INTRODUCTION

Background

Theory of the mind is the social cognitive milestone that allows humans “to predict and explain other people’s actions based on inferences about their mental states such as intentions, knowledge, or beliefs” (Tager-Flusberg & Plesa Skwerer, 2007, p. 97). Theory of mind is critical for navigating complex social dynamics and interactions because this skill allows individuals to theorize about what another person is thinking and take their perspective (Zinck, 2008). According to Meltzoff (1995), a child’s earliest understanding of theory of mind begins with reasoning about mental states like desires and intentions. The study of intentionality focuses on if and when children are able to understand the intentions of others (Meltzoff, 1995). Two populations that may exhibit unique development of the understanding of others’ intentions are children with Williams syndrome (WS) and children with Down syndrome (DS) because of the unique social cognitive profiles associated with each syndrome.

Research Problem

The purpose of this dissertation was to examine the understanding of intentionality in children with WS and DS, and whether these two syndromes predispose children to specific areas of strength or challenge in this area. In order to examine the understanding of intentionality in these two groups, Meltzoff’s (1995) behavioral reenactment procedure was used. The behavioral reenactment procedure is a nonverbal experimental paradigm where a child is presented with a series of objects. Prior to each presentation, the examiner either performs a successful action (e.g. the target action), such as putting beads in to a cup, or an unsuccessful action (e.g. the failed intentional action/failed attempt), such as trying, but failing to put beads in to a cup.
Study 1 was designed to test Tager-Flusberg and Sullivan’s (2000) hypothesis that there is a dissociation between social-perceptual abilities and social-cognitive abilities in individuals with WS when performing theory of mind tasks. The understanding of intentionality involves both social-perceptual and social cognitive skills. Social-perceptual skills include understanding and responding to nonverbal behaviors (such as directing facial expressions, directing vocalizations, responsive smiling, and shared affect) that involve dynamic matching and adapting to another’s behaviors (Tager-Flusberg & Sullivan, 2000). Social-cognitive skills are thought to be an extension of social-perceptual skills and include the abilities involved in the traditional conceptualization of theory of mind (e.g. shared and coordinated attention to an object or event, perspective taking, the attribution of mental states, etc.; Tager-Flusberg, 2005; Tager-Flusberg & Sullivan, 2000).

Young children with WS have been shown to demonstrate heightened levels of emotional responsivity and are more likely to imitate the emotional displays of their social partner (Fidler et al., 2007). However, previous studies have shown that knowledge of a social partner’s emotions does not appear to inform social decision making in many young children with WS (e.g. giving a disliked food even after the dislike has been indicated; Fidler et al., 2007). This suggests that children with WS may have difficulties taking the perspectives of others, despite being responsive to affective cues. Thus, there is evidence of a dissociation in social-perceptual abilities and social-cognitive abilities during childhood in this population (Fidler et al., 2007; Hepburn et al., 2011). Because understanding intentionality involves both social-perceptual and social-cognitive skills, characterizing skills in both areas in children with WS will further describe the proposed dissociation between these domains. For this study, the behavioral reenactment procedure was administered with and without affective cues to explore the
dissociation between social-perceptual and social-cognitive abilities in young children with WS. If this dissociation is evident in the understanding of intentionality, then children with WS who receive affective cues should attend more to the emotional information presented during the task and miss the necessary social-cognitive information needed to successfully complete the task.

Study 2 was motivated by past research suggesting that children with DS demonstrate deficits in some aspects of social cognition, even though many children with DS have strengths in other aspects of social-emotional functioning (Fidler, 2006). Based on existing work, it is unclear whether the understanding of intentionality is compromised in children with DS beyond the level of understanding that would be expected for their mental age (MA). In addition, the competent development of the understanding of intentionality in children with DS may be influenced by other related cognitive and social cognitive abilities. In early childhood, it is possible that skills related to intersubjectivity, like joint attention and affect sharing, are correlated to the understanding of intentionality. Infants with DS achieve the developmental milestones related to primary intersubjectivity and secondary intersubjectivity at MA appropriate levels (Fidler, 2006). The development of both primary and secondary intersubjectivity includes the development of joint attention and affect sharing. Joint attention occurs when individuals engage in a reciprocal interaction around an object or event (Mundy & Newell, 2007). Affect sharing is a combination of a joint attention behavior and emotional signals (Seibert, Hogan & Mundy, 1982; Mundy, Sigman, & Kasari, 1990).

For older children with DS, the understanding of intentionality may be related to executive function skills. Research on adults with DS suggests that theory of mind skills are positively related to their executive function skills (Zelazo, Burack, Benedetto, & Frye, 1996). ‘Executive function’ (EF) is an umbrella term used to describe the self-regulatory cognitive
processes integral to adaptive, goal-directed actions, including working memory, inhibition, shifting, and planning (Blair, Zelazo, & Greenberg, 2005; Carlson, 2005). Although the relationship between executive function skills and theory of mind abilities has been examined in adults with DS (Zelazo et al., 1996) and typically developing children (Carlson, Mandell, & Williams, 2004a; Carlson, Moses, & Claxton, 2004b; Hughes, & Ensor, 2007; Rakoczy, 2010), this relationship has yet to be examined in children with DS. It is possible that executive function abilities may be related to intentionality because successful completion of the target action involves planning regarding the proper completion of the action based on the failed intentional information they observed (by using working memory). Also, during the intentionality task children with DS may need to inhibit any processes telling them to imitate what was observed.

For this study, performance on the behavioral reenactment procedure of young children with DS was compared to that of older children with DS. In addition, the performance of children with DS on the behavioral reenactment procedure was compared to that of children with other developmental disabilities (DD). Finally, the relationship between the performance on the behavioral reenactment procedure and joint attention, affect sharing, and executive functioning was explored.

**Research Questions and Hypotheses**

**Study 1**

1. Is there a dissociation between social-perceptual and social-cognitive abilities in WS?
   a. Children with WS who receive affective cues in the administration of the intentionality task will perform more target actions on the target administration
and will imitate the failed intention more on the failed intention administration than children with WS who do not receive affective cues.

b. Children with WS who do not receive affective cues will perform more target actions on the failed intention administration than children with WS who receive affective cues.

c. When compared to children with DD, children with WS will imitate the failed intention more.

d. There will be an increased likelihood of affective responses for children with WS who experienced affective cues when compared to children with WS who do not receive affective cues.

2. What are the magnitudes of the association between chronological age, nonverbal-mental age, and mental age on task performance in children with WS?

a. Developmental status (chronological age, nonverbal-mental age, and mental age) will be positively correlated with performance of the target action on the failed intention administration.

b. Developmental status (chronological age, nonverbal-mental age, and mental age) will be negatively correlated with imitating the failed intention on the failed intention administration.

Study 2

1. What are the magnitudes of the associations between task performance and other domains of cognition in children with DS?

a. The task performance of younger children with DS will be correlated with joint attention and affect sharing performances.
b. The task performance of older children with DS on this task will correlate with executive function skills.

2. What are the age-related changes in the understanding intentionality in children with DS? Are the age-related changes similar or different from children with other developmental disabilities (DD)?

a. The understanding of intentionality in children with DS will improve with developmental status (chronological age, nonverbal-mental age, and mental age).

b. Performance on the intentionality task will be similar between young children with DS and young children with DD.

List of Keywords

Social Cognition – ability to think and reason about the social world (Cebula & Wishart, 2008; Tager-Flusberg et al., 2006; Trevathan & Aitken, 2001). This involves a wide range of abilities including interaction and social referencing, social attribution, interpretation of cues, face recognition, communication, and theory of mind (Tager-Flusberg et al., 2006).

Theory of Mind – the understanding that other minds exist, this ability allows humans “to predict and explain other people’s actions based on inferences about their mental states such as intentions, knowledge, or beliefs” (Tager-Flusberg & Plesa Skwerer, 2007, p. 97).

Intentionality – understanding the intentional of actions of others (Meltzoff, 1995).

Williams syndrome – a neurogenetic disorder caused by a deletion on chromosome 7 that results in a behavioral phenotype characterized by relative strengths in expressive language and
aspects of social functioning and relative weaknesses in spatial cognition and visual spatial processing.

**Down syndrome** – a neurogenetic disorder caused by an extra copy of the 21st chromosome that results in a behavioral phenotype characterized by relative strengths in visual short-term memory, receptive language, and some aspects of social functioning and relative weakness in areas related to communication and verbal short-term memory.

**Researchers Perspective**

My training has positioned me to be a post-positivist and for many years I did not question this approach, but as my understanding of research methodology and theory has grown, so has my understanding of post-positivism. Although there are aspects of the paradigm I do not completely agree with, the metaphysics of the paradigm lend themselves well to my research on neurogenetic disorders (e.g. Down syndrome and Williams syndrome). The field of neurogenetic disorders is focused on identifying the characteristics associated with various genetic causes of intellectual disability. The purpose of this dissertation was to examine the development of intentionality in children with WS and DS. Therefore, the most appropriate perspective to design this study from is post-positivism because of the underlying assumptions of the paradigm, which includes the ontology, epistemology, methodology, axiology, and teleology.

According to Guba (1990), ontology is what makes for reality. In post-positivism, there is the understanding that reality exists, but it can never be truly attained (Guba, 1990); nonetheless, as researchers we do the best we can to understand it (Green, 1990). Nonetheless, as researchers from this paradigm we do the best we can. When it comes to neurogenetic disorders, we know there is a chromosomal abnormality influencing the development of the
individual, which means these problems are originating from a genetic trait and not social
collection. For that reason, the post-positivistic view of ontology is an appropriate stance for
research into neurogenetic disorders.

What makes for knowledge of reality (Guba, 1990), or the epistemology, from a post-
positivist perspective is based on the generation of scientific facts developed from observation
and logic, with an emphasis on objectivity (Merriam, 1991). Thus the methodology of post-
positivism, or how knowledge is accumulated (Guba, 1990), takes the form of modified
experiments that are conducted in more natural settings, but still use statistics to approximate
reality (Guba & Lincoln, 2005). When it comes to the issue of objectivity we strive to be
objective as possible, but we are far from the traditional notion of the disinterested scientist. For
example, we take an objective stance in collecting our data by following protocols for the
administration and coding of the tasks we are studying. However, we also put the needs of the
child and family above our own goals as researchers, in order to establish rapport and make sure
the families are getting what they need from these visits with us. Although we want to gather
information in order to inform the field, our ultimate goal is to promote well-being in these
children and their families, which means we are far from disinterested scientists. Nonetheless,
we are still conducting experiments with these children following the basic ideals of the
scientific method, which aligns with the axiology of post-positivism.

Axiology is how we, as researchers, act in producing and applying knowledge (Guba &
Lincoln, 2005). Knowledge is produced via the scientific method in post-positivism (Merriam,
1991), which involves propositional knowing (Guba & Lincoln, 2005). Propositional knowing is
the understanding that the world is an end in itself, but we will never come to know it completely
(Guba & Lincoln, 2005). This view of propositional knowing aligns with the probabilistic
approach used in this dissertation to understand development in neurogenetic disorders (Dykens, 1995). The probabilistic approach is the idea that although we have identified certain behaviors and outcomes (i.e. behavioral phenotypes) for individuals with neurogenetic disorders, not all individuals with a specific disorder will display all these characteristics and behaviors (Dykens, 1995). In DS, for example, the behavioral phenotype is influenced by the context of the child (e.g. parenting styles, types of intervention, available resources, etc.) and the type of genetic abnormality (i.e. trisomy 21, translocation, or mosaicism).

Finally, the teleology, or how knowledge is applied (Guba & Lincoln, 2005), focuses on producing objective, scientific explanations (Merriam, 1991). Although this research is producing explanations about the development of individuals with neurogenetic disorders, it is not the only way this research is being applied. Our research is also used to promote well-being, develop interventions, and add information to practice and policy. Therefore this research is taking the next natural step in post-positivism by applying knowledge practically, not just scientifically, making it truly applied research.
CHAPTER 2 – LITERATURE REVIEW

Social cognition involves the ability to think and reason about the social world (Cebula & Wishart, 2008; Tager-Flusberg et al., 2006; Trevarthen & Aitken, 2001). Social cognitive skills influence many other areas of development (Bukowski et al., 1996, Carpendale & Lewis, 2006; Cebula & Wishart, 2008; Flavell et al., 2002) and have “implications for learning, for making friendships, and ultimately for social emotional well-being and quality of life” (Cebula & Wishart, 2008, p. 45). Within social cognition, researchers have studied numerous phenomena, including social referencing, social attribution, interpretation of cues, face recognition, communication, and the ability to understand the thoughts, feelings, and motivations of others (Tager-Flusberg et al., 2006).

From a developmental perspective, social cognition depends on interactions between the self and others, and begins in infancy with primary and secondary intersubjectivity (Meltzoff, Gopnik, & Repacholi, 1999). These early intersubjective skills serve as a foundation for more advanced social cognitive skills (i.e. understanding of intentions, theory of mind, social decision making) that allow humans to think, reason, and interact in the social world (Cebula & Wishart, 2008; Meltzoff, 2007 Tager-Flusberg et al., 2006; Trevarthen & Aitken, 2001). Specifically, the development of intersubjectivity provides young children with the skills needed to understand the intentions of others (Trevarthen, 1978; Trevarthen & Aitken, 2001) and, in turn, the understanding of the intentions of others supports the development of theory of mind (Meltzoff, 2007; 1995). Therefore, to understand the development of intentionality in children with WS and DS, we first must review what is known about intersubjectivity and intentionality in typically developing children.


**Intersubjectivity**

Intersubjectivity is the intuitive recognition and understanding of the impulses and desires of another’s mind (Trevarthen, 1978). The first signs of social understanding develop during infancy, involving emotional displays and affect sharing between the infant and caregiver during reciprocal interactions (Trevarthen & Aitken, 2001). At 2 months, infants and caregivers actively participate in mutually regulated, dynamic interactions. This includes taking turns initiating and responding to the feelings and interests that are being displayed by each social partner during a social interaction (Trevarthen, 1979). These interactions serve as a foundation for the development of primary intersubjectivity (Trevarthen, 1979). From these interactions, an infant begins to predict what their social partner knows and what they will do, and they begin to share mental control of social interactions with another person (Trevarthen & Aitken, 2001).

By the middle of the first year, infant interests in objects increases with their use of protoconversations, leading to more elaborate games with objects (Trevarthen & Aitken, 2001). Joint attention emerges before the end of the first year, which is instigated by a shift in infants attention to their surroundings and the world (Hubley & Trevarthen, 1979), and has tremendous consequences for learning by changing the way caregivers interact with their infant (Trevarthen & Aitken, 2001). The emergence of joint attention leads to two types of awareness, one for objects and one for people (Trevarthen, 1998), which in turn instigates the emergence of secondary intersubjectivity, or person-person-object awareness, around 9 months of age (Trevarthen & Hubley, 1978). Together, the development of primary and secondary intersubjectivity provides infants and young children with the necessary skills to function in the social world (Meltzoff & Moore, 1994; 1998; Trevarthen & Aitken, 2001).
What Infants Gain from Intersubjectivity

Though intersubjectivity, infants learn to react dynamically to another person’s expressions in order to match their distinct pattern and rhythm. They also orient to and imitate how others use and handle objects (Trevarthen & Aitken, 2001), which highlights the central role of imitation in the infant’s intersubjective development (Meltzoff & Moore, 1998; Trevarthen & Aitken, 2001). Early in development, imitation serves to qualify attachments (Meltzoff & Moore, 1994) and emphasize an infant’s innate motivation for human contact and engagement (Trevarthen & Aitken, 2001). For older infants and toddlers, imitation is used to demonstrate and reinforce relationships (Trevarthen & Aitken, 2001). Imitation leads older infants and toddlers to begin to see people as intentional beings because people can be imitated (Meltzoff & Moore, 1998). Thus, through the development of intersubjectivity skills, young children begin to attribute emotions, desires, and intentions to the actions people produce (Trevarthen, 1978; Trevarthen & Aitken, 2001). According to Tager-Flusberg (2005), “understanding people as intentional, mental beings is at the core of social cognition” (p. 276).

Development of the Understanding of Others’ Intentions

In the first few years of life, young children come to understand others as intentional agents who act on the world in similar ways to the child’s actions on the world (Meltzoff, 2007, Meltzoff et al., 1999). Once children begin to see others as intentional, they are able to read through the surface behavior of an action to the underlying intention (Johnson, 2005; Meltzoff et al., 1999). Because of the importance of intention reading in the development of more complex theory of mind (Meltzoff, 2007), researchers are interested in when children begin to reason about the intentions of others (Meltzoff, 1995).
Intentionality

To investigate when young children begin to understand the intentions of others, Meltzoff (1995) developed a research paradigm called the ‘behavioral reenactment’ procedure. The behavioral reenactment procedure is a nonverbal task designed to examine whether children will interpret an individual’s behavior literally or if they can read through their behavior and identify the intended goal of the individual’s actions. In Meltzoff’s (1995) procedure, an experimenter presented the child with five novel objects (e.g. dumbbell, box and stick tool, prong and loop, cylinder and beads, square and post) and performed specific actions with the objects. In the target condition, the examiner successfully completed the action with the objects in full (i.e. the dumbbell was picked up and held at each end and then pulled outward so that it came apart at the mid-point; Meltzoff, 1995). In the failed intention condition, however, the examiner did not successfully accomplish the final goal state with the objects (Meltzoff, 1995). The experimenter tried, but failed, to complete the target action.

In Meltzoff’s (1995) original study, he reported that typically developing 18-month-old children were able to infer the intended actions of others as measured by the behavioral reenactment procedure (Meltzoff, 1995). In this study there were two control conditions, the baseline condition and the adult manipulation condition. In the baseline condition, the objects were presented to the children, but the examiner performed no action (Meltzoff, 1995). This condition controlled for the possibility that the objects may afford certain actions and to see if children would spontaneously produce the target action. In the adult manipulation condition, the examiner would handle the objects, but the examiner did not perform an action with them (Meltzoff, 1995). This condition was included to control for the possibility that children may be more likely to manipulate the objects if they observed someone else manipulating them.
Participants in both the target and failed intention conditions performed significantly more target actions than subjects in the control conditions (Meltzoff, 1995). Meltzoff (1995) concluded that at 18 months, infants could infer intentionality in others. The ability to understand intentionality suggests that infants have begun to differentiate surface behavior (what an individual does) from what an individual is trying to do (Meltzoff, 1995).

**Development of Intentionality.** While Meltzoff (1995) reported the presence of intentionality skills at 18 months, subsequent studies have examined whether these skills emerge earlier in development. In a cross-sectional examination of intentionality in typically developing infants, Bellagamba and Tomasello (1999) reported evidence of an emerging understanding of intentionality as early as 12 months. Meltzoff’s (1995) study procedures and conditions were used in this investigation, with the addition of an endstate condition (i.e. presenting the object to the infant as if the target action had been successfully completed). Bellagamba and Tomasello (1999) noted that 12-month-olds would reproduce full target actions when they saw them performed by the examiner, but infants would not reproduce target actions when they saw the examiner trying and failing to perform an action. This indicates that 12-month-olds may not share the ability of 18-month-olds to understand unsuccessful goal directed actions as intentional (Bellagamba & Tomasello, 1999). Nonetheless, this study provides suggestive evidence that the understanding of intentionality improves with age (Bellagamba & Tomasello, 1999).

This suggested improvement in intentionality as infants’ age is further supported by a longitudinal investigation of intentionality from 12-to-15-months (Bellagamba, Camaioni, & Colonnesi, 2006). In this study, infants produced more target actions at 15 months than they did at 12 months (Bellagamba et al., 2006). However, 15-month-olds also performed more approximations of the target action, which suggests a partial understanding of intentionality
(Bellagamba et al., 2006) that was not observed in 18-month-olds (Bellagamba & Tomasello, 1999; Meltzoff, 1995). Taken together, research on intentionality in children under 18 months suggests that the ability to interpret an unsuccessful goal directed action as intentional is still emerging between 12 and 15 months (Bellagamba & Tomasello, 1999; Bellagamba et al., 2006).

**Critiques of Meltzoff’s Paradigm.** Although the research conducted by Bellagamba and Tomasello (1999) and Bellagamba and colleagues (2006) has added support to the efficacy of Meltzoff’s (1995) paradigm, Huang, Heyes, and Charman (2002; 2005) questioned the effectiveness of the behavioral reenactment procedure for measuring intentionality. The criticisms proposed by Huang and colleagues (2002; 2005) are important to consider because from these studies it is still unclear if there are other skills influencing the ability of children to understand intentionality when using the behavioral reenactment procedure.

First, Huang and colleagues (2002) posed an important critique of Meltzoff’s paradigm about whether children performed the target action because they understand intentionality or if they performed the target action because the task leads them to produce the target action through expanded imitation. In order to test whether expanded imitation was the reason children performed the target action during the behavioral reenactment procedure, two new conditions: emulation learning and stimulus enhancement. In the emulation learning condition, the examiner would show the infant the startstate and endstate of the action, but not the action itself. In order to examine stimulus enhancement, Huang and colleagues (2002) used spatial contingency (e.g. the examiner presented the object to the infant with the target relevant pieces close together spatial).

This study found that there were no statistically significant differences in the amount of target actions produced between the target condition, the failed intention condition and the
emulation learning condition (Huang et al., 2002). This challenges previous findings and suggests that emulation learning may play a role in children’s performance on the behavioral reenactment procedure (Huang et al., 2002). It is possible that emulation learning occurred during the failed intention condition by providing infants with observational information that afforded target-relevant information about how to successfully manipulate the objects (Huang et al., 2002). Also, infants in the stimulus enhancement condition produced more target actions during the target administration than infants in the other three conditions. Thus, Huang and colleagues (2002) concluded that emulation learning and stimulus enhancement might be factors in children’s performances on the behavioral reenactment procedure because these two skills lead to the production of target actions (Huang et al., 2002); however, it is unclear from this study how they lead to the production of target actions.

Next, to further examine the theoretical and methodological foundations of Meltzoff’s (1995) behavioral reenactment paradigm, Huang, Heyes, and Charman (2005) investigated the role of intention reading, emulation learning, and mimicry in preschool children (31-to-41-month-olds). This investigation was driven by the hypothesis that if intention reading is what causes children to perform target actions after observing a failed intentional action then older children who see the failed intention should produce as many target actions as children who view the target action (Huang et al., 2005). However, if performing the target action depends on both intention reading and detection of affordances, then older children should produce more target actions when viewing the target action in full than when viewing the failed intention (Huang et al, 2005).

This study adds further support to the suggestive evidence that the understanding of intentionality improves with age (Bellagamba & Tomasello, 1999, Bellagamba et al., 2006;
Huang et al., 2005). A similar pattern of performance of target actions that was observed in 19-months olds emerged for 31-to-41-month-olds, such that children in the target condition produced more target actions then children in the other three conditions (i.e. failed intention, emulation learning, and adult manipulation condition; Huang et al., 2005).

Huang and colleagues’ (2002; 2005) findings challenge the previous conclusions that children in the failed intention condition produce the target action by inferring the examiners intentional action (Bellagamba et al., 2006; Bellagamba & Tomasello, 1999; Meltzoff, 1995). Huang and colleagues (2005) concluded that in certain situations, infants and preschoolers being to understand intentions, but there are social and non-social learning processes affecting how a child responds to the observed action. Specifically, three constructs appear to be active in a child’s response: intentional imitation, emulation, and mimicry. Although it is true that these skills do appear to be a factor in performance, it is not clear if they are factors in understanding intentionality or if they are just factors in understanding cause and effect because the method and findings of these studies do not seem to investigate all of these constructs. It may be more accurate to conclude that the findings presented in this study provide suggestive evidence that intentional imitation, emulation, and mimicry are active processes in understanding intentionality. More research will be needed to determine how these constructs influence infants and young children’s understanding of intentionality.

**Conclusions on Intentionality.** Based on previous research, it is clear there are many questions still to be answered about children’s understanding of intentionality. First, it is still unclear whether children are actually inferring others’ intentions or if they are responding to what they observe through emulation learning and affordance detecting during the behavioral reenactment procedure (Huang et al., 2002; 2005). It is also unclear how intentionality develops
in more diverse samples (i.e. children from different cultural and socioeconomic backgrounds, and children with developmental disabilities).

Conclusions

The research that has been described on intersubjectivity and intentionality in typically developing children indicates that primary and secondary intersubjectivity develop over the course of the first year (Cebula & Wishart, 2008; Meltzoff, 2007 Tager-Flusberg et al., 2006; Trevarthen & Aitken, 2001) and lead to the development of intentionality around 18 months (Bellagamba & Tomasello, 1999; Bellagamba et al., 2006; Meltzoff, 2007; 1995; Trevarthen, 1978; Trevarthen & Aitken, 2001). Understanding the developmental course of intersubjectivity in typically developing children provides the foundational knowledge needed to examine intentionality in children with WS and DS. However, to date, intentionality has yet to be examined in children with WS and DS. Therefore, research on intentionality in typically developing children can help to guide research on intentionality in children with WS and DS. In the next section, the relevant literature on the behavioral phenotypes and social cognitive abilities of individuals with WS and DS is reviewed.

Neurogenetic Disorders

Neurogenetic disorders are the result of genetic abnormalities from either single gene mutations (e.g. fragile X syndrome) or from having an entire chromosome or segments of a chromosome that are missing (e.g. Williams syndrome) or duplicated (e.g. Down syndrome; Tager-Flusberg, 2005). The presence of these genetic abnormalities can influence development, especially brain development, which has a cascading effect on numerous areas of functioning within the individual (Tager-Flusberg, 2005). The effects of a genetic abnormality can be direct, by predisposing the individual to be more likely to display particular behaviors and abilities than
individuals with developmental disabilities of unknown etiology (Dykens, 1995; Hodapp, 2004). However, the effects can also be indirect via an evocative genotype-phenotype interaction (Scarr & McCartney, 1983), which suggests that characteristics of the individual, such as specific etiology-related behaviors, may elicit certain reactions and response from others (Hodapp, 2004; Hodapp & DesJardin, 2002). For example, the sociable behaviors and facial expressions of children with DS may elicit more positive reactions from others (Hodapp, 2004; Hodapp & DesJardin, 2002). Further, as children with DS continue to receive these responses from their social partners they start producing these behaviors with more frequency, leading to the development of a sociable and positive personality profile in individuals with DS (Hodapp, 2004; Hodapp & DesJardin, 2002). In the last few decades, researchers have started to investigate the complex issue of how neurogenetic disorders influence behavior by identifying and examining the behavioral phenotypes associated with different neurogenetic disorders (Dykens, 1995; Hodapp, 2005; Hodapp & DesJardin, 2002).

**Behavioral Phenotypes**

Behavioral phenotypes are the measureable behavioral outcomes observed in individuals with neurogenetic disorders (Dykens, 1995; O’Brien, 1995). According to Dykens (1995), a behavioral phenotype is conceptualized as “the heightened probability or likelihood that people with a given syndrome will exhibit certain behavioral or developmental sequelae relative to those without the syndrome” (p. 523). Therefore, for a specific syndrome, there are behaviors that are more probable or “characteristic”, but these behaviors may not emerge for all individuals with that syndrome (Dykens, 1995; Hodapp, 2004; Hodapp & DesJardin, 2002). In addition, while certain behaviors may be associated with a specific syndrome, they may not be completely unique to that syndrome because there are behavioral overlaps between genetic syndromes (e.g.
sociability and friendliness in WS and DS; Dlits, Morries, & Leonard, 1990). Because this dissertation focused on the development of intentionality in children with WS and DS, and intentionality has yet to be examined in these populations, it is important to consider how the behavioral phenotypes associated with these syndromes may influence the ability to understand intentionality.

**Williams Syndrome**

Williams syndrome (WS) is one of the most widely researched neurogenetic disorders (Bellugi, Lichtenberger, Jones, Lai, & St. George, 2001) with an incidence rate of 1 in 7500 (Stromme, Bjornstad, & Ramstad, 2002). WS is the result of a microdeletion of 16 genes on chromosome 7, and in most of these cases (>98%), the same deletion is observable (Mervis & Klein-Tasman, 2000). WS leads to mild to moderate cognitive and developmental impairment that is characterized by peaks and valleys in ability and functioning (Bellugi & St. George, 2001). Individuals with WS have distinct craniofacial features (Mervis & Klein-Tasman, 2000), giving them a pixie-like or elfin appearance (Bellugi & St. George, 2001; Dykens et al., 2000). These craniofacial features include: a broad forehead, high prominent cheekbones, wide mouth, full lips, flat nasal bridge, and upturned nose (Dykens, Hodapp, & Finucane, 2000; Kaplan, Wang, Franck, 2001). Many individuals with WS experience physical problems such as a defect in the production of elastin, hypercalcemia, hypercusis, heart problems (e.g. arterial stenosis, hypertension), and musculoskeletal problems (Bellugi & St. George, 2001; Dykens et al., 2000; Kaplan et al., 2001). In the last few decades, researchers have focused on the examination of the specific behavioral phenotype associated with WS in order to identify areas of strength and challenge in this population (see Dykens, 2003; Martens, Wilson, & Reyten, 2008; Mervis & Klein-Tasman, 2000 for a review).
**WS Behavioral Phenotype.** Individuals with WS tend to have relative strengths in expressive language, communication, facial production and recognition skills, empathy, interpersonal skills, and auditory rote memory (Bellugi & St. George, 2001; Dykens, 2003; Dykens & Rosner, 1999; Mervis & Klein-Tasman, 2000). However, individuals with WS also tend to experience relative weaknesses in visuospatial construction (i.e. pattern construction), motor skills (Bellugi & St. George, 2001; Mervis & Klein-Tasman, 2000), and extreme weakness in visual spatial-processing (Bellugi & St. George, 2001; Dykens, 2003). There is a distinct social profile associated with WS, which is characterized by an overly friendly, highly sociable (termed hypersociability; Gosch & Pankau, 1997), highly approachable, highly gregarious, and people oriented personality style (Mervis & Klein-Tasman, 2000). Children and adults with WS are eager to interact with others and do not seem to experience stranger anxiety, leading them to indiscriminately approach people (Jarvinen-Pasley et al., 2008; Mervis & Klein-Tasman, 2000). This combination of strengths, weaknesses, and personality leads to the emergence of a unique social cognitive profile, which was the focus of this dissertation.

**Social Cognition Abilities.** Social cognition in children and adults with WS, like other domains of development in this population, is characterized by peaks and valleys in performance (Bellugi & St. George, 2001). Most of the research on social cognition in WS has focused on describing the distinctive social phenotype associated with WS in order to understand the neurocognitive mechanisms behind their social strengths and weakness (Tager-Flusberg & Plesa Skwerer, 2007). The results of this research have demonstrated widespread impairments in social cognition starting in infancy and early childhood in WS (Jarvinen-Pasley et al., 2008). The present review of social cognition focuses on joint attention, theory of mind, and
hypersociability in WS because these abilities are the most relevant to the study of intentionality in this population.

Joint Attention. A distinct social phenotype emerges during infancy in WS, including increased frequency of smiling behavior and an increased frequency of attending to the faces of others during infancy (Jarvinen-Pasley et al., 2008). Infants and toddlers with WS also have a strong attraction to people and prefer social stimuli to nonsocial stimuli. The combination of a strong interest in people and atypically intense looking behavior may contribute to difficulties in joint attention, which in turn may reduce the opportunity for infants with WS to learn from the environment (Laing, et al., 2002, Mervis et al., 2003). During turn-taking tasks, toddlers with WS have higher performance than MA-matched typically developing peers and are more interactive during social games, which suggests dyadic interaction strengths in this population (Laing et al., 2002). However during triadic interactions, toddlers with WS display fewer instances of initiating joint attention (referential/declarative pointing) and initiation requests (instrumental pointing) then MA-matched typically developing peers (Laing et al., 2002). During these interactions, children with WS quickly turn what should be a triadic interaction into a dyadic interaction, especially when a novel person is present (Doyle, Bellugi, Korenberg, & Graham, 2004), which may result from poor joint attention skills (Laing et al. 2002) or it may be related to an overall preference for dyadic interaction (Jarvinen-Pasley et al., 2008). Other studies have indicated that responses to joint attention appeared to be developing typically in young children with WS, while initiating joint attention skills appear to be delayed (Hepburn, Fidler, Hahn, & Philofsky, 2011). This split in joint attention abilities could disrupt the development of intersubjectivity, which in turn would influence later development of theory of mind (Hepburn et al., 2011). The combination of poor joint attention skills and a tendency to use
dyadic interactions even when triadic interactions are necessary may influence future difficulties in perspective taking and theory of mind, including the understanding of intentionality.

Placed within the context of research on intersubjectivity and joint attention in typically developing infants, it would appear that children with WS may be missing critical opportunities to develop the secondary intersubjectivity skills (i.e. person-person-object awareness) needed to develop the understanding of others’ minds. Also, difficulties in secondary intersubjectivity may also lead to difficulties in seeing people as intentional beings (Meltzoff & Moore, 1998) and in interpreting the behavior of others in terms of mental states (Tager-Flusberg, 2005). This provides suggestive evidence that children with WS may struggle with the understanding of intentionality because as infants and toddlers, they are lacking the foundational intersubjectivity skills needed to understand intentionality. Furthermore, if children with WS are struggling with these early social cognitive abilities, they may also experience difficulties in more complex social cognitive skills, such as theory of mind (Cebula & Wishart, 2008; Meltzoff, 2007; Tager-Flusberg et al., 2006; Trevarthen & Aitken, 2001).

Theory of Mind. Early in the pursuit of research on theory of mind in WS, researchers believed there was relative sparing of theory of mind because most individuals with WS would pass first order and higher order false belief tasks (Karmiloff-Smith, Klima, Bellugi, Grant, & Baron-Cohen, 1995; Tager-Flusberg & Sullivan, 1998). However, more recent research has called into question the notion of relative sparing in theory of mind. For example, studies examining first order false belief in children with WS have uncovered that children with WS perform no better on these tasks than matched comparison groups (i.e. children with Prader-Willi syndrome and children with idiopathic developmental disabilities; Tager-Flusberg, Sullivan, & Boshart, 1997; Tager-Flusberg & Sullivan, 2000; Plesa Skwerer & Tager-Flusberg, 2006). Also,
there appears to be no difference in the ability of individuals with WS to distinguish lies from jokes (Sullivan, Winner, & Tager-Flusberg, 2003) or in their ability to use trait information to attribute intentionality (Plesa Skwerer & Tager-Flusberg, 2006), which are important precursors to understanding theory of mind.

In fact it seems that individuals with WS actually have difficulty with higher-order false belief tasks (Tager-Flusberg, Boshart, & Baron-Cohen, 1998; Sullivan & Tager-Flusberg, 1999). Similarly, studies on mentalizing abilities (i.e. quickly attributing someone’s mental state based on their facial and vocal expressions) in children, adolescents, and adults with Williams syndrome suggests that there is no evidence for the relative sparing of these abilities (Plesa Skwerer, Verbalis, Schofield, Faja, & Tager-Flusberg, 2006; Tager-Flusberg et al., 1998; Tager-Flusberg & Sullivan, 2000). In one study, adults with WS performed worse on attributing mental states to others than their typically developing peers, but also performed better than developmentally matched adults with Prader-Willi syndrome (Tager-Flusberg et al., 1998). This study provided some of the preliminary evidence that there may not be relative sparing in theory of mind. Furthermore, this study suggests that instead of relative sparing there may be selective sparing in some of the social cognition skills required to attribute mental states (Tager-Flusberg et al., 1998). These studies suggest that individuals with WS have problems “making inferences to narratives to interpret mental state information” (Tager-Flusberg & Plesa Skwerer, 2007, p. 98).

Difficulty understanding and interpreting mental state information may be observed in individuals with WS because such information is usually inferred by interpreting social cues from faces, voices, and bodily movements, a combination of preserved and impaired abilities in this population (Gagliardi, Frigerio, Burt, Cazzaniga, Perrett, & Borgatti, 2003; Riby & Black,
2010; Tager-Flusberg & Plesa Skwerer, 2007; Tager-Flusberg, Plesa Skwerer, Faja, & Joseph, 2003; Tager-Flusberg et al., 1998; Plesa Skwerer et al., 2006; Udwin & Yule, 1991). Also, the perception that individuals with WS can tune into mental states may be more related to the emotions presented during the interaction than an actual understanding of the mental state behind those emotions (Dykens & Rosner, 1999; Gosch & Pankau, 1997; Hodapp & DesJardin, 2002; Jones et al., 2000; Plesa Skwerer et al., 2006). Individuals with WS may have difficulties interpreting mental states because they lack the social knowledge that is normally acquired through theory of mind (Davies, Udwin, & Howlin, 1998; Dykens & Rosner, 1999). A gap in understanding may lead individuals with WS to have impaired social judgment and difficulties in perspective taking, applying emotional information, making and maintain friendships, and other important social cognitive abilities (Davies et al., 1998; Dykens & Rosner, 1999; Jarvinen-Pasley et al., 2008; Tager-Flusberg & Sullivan, 2000).

Based on these studies and research on the social profile associated with WS, Tager-Flusberg and Sullivan (2000) proposed a componential view of theory of mind, delineating two processes that underlie the development of theory of mind: social-perceptual skills and social-cognitive skills (Tager-Flusberg & Sullivan, 2000). Social-perceptual skills are closely related to the affective system and involve making judgments based on the perceptual and social information available during a social interaction (Tager-Flusberg, 2005; Tager-Flusberg & Sullivan, 2000). Social-cognitive skills build on social-perceptual skills and involve the higher order thinking abilities that make up the traditional conceptualization of theory of mind (Tager-Flusberg, 2005; Tager-Flusberg & Sullivan, 2000). Together these skills allow for cognitive inferences to be made about the content of mental states by applying information from past interactions and events (Tager-Flusberg, 2005).
Using this model, Tager-Flusberg and Sullivan’s (2000) research on theory of mind in WS indicates that there is relative sparing of social-perceptual abilities in this population, while social-cognitive abilities are relatively weaker. Because existing research in this area has focused on older children with WS, it is unclear if a dissociation in social-perceptual and social-cognitive abilities emerges in infancy and early childhood. However, research on young children with WS provides suggestive evidence that this dissociation does begin in early childhood (Fidler et al., 2007; Laing et al., 2002). In order to explore the emergence of this dissociation, Hepburn, Fidler, Hahn, and Philofsky (2011) examined the social-perpetual and social-cognitive behaviors of young children with WS (under the age of 5), autism, and typically developing children. Results indicated that young children with WS demonstrated intact social-perceptual skills and emerging deficits in social-cognitive skills (Hepburn et al., 2011). What is still unclear from this study is whether there is a dissociation in social-perpetual and social-cognitive skills in young children with WS understanding of early mental states, like desires and intentions. Due to the affective nature of social-perceptual abilities it is important to consider how hypersociability in WS may influence the understanding of intentionality in WS.

**Hypersociability.** The combination of relative strengths in interpersonal skills and the distinct social profile associated with WS drives individuals with WS to socially engage with others (Hodapp & DesJardin, 2002; Jones et al., 2000; Mervis & Klein-Tasman, 2000). As previously mentioned, a strong interest in people emerges during infancy (Tager-Flusberg et al., 2006), with infants with WS attending to the faces of others with more frequency and smiling more (Jarvinen-Pasley et al., 2008). Infants and toddlers with WS spend more time looking at their mother and at strangers then infants and toddlers of the same chronological or developmental age (Mervis et al., 2003). Furthermore, older infants and toddlers with WS look
more intensely at strangers in a manner that is not observed in typically developing children (Mervis et al., 2003). Children with WS are described as good social partners who are very friendly, highly empathic, and able to tune into other people’s mental states because they seem to have special sensitivity to the emotional states of others (Dykens & Rosner, 1999; Gosch & Pankau, 1997; Hodapp & DesJardin, 2002; Jones et al., 2000; Plesa Skwerer et al., 2006).

Given their high levels of empathy, emotional responsivity, and attunement to others, it would be plausible to speculate that children with WS might show relative sparing or even strength in perspective taking. However, there is evidence that young children with WS do not use knowledge of their social partners’ emotions to guide their behavior (Fidler et al., 2007). Thus, even if children with WS imitate their social partners’ affect, for example dislike towards a type of food, they may still earnestly offer their social partner the disliked item (Fidler et al., 2007). This suggests that children with WS may have difficulties applying emotional information and taking the perspectives of others, which is a critical skill for understanding the mental states of others (Trevarthen, 1978; Trevarthen & Aitken, 2001).

**Present Study.** The first study in this dissertation was designed to test Tager-Flusberg and Sullivan (2000)’s hypothesis that there is a dissociation between social-perceptual abilities and social-cognitive abilities in children with WS. In order to understand the intentions of others, both social-perceptual and social-cognitive skills are needed. To examine the possible presence of a dissociation between these two domains, Meltzoff’s (1995) behavioral reenactment procedure was administered with and without experimenter affective cues (e.g. facial, vocal, and gestural cues). Based on past research on theory of mind (Hepburn et al., 2011; Plesa Skwerer et al., 2006; Tager-Flusberg et al., 1998; Tager-Flusberg & Sullivan, 2000) and emotion recognition (Dykens & Rosner, 1999; Fidler et al., 2007; Gosch & Pankau, 1997; Hodapp &
DesJardin, 2002; Jones et al., 2000; Laing et al., 2002; Plesa Skwerer et al., 2006), it is possible
that the presence of emotional cues will serve as a further distraction in understanding the
intentions of others leading children with WS to be more likely to misinterpret the social-
cognitive information they observe and instead focus on sharing in the emotional experience
with their social partner.

Down Syndrome

Down syndrome (DS) is the most identifiable and common chromosomal abnormality
associated with intellectual disability, with an incidence rate of 1 in every 732 (Canfield et al.,
2006). DS is caused by the presence of three copies of the 21st chromosome, resulting in three
distinct genotypes: trisomy 21, mosaicism, and translocation. Individuals with DS also have
distinct craniofacial features, which include an upward and outward slant to the eyes, wide nasal
bridge, and brachycephaly (Dennis, 1995). Other physical characteristics include small stature,
low muscle tone, short neck, hypotonia in infancy, and joint laxity (Dennis, 1995). About one-
third of individuals with DS are born with heart defects (Chapman & Hesketh, 2000; Dennis,
1995). Individuals with DS are also at an increased risk for other medical conditions such as
respiratory and hearing problems, childhood leukemia, Alzheimer’s disease, and thyroid
conditions (Chapman & Hesketh, 2000; Dennis, 1995). In addition to the investigation of
physical characteristic and health conditions associated with DS, researchers have also focused on
identifying the specific behavioral phenotype associated with DS in order to identify areas of
behavioral strength and challenge in individuals with DS (see Chapman & Hesketh, 2000; Fidler,
2005; 2006 for a review).

DS Behavioral Phenotype. Individuals with DS tend to show relative strengths in
visuospatial processing, visual short-term memory, social functioning (i.e. more empathy and
prosocial responses; Kasari, Freeman, & Bass, 2003), receptive language, nonverbal social communication, primary intersubjectivity, reciprocal turn taking, and other social initiations (Fidler, 2005; 2006; 2008; Hodapp & DesJardin, 2002). It has been noted that young children with DS show strengths in social relatedness (Fidler, 2005; Kasari et al., 1990; Ruskin et al., 1994), and have a preference for people over objects. However, during cognitively challenging tasks children with DS may overuse their social relatedness skills to compensate for weaknesses in other domains (Cebula & Wishart, 2008; Fidler, 2006). Individuals with DS also experience relative weaknesses in verbal processing, expressive language, instrumental communication, verbal short-term memory, emotion recognition, and some areas of motor development (Chapman & Hesketh, 2000; Fidler, 2005; 2006; Wishart, 2007). Also, researchers note a particular motivation profile such that individuals with DS engage in lower levels of task persistence and higher levels of off task behavior (Fidler, 2005). The domain of the DS behavioral phenotype that is most relevant to the development of intentionality in children with DS is the development of social cognition in this population.

**Social Cognition.** Social cognition in DS has not received as much research attention when compared to other areas of the DS behavioral phenotype, because historically it was thought that this domain was spared (Cebula & Wishart, 2008). Because individuals with DS are characterized as having intact social skills (Fidler, Most, Booth-LaForce, & Kelly, 2008; Gilmore, Campbell, & Cuskelly, 2003), it has been assumed that social understanding is also relatively intact (Cebula, Moore, & Wishart, 2010; Wishart, 2007). However, more recent research into the social cognitive profile associated with DS suggests that social cognition does not seem to be “playing the same supporting role” (p.45) in the overall development of children with DS as it does in typically developing children (Cebula et al., 2008) and, therefore, may
actually be compromised in this population (Wishart, 2007). Although the developmental sequence of social cognitive skills in DS follows a similar pattern to that observed in typically developing infants and children (Cebula et al., 2010; Cebula & Wishart, 2008), these skills are qualitatively different and take longer to develop in infants with DS, which may influence the development of more complex social cognitive skills in later development (Cebula & Wishart, 2008). This differentiation in the developmental of social cognition can be observed in the development of intersubjectivity in DS.

*Intersubjectivity.* Infants with DS achieve the developmental milestones associated with primary intersubjectivity (i.e. mutual gaze, joint attention, etc.; Fidler, 2006), but with noticeable differences from the typical trajectory. Mutual gaze in infants with DS is slow to emerge, but then it persists at a high level into the middle of the 1st year at a time when typical infants begin to shift their focus from people to the world around them (Cebula et al., 2010; Cebula & Wishart, 2008; Berger & Cunningham, 1981; Carvajal & Iglesias, 2000). By the middle of their first year, infants with DS show increased looking behavior (Crown, Feldstein, Jasnow, & Beebe, 1992; Gunn, Berry, & Andrews, 1982). This longer looking time may come at the expense of the development of other social cognition skills, such as dividing and switching attention, leading to difficulties in the development of joint attention (Cebula & Wishart, 2008; Legerstee & Weintraub, 1997). Once joint attention skills develop, infants with DS are more likely to be a passive participant, sharing attention to objects, rather than actively participating in the coordination of attention by pointing to objects (Legerstee & Weintraub, 1997). This suggests that the coordination of attention between people and objects progresses more slowly in infants with DS, leading to fewer instances of joint attention (Legerstee & Weintraub, 1997).

Conversely, other studies suggest that young children with DS have MA appropriate levels of
joint attention (Mundy, Sigman, Kasari, & Yirmiya, 1988; & Sigman & Ruskin, 1999) and levels of coordinated joint attention that is similar to MA-matched typically developing children (Kasari et al., 1995).

Generally, toddlers with DS appear to use pointing gestures competently and more frequently than typically developing children (Cebula et al., 2010, Cebula & Wishart, 2008), (Franco & Wishart, 1995). Because of expressive language delays, this may be an effective form of nonverbal communication for toddlers with DS (Cebula & Wishart, 2008). Even though toddlers with DS have strengths in pointing gestures, they also use fewer spontaneous requesting gestures than MA-matched peers (Cebula & Wishart, 2008; Fidler et al., 2005; Franco & Wishart, 1995; Mundy et al., 1988). However, the difficulties children with DS experience in nonverbal requesting are less pronounced during social games (e.g. tickle game) than they are in other games and interaction with toys (Fidler et al., 2005). Furthermore, children with DS have been found to be less engaged in situations created to promote requesting and commenting than they were in situations created to promote simple interaction suggesting language based joint attention may be a problem (Adamson et al., 2009). These strengths and weakness in nonverbal communication could influence the ability of children with DS to understand intentionality.

Conclusions on Intersubjectivity. Taken together, Cebula and Wishart (2010) conclude that developing intersubjectivity skills (i.e. joint attention, non-verbal requesting, imitation, and social referencing) support the ongoing development of person-person-object awareness, which in turn influences the theory of mind abilities in DS. The deficits related to theory of mind in DS are more subtle than in other syndromes with theory of mind deficits, like autism, which is why theory of mind may not have been identified as an area of weakness (Abbeduto et al., 2001;
Theory of Mind. Early research on theory of mind in children with DS suggested that first and second order false belief abilities are intact when matched to typically developing children on receptive language ability (Baron-Cohen, 1989; Baron-Cohen, Leslie, & Frith, 1985). However, Cebula and Wishart (2008) propose that these findings may be result of using receptive language ability instead of a measure of mental age when examining theory of mind because other studies on theory of mind in children and adults with DS have not replicated these results.

In one study of theory of mind in individuals with DS, autism, developmental disabilities, and typically developing children, results indicated that individuals with DS had similar performance to typically developing children on the value task (e.g. asking the child whether the examiner liked or disliked a food item after the experimenter displayed facial like or dislike for the food; Yirmiya et al., 1996). However, typically developing children performed better than children with DS on the fact task (e.g. having knowledge that a candy box was full while the experimenter did not know whether or not the box was full). This suggests that individuals with DS are more successful at easier theory of mind tasks. When compared to receptive language matched typically developing children, individuals with DS also appear to have difficulties with standard theory of mind tasks, failing 9 of 12 theory of mind tasks (Zelazo et al., 1996).

All of these studies have used more complex theory of mind tasks. To date there has been no research on early aspects of theory of mind, like desires and intentions. In order to fully conceptualize theory of mind abilities and social cognitive abilities in children with DS, it is necessary to examine how these early skills develop. Also, it has been suggested that theory of
mind abilities in DS are influenced by the individual’s executive function abilities, specifically rule use (Zelazo et al., 1996). This study found that the ability to understand theory of mind and the ability to understand rule use are related to one another, such that if individuals with DS failed one task they were more likely to fail the other task. Also, this understanding did not appear to be related to mental age; therefore, there is some other process that may be influencing understanding of these skills. However, this study does not take into account how other aspects of executive functioning, such as working memory, inhibition, and planning, may play a role in theory of mind abilities of individuals with DS.

Executive Function. Research on executive function in DS is limited and there is even less research on children with DS under the age of 10 (Lee, Fidler, Blakeley-Smith, Daunhauer, Robinson, & Hepburn, 2011). The EF domain that has received the greatest amount of research attention is working memory, which has been characterized as an area of weakness including pronounced weaknesses reported in the domain of verbal working memory (Jarrold & Braddeley, 2001; Kogan et al., 2009; Lanfranchi, Cornoldi, & Vianello, 2004; Lanfranchi, Jerman, Dal Pont, Alberti, & Vianelo, 2010; Lanfranchi, Jerman, & Vianello, 2009; Vicari, Carlesimo, & Caltagirone, 1995). This pattern of working memory deficits has also been observed in caregiver report of executive function in early childhood (Lee et al., 2011). Similarly, research on inhibition indicates that children, adolescents, and adults with DS displayed weaker performance than chronological and MA-matched comparison groups (Kopp, Krakow, & Johnson, 1983; Lanfranchi et al., 2010; Rowe, Lavender, & Turk, 2006). However, in early childhood inhibition has not been found to be an area of weakness (Lee et al., 2011).

During laboratory planning tasks, individuals with DS tend to have less efficient problem solving strategies than chronological and MA-matched comparison groups (Kasari & Freeman,
2001; Fidler, Hepburn, Mankin, & Rogers, 2005; Lanfranchi et al., 2010; Rowe et al., 2006). In addition, these deficits have been identified in caregiver reports of planning abilities (Lee et al., 2011). Individuals with DS performed worse on shifting tasks, especially when they had to sort by more than one rule (i.e. short by shape and color) or had to switch rules mid-task (i.e. short by color, now sort by shape; Lanfranchi et al., 2010; Rowe et al., 2006; Zelazo et al., 1996). Problems with shifting have been observed during social referencing situations, where children with DS have been shown to exhibit less shifting of attention between their social partner and the indented stimulus (Kasari et al., 1995).

Taken together, these studies provide suggestive evidence for deficits in all domains of executive function. These skills may influence the ability of children with DS to understand intentionality. First, in order to complete a successful target action, the child will have to plan how to correctly complete the action based on the intentional information they are presented with and inhibit any inclination to imitate the failed intention. Also, if children with DS experience difficulties with shifting in general, (Lanfranchi et al., 2010; Rowe et al., 2006; Zelazo et al., 1996) and in social referencing situations (Kasari et al., 1995), it is possible children with DS will have difficulties with intentionality. This may be especially true when using the behavioral reenactment procedure because during the task, the child is using social referencing to interpret what the examiner is trying to do. Finally, this whole process of interpreting the intentional action and performing the target action may depend on the child’s working memory abilities because the child will need to hold and apply the relevant pieces of information they have observed in order to successfully perform the target action.

**Present Study.** Based on past research on intersubjectivity and theory of mind in DS, it is unclear whether intentionality will be a strength, if it will be compromised, or if it will be
similar to MA-matched peers with DD. In the second study of this dissertation, task performance on Meltzoff’s (1995) behavioral reenactment procedure of children with DS was compared to children with DD. Using the behavioral reenactment procedure may also account for past criticisms of theory of mind tasks being too language intensive and, therefore, the reason children with DS do not perform well (Cebula & Wishart, 2008; Zelazo et al., 1996). However, the behavioral reenactment procedure has not been used to examine intentionality in children over 4 years of age. Therefore, it is possible that this task may not be developmentally appropriate for older children. Nonetheless, there is currently no measure of intentionality for older children. Finally, it is possible that the understanding of intentionality is also influenced by other social cognitive skills during early childhood (i.e. joint attention and affect sharing; Cebula & Wishart, 2010) and executive function skills in middle childhood (Kasari et al., 1995; Zelazo et al., 1996).
CHAPTER 3 – GENERAL METHOD

Research Design and Rationale

A quantitative research methods approach was implemented because the outcome variable in the present dissertation was performance on the intentionality task, which was measured by the proportion of actions completed during the task. A quantitative approach was also supported by previous research on intentionality when using Meltzoff’s (1995) behavioral reenactment procedure (Bellagamba, & Tomasello, 1999, Bellagamba et al., 2006; Huang et al., 2002; 2005; Meltzoff, 1995). Specifically, a comparative research approach was used because participants were not randomly assigned to groups, which means that cause and effect conclusions could not be drawn (Gliner, Morgan, & Leech, 2009). The general design classification was a between-group comparison (i.e. each participant is in only one condition). Study 1 compared the performance of children with WS who received affective cues to children with WS who did not receive affective cues. In addition, study 1 compared performance of children with WS who did not receive affective cues to MA-matched children with DD. For Study 2, we compared the performance of young children with DS to older children with DS. In addition, the performance of young children with DS was compared to the performance of MA-matched children with DD.

There was also an associational component to the present dissertation. Study 1 explored the relationship between performance on the intentionality task and the child’s chronological age (CA), verbal mental age (VMA), and nonverbal mental age (NVMA). For Study 2, the relationship between performance on the intentionality task and joint attention and affect sharing was examined in young children with DS. Study 2 also examined the relationship between performance on the intentionality task and executive function skills in older children with DS.
Finally, the relationship between performance on the intentionality task and the child’s CA, VMA, and NVMA was explored.

Validity

With regard to internal validity, there are some issues with this dissertation that need to be addressed. Both Study 1 and Study 2 compared the performance of children with WS and DS to children with developmental disabilities, which means that there are uncontrolled characteristics of the individuals that may threaten the internal validity of the study (Gliner et al., 2009). However, participants were matched as closely as possible on nonverbal mental age and verbal mental age when possible. Another aspect of internal validity that could be a potential problem was that some participants were tested in a lab and others were tested at their homes or at conferences. This method was necessary to achieve a sample of this size from these populations, but this could contribute to differences in performance.

With regards to external validity, convenience sampling and snowball sampling methods were used to obtain this sample, which could affect the generalizability of the findings (Gliner et al., 2009). As mentioned before, data from participants were collected in different locations. Therefore, it was not possible to control for other external factors like the time of day or the conditions of the room. This could affect whether these results can be generalized to real-life outcomes (Gliner, Morgan, & Leech, 2009). However, all of the tasks (i.e. intentionality task and the Early Social Communication Scales) were designed to instigate assessment within the context of playful activities. Using this approach could help the tasks, and the responses, to be less artificial because the child feels as though they are just playing with toys. Finally, all the developmental measures and the executive function measure used were assessments that have
been standardized and normed on typical and atypical populations with acceptable validity and reliability coefficients (see measures sections below for details).
CHAPTER 4 - STUDY 1 METHOD

Participants

Participants for Study 1 were 25 children with a confirmed diagnosis of WS. Within in the WS group, half of the participants received affective cues (i.e. facial, vocal, and gestural cues) during administration of the intentionality task and the other half did not. Children with WS who received affective cues (N = 13) had a mean CA of 46.31 months, a mean VMA of 28.20 months, and a mean NVMA of 22.59 months (See Table 4.1 for developmental and demographic information). Children with WS who did not receive affective cues (N = 12) had a mean CA of 44.55 months, a mean VMA of 32.77 months, and a mean NVMA of 26.77 months. Independent Samples t-tests were performed to examine differences between the children with WS who received affective cues and children with WS who did not receive affective cues on CA, MA, and NVMA. There were no statistically significant differences observed between the two groups on any of these dimensions.

Table 4.1

Participant Characteristics Study 1

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Williams Syndrome Affect (N=13)</th>
<th>Williams Syndrome No Affect (N=12)</th>
<th>Developmental Disabilities No Affect (N=12)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Child CA (in months)</td>
<td>46.31 17.56</td>
<td>44.55 11.71</td>
<td>39.91 12.10</td>
</tr>
<tr>
<td>Child Verbal MA (in months)</td>
<td>28.20  7.99</td>
<td>32.77  7.97</td>
<td>29.32  7.23</td>
</tr>
<tr>
<td>Child Nonverbal MA (in months)</td>
<td>22.59  5.74</td>
<td>26.77  6.57</td>
<td>29.60  8.13</td>
</tr>
<tr>
<td>Child gender (% male)</td>
<td>38.5</td>
<td>38.5</td>
<td>33.3</td>
</tr>
<tr>
<td>Child Ethnicity (%)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>84.6</td>
<td>100</td>
<td>58.4</td>
</tr>
<tr>
<td>Hispanic</td>
<td>7.7</td>
<td>-</td>
<td>8.3</td>
</tr>
<tr>
<td>More than 1 race</td>
<td>7.7</td>
<td>-</td>
<td>8.3</td>
</tr>
<tr>
<td>Unknown/Choose not to</td>
<td>-</td>
<td>-</td>
<td>25</td>
</tr>
</tbody>
</table>
In addition, the performances of the 12 children with WS who did not receive affective cues during the intentionality administration were compared to those of 12 children with developmental disabilities (DD; idiopathic DD = 3, questionable/unknown etiology = 2, speech language delay = 1, DD with sensory integration disorder = 1, Smith-Magenis syndrome = 1, velocardiofacial syndrome = 1, Sturge-Webber with Psychosis = 1, Cochayne syndrome = 1, Chromosome 18 = 1). Participants in these two groups were matched on CA, VMA, and NVMA (see Table 4.1). Children with DD had a mean CA of 39.91 months, a mean VMA of 29.32 months, and a mean NVMA of 29.60. Group differences were examined using Independent Samples t-tests. No statistically significant differences were observed between the two groups on the matching variables (i.e. CA, VMA, and NVMA).

Participants for both the WS and DD groups were recruited through the Autism and Developmental Disability Research Groups at the University of Colorado Medical School, JFK Partners Center for Excellence in Developmental Disabilities, parent support groups (Williams Syndrome Association; Mile High Down Syndrome Association; Rocky Mountain Chapter of the Williams Syndrome Association), and local agencies.

Data Collection

Before consent was obtained, consent forms were reviewed with each family and all parent questions regarding the study were answered. All of the examiners for these studies were Master’s or doctoral level researchers who had several years of experience working with children with developmental disabilities. During administration of the test battery, two examiners were present: one to administer the test and one to videotape the administration. Assessments were
counter-balanced and administered in at least two visits in order to reduce fatigue and maintain participant engagement and attention.

**Matching.** According to Chapman and Hesketh (2000), mental age matching (MA-matching) is a way to account for developmental level or what one has learned, and thus is a better dimension for matching than chronological age. MA-matching has been used in numerous other studies involving children with developmental disabilities (e.g. Fidler et al., 2005; 2007; 2008; Kasari et al., 1995; 2001; Kasari & Freeman, 2001; Klein-Tasman & Mervis, 2003; Mervis et al., 2003; Tager-Flusberg & Sullivan, 2000; Tager-Flusberg et al., 1998; Zelazo et al., 1996).

In the present study, children with WS who received affective cues were matched to children with WS who did not receive affective cues on non-verbal mental age (NVMA), verbal mental age (VMA), and chronological age (CA). Also, children with WS who did not receive affective cues were matched to children with DD on CA, VMA, and NVMA.

**Group Differences.** Independent samples *t*-tests were performed to compare performance of children with WS who received affective cues and children with WS who did not receive affective cues on the matching variables (i.e. CA, VMA, and NVMA). The results of the independent samples *t*-tests indicated that there were no statistically significant differences between the two WS groups. Similarly, independent samples *t*-tests were performed to compare the performance of the children with WS who did not receive affective cues to children with DD on the matching variables. The independent samples *t*-tests results indicated that there were no statistically significant differences between children with WS and children with DD. The non-significant results of these analyses enable the researchers to make comparisons between the two groups.
Measures

Child information sheet. Parents were asked to provide information about their age, education level, income, and the child’s ethnicity.

Mullen Scales of Early Learning (Mullen, 1995). The Mullen Scales of Early Learning (MSEL) is a standardized, observational test for children ages 3 to 60 months assessing cognitive abilities and motor skills. All of the items on the MSEL are performance-based and are designed to pose a challenge only in the skill being assessed. Scoring is based on T scores (M = 50) and age equivalent scores. Strong concurrent validly with other well-known developmental tests (e.g. Bayley Scales of Infant Development, Peabody Developmental Motor Scales, Birth to Three Scale) has been established. Also, content validity, construct validity, and predictive validity has been established for this measure (Mullen, 1995). Internal consistency coefficients range from .83 to .95, test-retest reliability coefficients ranged from .82 to .85, and interrater reliability coefficients range from .91 to .99.

Leiter International Performance Scale-Revised (Roid & Miller, 1995, 1997). The Leiter International Performance Scale-Revised (Leiter-R) is a standardized measure of nonverbal intelligence. In the present study, the Leiter-R Brief IQ Composite was used to measure nonverbal intelligence. The Brief IQ is constructed from the four subtests: Figure Ground, Form Completion, Sequential Order, and Repeated Patterns. The Leiter-R has been standardized on a national sample of almost 2,000 individuals from 2.0 to 20.11 years old and has established evidence for fairness across different ethnic and socioeconomic groups (Roid & Miller, 2002). The Leiter-R Brief IQ Composite demonstrated adequate concurrent validity correlating with the WISC-III Full Scale and Performance IQs (.85) and high test-retest reliability (upper .80s-.90s).
**Intentionality Task.** This task was adapted from Meltzoff’s (1995) behavioral reenactment procedure. The purpose of this task is to assess a child’s understanding of the intentions of others. Specifically, this task examined if children understand the intended action that an examiner is trying to complete. In the present study, nine sets of intentionality toys were used: prong and two (2) loops; cylinder, stick and beads; Plastic square with hole, post and ball; two (2) small plastic nesting cups; dumbbell; two (2) wooden blocks with holes, rope; two (2) nets and toy grasshopper; Winnie the Pooh doll and chair; lily pad, frog/fish and cotton ball. The order of presentation of the toys was randomized. All toys were kept hidden before they were brought to the table for demonstration. The toys were then returned to a container before the next toy was presented. There were six trials for this task: three administrations of the target condition and three administrations of the failed intention condition. These administrations were also randomized. For 27% of the administrations, there were only 2 administrations of the target action because of problems with the toys. In the target condition, the experimenter modeled the entire target action with the toy successfully. In the failed intention condition, the experimenter tried to do the target action, but was unsuccessful.

Before each of the trials, the child was presented with each toy for that trial for at least a 20-second baseline period, or until the child had a chance to explore the toy. This initial baseline period established that they would not spontaneously make the target actions with the toys on their own without the experimental model. In the event that the child spontaneously produced the action, the examiner performed a different target action. After this period, the examiner would take back the toy, saying, “Can I have a turn?” The experimenter then modeled the appropriate experimental condition with the toy, either the target condition or the failed intention condition. The action was repeated three times, in approximately 20-seconds, and then the child
was given the toy for a 20-second response period. Timing started from when the child touched the object. If, during the baseline period, the child demonstrated the target behavior, then an alternate behavior was presented.

**Emotional displays by experimenter.** To investigate how the presence of affective cues influenced performance on the intentionality task in WS, in half of the administrations the examiner did not show emotional displays (no facial, vocal or gestural displays) during the intentionality trials. For the target action administration the examiner showed facial displays of joy (wide smile) or surprise (eyes wide, slight smile), they said “ta da” or “yea”, and they clapped or showed surprise with their hands (hands out palms up). Also, for the action with the dumbbell, they made a motor noise while putting the pieces together. For the failed intention administration, the examiner showed signs of concentration or trying (furrowed brow, bite lip, frown), they said “shoot” or “ugh”, and for gestural display they would hang their head or slammed this fist/hand down.

**Coding.** Two coders (undergraduate research assistants) who were naïve to the hypotheses in this study coded the data (see Appendix 1 for coding scheme). Cohen’s kappa was calculated for 20% of the videos to establish interrater reliability (kappa = .81). The coders met with the Co-PI to reconcile any discrepancies or difficult administrations. The PI was asked for help in reconciliation if the discrepancies could not be solved between them. For each trial, they coded the action of the examiner and the action of the child. As per Bellagamba and colleagues (2006) and Huang and colleagues (2002; 2005), when the examiner performed the target action, there were three responses coded for the child (target action, no action, other action) and four responses coded for the child when the examiner performs the failed intention condition (target action, no action, other action, imitate the failed intention).
A target action was coded if the child successfully performed the target action as the examiner did or as the examiner intended to do on the failed intention administration (e.g. successfully putting the beads into the cup). For some children performing the target action was difficult because they were not strong enough or had poor motor skills. Therefore, trying was defined as actively trying to complete the action, such that the child would have completed the task if they were stronger or had better motor skills. An imitation of the failed intention was coded when the child imitated the examiner’s failure (e.g. missing the cup and letting the beads slide down the side of the cup). For coding purposes, an “other action” was defined as any action the child produced with the toys other than the target or imitation of the failed intention. Similarly, a code of “no action” meant that the child did not perform any actions with the toys (i.e. the child either did not touch the toys or just held them in their hands).

In addition, the first action performed by the children was coded, as well as all of the subsequent actions performed during the 20-second response time, as per Huang and colleagues (2002; 2005). By coding the first action and all the subsequent actions, it was possible to examine whether children used a trial and error process, or whether certain actions that preceded the completion of the target action (Huang et al., 2002; 2005). For each trial the coders also indicated whether or not the target action and imitation of the failed intention was ever performed during the trial. This allowed for an overall summary of performance on each trial.

For affective displays, the facial, vocal, and gestural displays of the children were coded. The facial displays that were coded were concentration/trying (frows brow, eyes drawn down), surprise (eyes wide, eye brows up, mouth open, maybe smile), happy/joy (smile), frown (corner of the lips turned downward), and pursing or biting lips. For vocalizations, coding focused on the vocalizations that would be made by the examiner (e.g. “look what I can do/can I have a
turn?”, “ta da”, “yay”, “ugh”, “shoot” and the motor sound). The gestural display codes were, clap, hands out and open (surprise), hanging their head, and clenching their fist. Also, for all of the affective displays there was the code of “other”. This code was used for any facial, vocal, or gestural displays the child produced. Whenever “other” was coded, the coders would also indicate what this other behavior was (e.g. facial displays of anger, saying “let me show you”, etc.).

Data Analysis

Following Huang and colleagues (2002; 2005) the proportion of target action, imitations of the intention, and other actions for the target and intention administrations were created. For example, if the child produced the target action on the three target administrations then they had a proportion of 1. Using this method allows for the use of data from participants who only received five administrations.

To investigate whether there was a dissociation between social-perceptual and social-cognitive abilities in children with WS who did and did not receive affective cues, relative likelihood statistics and the confidence intervals associated with those statistics were used. Relative likelihood ratios were used because these statistics may be more informative than the magnitude of the difference in the number of actions performed between the two groups of children with WS. Likelihood ratios are especially informative when the proportions are close to zero (Agresti & Finlay, 2009), which was the case in this study. Also, use of this statistic allows for an easy interpretation of the relative differences between the two groups because a relative likelihood statistic of 3, for example, means that one group was three times as likely to performed an action than the other group (Fidler et al., 2007). Relative likelihood statistics and the confidence intervals associated with those statistics were also used in this study to compare
performance on the intentionality task between children with WS and children with DD. The relationship between developmental status and performance on the intentionality task was examined using Spearman rho correlations because the proportion created for performance was not continuous and, therefore, it was treated as an ordinal variable.
CHAPTER 5 – STUDY 1 RESULTS

Research Question 1 – Dissociation between Social-Perceptual and Social-Cognitive Skills

It was hypothesized that children with WS who received affective cues would perform more target actions on the target administration than children with WS who did not receive affective cues. Indeed, 100% of children with WS who received affective cues performed the target action on the target administration, while 66.7% of children with WS who did not receive affective cues performed the target action on the target administration (see Table 5.1). This suggests that when observing the target action performed correctly, the presence of affective cues does improve performance (relative likelihood = 1.5, 95% CI = 1.01-2.24). It was further hypothesized that children with WS who received affective cues would imitate the failed intention more on the failed intention administration than children with WS who did not receive affective cues. Of children with WS who received affective cues, 76.9% imitated the failed intention, while 41.7% of children with WS who did not receive affective cues imitated the failed intention (relative likelihood = 1.85, 95% CI = .89 to 3.84). This suggests that children with WS who received affective cues were 85% more likely to imitate the failed intention as children with WS who did not receive affective cues. Finally, it was hypothesized that children with WS who did not receive affective cues would perform more target actions on the failed intention administration than children with WS who received affective cues. However, children with WS in both groups produced the same number of target actions on the failed intention administration.
Table 5.1

*Proportion of Performance of Children with WS during the Intentionality Task*

<table>
<thead>
<tr>
<th>Group</th>
<th>Action Performed</th>
<th>N</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Affective Cues</td>
<td>Proportion of Target Act on Target Administration</td>
<td>13</td>
<td>100</td>
</tr>
<tr>
<td></td>
<td>Proportion of Target Acts on Failed Intention Administration</td>
<td>9</td>
<td>69.2</td>
</tr>
<tr>
<td></td>
<td>Proportions of Imitations of the Failed Intention Administration</td>
<td>10</td>
<td>76.9</td>
</tr>
<tr>
<td></td>
<td>Proportion of Other Acts on Target Administration</td>
<td>13</td>
<td>100%</td>
</tr>
<tr>
<td></td>
<td>Proportion of Other Acts on Failed Intention Administration</td>
<td>13</td>
<td>100</td>
</tr>
<tr>
<td>No Affective Cues</td>
<td>Proportion of Target Act on Target Administration</td>
<td>8</td>
<td>66.7</td>
</tr>
<tr>
<td></td>
<td>Proportion of Target Acts on Failed Intention Administration</td>
<td>9</td>
<td>75</td>
</tr>
<tr>
<td></td>
<td>Proportions of Imitations of the Failed Intention Administration</td>
<td>5</td>
<td>41.7</td>
</tr>
<tr>
<td></td>
<td>Proportion of Other Acts on Target Administration</td>
<td>12</td>
<td>100</td>
</tr>
<tr>
<td></td>
<td>Proportion of Other Acts on Failed Intention Administration</td>
<td>12</td>
<td>100</td>
</tr>
</tbody>
</table>

As previously discussed, children with WS have demonstrated heightened levels of emotional responsivity during interactions with a social partner (Fidler et al., 2007). In the present investigation, we hypothesized that there would be an increased likelihood of affective responses for children with WS who received affective cues when compared to children with WS who did not receive affective cues. There were three forms of affect examined in the present study: facial, vocal, and gestural displays. Although both groups produced some form of affect on almost every administration regardless of whether it was the target or failed intention administration, children with WS who received affective cues produced more affective responses than children with WS who did not receive affective cues (Affect M = 17.08, SD = 5.45; No Affect M = 14.67, SD = 2.96; see Table 5.2). As hypothesized, the simple presence of affective
displays by the social partner led children with WS to produce more affect than when the social partner did not produce any affect. There was also some variation in the types of affect between groups. Most interestingly, 53.8% of children with WS who received affective cues performed gestural displays, while none of the children in the no affect group produced gestural displays of any kind. Children with WS who received affective cues were more likely to produce facial displays than children with WS who did not receive affective cues (Affect M = 11.77, SD = 3.35; No Affect M = 9.33, SD = 3.42). Also, children with WS who did not receive affective cues also performed slightly more vocal displays than children with WS who received affective cues (No Affect M = 5.33, SD = 2.87; Affect M = 4.77, SD = 3.65).

Table 5.2

Average Number of Facial, Vocal, and Gestural Displays

<table>
<thead>
<tr>
<th>Type of Affect</th>
<th>Williams Syndrome Affect (N=13)</th>
<th>Williams Syndrome No Affect (N=12)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Facial</td>
<td>M 11.77 SD 3.35</td>
<td>M 9.33 SD 3.42</td>
</tr>
<tr>
<td>Vocal</td>
<td>M 4.77 SD 3.65</td>
<td>M 5.33 SD 2.87</td>
</tr>
<tr>
<td>Gestural</td>
<td>.54 SD .52</td>
<td>0 SD 0</td>
</tr>
</tbody>
</table>

Children with WS who did not receive affective cues were also compared to an MA-matched group of children with DD in order to gain insight into whether there is a different pattern of task performance for children with WS. It was hypothesized that children with WS who did not receive affective cues would imitate the failed intention more than children in the DD comparison group. Interestingly, children with WS who did not receive affective cues and children with DD both imitated the failed intention 41.7% of the time (see Table 5.3).
Table 5.3

*Proportion of Performance of Children during the Intentionality Task*

<table>
<thead>
<tr>
<th>Group</th>
<th>Action Performed</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N</td>
<td>%</td>
<td></td>
</tr>
<tr>
<td>Williams Syndrome</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Target Act</td>
<td>8</td>
<td>66.7</td>
<td></td>
</tr>
<tr>
<td>on Target Administration</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Target Acts</td>
<td>9</td>
<td>75</td>
<td></td>
</tr>
<tr>
<td>on Failed Intention</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportions of Imitations</td>
<td>5</td>
<td>41.7</td>
<td></td>
</tr>
<tr>
<td>of the Failed Intention</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Other Acts</td>
<td>12</td>
<td>100</td>
<td></td>
</tr>
<tr>
<td>on Target Administration</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Other Acts</td>
<td>12</td>
<td>100</td>
<td></td>
</tr>
<tr>
<td>on Failed Intention</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Developmental Disabilities</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Target Act</td>
<td>11</td>
<td>91.7</td>
<td></td>
</tr>
<tr>
<td>on Target Administration</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Target Acts</td>
<td>9</td>
<td>75</td>
<td></td>
</tr>
<tr>
<td>on Failed Intention</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportions of Imitations</td>
<td>5</td>
<td>41.7</td>
<td></td>
</tr>
<tr>
<td>of the Failed Intention</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Other Acts</td>
<td>12</td>
<td>100</td>
<td></td>
</tr>
<tr>
<td>on Target Administration</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Other Acts</td>
<td>12</td>
<td>100</td>
<td></td>
</tr>
<tr>
<td>on Failed Intention</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

**Research Question 2 – Relationship between Developmental Status and Performance**

To examine if there was an association between developmental status (CA, VMA, and NVMA) and task performance (proportions of the target actions on the failed attempt administration, proportion of imitations of the failed intention on the failed intention administration, and proportion of other actions on both administrations), Spearman rho correlations were performed separately for both children with WS who received affective cues and those who did not receive affective cues (see Table 5.4 and 5.5). It was hypothesized that developmental status would be positively associated with performing the target action on the failed intention administration; however, no statistically significant correlations were observed for either group. For children with WS who did not receive affective cues, performing the target
action on the target administration was positively associated NVMA ($r_s(11) = .70, p < .05$) and VMA ($r_s(11) = .57, p = .07$). This indicates that as VMA and NVMA increases, children with WS who did not receive affective cues performed more target actions on the target administration and vice versa. This pattern of performance was not statistically significant for children with WS who received affective cues. Nonetheless, these correlation coefficients were in the same positive direction that was observed in children with WS who did not receive affective cues. Thus, it is unclear whether this pattern of performance is specific to children who did not receive affect or if there was not enough power to reach statistical significance in the affect group. Instead, CA was positively associated with performance on the target action on the target administration for children with WS who received affective cues ($r_s(13) = .70, p < .05$). This suggests that for children with WS who received affective cues, as CA increased, more target actions were performed on the target administration.

Table 5.4

*Relationship between Developmental Status and Performance – Affective Cues (N = 13)*

<table>
<thead>
<tr>
<th></th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Chronological Age</td>
<td>-</td>
<td>.75*</td>
<td>.70*</td>
<td>.64*</td>
<td>.14</td>
<td>.34</td>
<td>-.21</td>
<td>.00</td>
</tr>
<tr>
<td>2. Verbal Mental Age</td>
<td>-</td>
<td>-</td>
<td>.81**</td>
<td>.22</td>
<td>.18</td>
<td>.41</td>
<td>.14</td>
<td>-.24</td>
</tr>
<tr>
<td>3. Nonverbal Mental Age</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>.03</td>
<td>.24</td>
<td>.05</td>
<td>.13</td>
<td>.13</td>
</tr>
<tr>
<td>4. Proportion of Target Act on Target Administration</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-.28</td>
<td>.43</td>
<td>-.56*</td>
<td>-.20</td>
</tr>
<tr>
<td>5. Proportion of Target Acts on Failed Intention Administration</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-.29</td>
<td>.26</td>
<td>-.20</td>
</tr>
<tr>
<td>6. Proportions of Imitations of the Failed Intention</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-.49</td>
<td>-.35</td>
</tr>
<tr>
<td>7. Proportion of Other Acts on Target Administration</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>.12</td>
</tr>
<tr>
<td>8. Proportion of Other Acts on Failed Intention Administration</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
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</tr>
</tbody>
</table>

*p < .05, **p < .01*
Table 5.5

*Relationship between Developmental Status and Performance – No Affective Cues (N = 12)*

<table>
<thead>
<tr>
<th></th>
<th>1</th>
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<th>4</th>
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<th>6</th>
<th>7</th>
<th>8</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Chronological Age</td>
<td>-</td>
<td>.42</td>
<td>.36</td>
<td>.16</td>
<td>-.35</td>
<td>-.80**</td>
<td>-.28</td>
<td>-.17</td>
</tr>
<tr>
<td>2. Verbal Mental Age</td>
<td>-</td>
<td>-</td>
<td>.84**</td>
<td>.57#</td>
<td>.04</td>
<td>-.65*</td>
<td>-.37</td>
<td>-.32</td>
</tr>
<tr>
<td>3. Nonverbal Mental Age</td>
<td>-</td>
<td>-</td>
<td>.70*</td>
<td>-.02</td>
<td>-.65*</td>
<td>-.35</td>
<td>-.44</td>
<td></td>
</tr>
<tr>
<td>4. Proportion of Target Act on Target Administration</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>.33</td>
<td>-.41</td>
<td>-.40</td>
<td>-.80**</td>
<td></td>
</tr>
<tr>
<td>5. Proportion of Target Acts on Failed Intention Administration</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>.22</td>
<td>-.08</td>
<td>-.57</td>
</tr>
<tr>
<td>6. Proportions of Imitations of the Failed Intention</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>.22</td>
<td>.27</td>
</tr>
<tr>
<td>7. Proportion of Other Acts on Target Administration</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>.16</td>
</tr>
<tr>
<td>8. Proportion of Other Acts on Failed Intention Administration</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
</tbody>
</table>

*p < .10, *p < .05, **p < .01

It was also hypothesized that developmental status would be negatively correlated with imitating the failed intention. For children with WS who did not receive affective cues, imitating the failed intention was negatively associated with CA ($r_{s}(11) = -.80$, $p < .01$), VMA ($r_{s}(11) = -.65$, $p < .05$), and NVMA ($r_{s}(11) = -.65$, $p < .05$). This suggests that as developmental status increased, children who did not see affective cues performed fewer imitations of the failed intention. This pattern of performance was not seen in children with WS who received affective cues.
CHAPTER 6 – STUDY 1 DISCUSSION

Past research has indicated that there may be a dissociation between social-perceptual and social-cognitive abilities in WS (Hepburn et al., 2011; Tager-Flusberg & Sullivan, 2000). The present study provides suggestive evidence of a dissociation of these abilities in the understanding of intentionality in this population. We investigated this possible dissociation by examining differences in performance on an intentionality task, as measured by the behavioral reenactment procedure (Meltzoff, 1995), between two groups of children with WS: one that received affective cues and one that did not. In this study, children with WS who received affective cues imitated the failed intention more than children with WS who did not receive affective cues. This indicates that the presence of affective cues led children with WS to imitate the examiner more. This is further supported by the findings that children with WS who received affective cues also produced the target action more on the target administration than children with WS who did not receive affective cues. These findings suggest that the simple presence of affective cues led children with WS to imitate the examiner more during the behavioral reenactment procedure.

Taken together, introducing affect to the behavioral reenactment procedure may have changed the nature of the task for children with WS who were presented with affect cues. The presence of affective cues may have elicited a different set of responses for children with WS that are based within the affective system (i.e. the processing of emotional information separate from cognition or action; Izard, Kagan, & Zajonc, 1984) leading children with WS to imitate the examiner more. Furthermore, these findings suggest that the presence of affect may not add support to social cognitive processes. If this is the case, then the present study provides further evidence of a dissociation between social-perceptual and social-cognitive abilities in WS.
(Hepburn et al., 2011; Tager-Flusberg & Sullivan, 2000) because these abilities appear to be working independently during perspective taking tasks when affect is present. However, more research is needed to further explore and describe this dissociation in WS.

Nonetheless, these findings add to the growing literature on intersubjectivity in WS and suggest that emotional cues may not facilitate the understanding of intentionality. The present investigation further suggests that children with WS may be more skilled at sharing emotional experiences than interpreting what their social partner is intending to do. While attending to the emotional aspects of the situation, children with WS may overlook the information they need to understand the actions and the perspective of their social partner. In the present study, the presence of affect should have provided children with more information to use in order to interpret the failed intention of the examiner. However for children with WS, affect did not facilitate interpretation. Although the presence of affect facilitated higher rates of imitation in children with WS, this imitation did not seem to provide them with information to use to interpret their social partners mental state. This finding is further supported by previous research on perspective taking and emotional responsivity in children with WS (Fidler et al., 2007; Hepburn et al., 2011; Laing et al., 2002).

The ability to interpret the behavior of others in terms of mental states is critical for interacting in the social world (Cebula & Wishart, 2008; Meltzoff, 2007 Tager-Flusberg et al., 2006; Trevarthen & Aitken, 2001). To be able to interpret the mental state of a social partner, information is gathered from the emotional cues that are displayed during a social interaction (Meltzoff & Moore, 1998; Tager-Flusberg, 2005). In many situations, emotional cues are a means to an end during social interaction because emotional displays can help to facilitate the understanding of others mental states and guide our reactions and responses. However, this may
not be the case for individuals with WS. More research is needed to examine the role of emotion in interpreting the mental states of others’ in individuals with WS, to begin to understand how emotion is or is not supporting social cognition in this population. Examination of the relationship between emotion and the understanding of others’ mental states would also provide further insight into the proposed dissociation between social-perceptual and social-cognitive abilities in WS.

The present study also suggests that the understanding of intentionality does improve with developmental status for children with WS who did not receive affective cues, which is supported by previous research on the understanding of intentionality in typically developing children (Bellagamba & Tomasello, 1999, Bellagamba et al., 2006; Huang et al., 2005). However, this pattern of performance was not observed in children with WS who did receive affective cues. This finding suggests that the presence of affective cues also altered the pattern of performance for children with WS, providing further support that emotional displays serve as a distraction for children with WS during this type of intersubjectivity task.

When children with WS who did not receive affective cues were compared to children with DD, both groups performed the target action on the failed intention administration at the same rate. This suggests that children with WS are not more skilled than children with DD on this type of intersubjectivity tasks. One limitation of the present study was that there was no comparison between children with WS who received affective cues to children with DD who received affective cues. Comparing performance on the intentionality task between children with WS and DD who received affective cues would provide further insight into how the presence of affective cues influences the understanding of intentionality. Specifically, a comparison between children with WS and children with DD who received affective cues would
provide insight into whether the frequency of imitations on the failed intention administration was similar or different between the two groups.
CHAPTER 7 - STUDY 2 METHOD

Participants

Participants for this study were 40 children with a confirmed diagnosis of DS. There were two subgroups, young children with DS (24 to 57 months, N = 16) and older children with DS (59 to 136 months, N = 24). The 16 young children with DS were compared to 16 CA, VMA and NVMA matched young children with DD (Idiopathic DD = 3, questionable/unknown etiology = 3, Cochayne syndrome = 2, Chromosome 18 = 2, Angelman’s syndrome = 1, DD with sensory integration disorder = 1, Smith Magenis syndrome = 1, velocardiofacial syndrome = 1, Sturje-Webber with Psychosis = 1, speech language delays = 1). Young children with DS had a mean CA of 40.38 months, a mean VMA of 23.28 months, and a mean NVMA of 25.38 months (See Table 7.1 for developmental and demographic information). Young children with DD had a mean CA of 39.69 months, a mean VMA of 27.59 months, and a mean NVMA of 28.07 months. Independent Samples t-tests were performed to examine differences between the two groups on CA, VMA, and NVMA. No statistically significant differences were observed between young children with DS and children with DD on any of these dimensions. Older children with DS had a mean CA of 92.67 months, a mean VMA of 36.48 months, and a mean NVMA of 49.33 months.

Table 7.1

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Young Down Syndrome (N=16)</th>
<th>Young Developmental Disabilities (N= 16)</th>
<th>Older Down Syndrome (N=24)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Child CA (in months)</td>
<td>40.38 ± 10.60</td>
<td>39.69 ± 11.79</td>
<td>92.67 ± 21.17</td>
</tr>
<tr>
<td>Child Verbal MA (in months)</td>
<td>23.28 ± 7.38</td>
<td>27.59 ± 12.20</td>
<td>36.48 ± 18.93</td>
</tr>
</tbody>
</table>

Participant Characteristics Study 2

58
<p>| | | | | |</p>
<table>
<thead>
<tr>
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<th></th>
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<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Nonverbal MA (in months)</td>
<td>25.38</td>
<td>7.34</td>
<td>28.07</td>
<td>8.73</td>
</tr>
<tr>
<td>Child gender (% male)</td>
<td>64.7</td>
<td>4.8</td>
<td>75.0</td>
<td></td>
</tr>
<tr>
<td>Child Ethnicity (%)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>70.6</td>
<td>62.5</td>
<td>87.5</td>
<td></td>
</tr>
<tr>
<td>Hispanic</td>
<td>5.9</td>
<td>12.5</td>
<td>4.2</td>
<td></td>
</tr>
<tr>
<td>More than 1 race</td>
<td>11.8</td>
<td>6.3</td>
<td>-</td>
<td></td>
</tr>
<tr>
<td>Unknown/Choose not to respond</td>
<td>11.7</td>
<td>18.7</td>
<td>8.3</td>
<td></td>
</tr>
</tbody>
</table>

Participants were recruited through the Autism and Developmental Disability Research Groups at the University of Colorado Medical School, JFK Partners University Center for Excellence in Developmental Disabilities, parent support groups (Mile High Down Syndrome Association, Wyoming Down Syndrome Association), and local agencies.

**Data Collection**

The same data collection procedures that were used in Study 1 were also used in Study 2. As in Study 1, these tasks were counter-balanced and administered in two sessions in order to reduce participant fatigue and promote participant attention and engagement.

**Matching.** Young children with DS in were matched to children with DD on CA, VMA, and NVMA. This type of matching has been used in other studies involving children with developmental disabilities (e.g. Fidler et al., 2005; 2007; 2008; Kasari et al., 1995; 2001; Kasari & Freeman, 2001; Klein-Tasman & Mervis, 2003; Mervis et al., 2003; Tager-Flusberg & Sullivan, 2000; Tager-Flusberg et al., 1998; Zelazo et al., 1996).

**Group Differences.** As in Study 1, group differences were assessed by using an independent samples *t*-test to compare young children with DS to children with DD on the matching variable (i.e. CA, VMA, and NVMA). No statistically significant differences were
observed between young children with DS and children with DD, which enabled the researchers to make comparisons between the two groups.

**Measures**

The same measures used in Study 1 were also used in Study 2 (Child information sheet, Mullen Scales of Early Learning [Mullen, 1995], Leiter International Performance Scale-Revised [Roid & Miller, 1995; 1997], and the Intentionality Task). However in Study 2, there was no administration of affective display procedures for the intentionality task. Also, for 40% of the administrations of the intentionality task, there were only 2 administrations of the target actions because of problems with the toys. In addition to these measures, the Oral and Written Language Scales was administered as a measure of verbal mental age for children who only received the Leiter-R. Also, the Early Social Communication Scale was used to examine joint attention and affect sharing in young children with DS. Finally, the Behavior Rating Inventory of Executive Function – Preschool Version was used to examine executive function abilities in older children with DS. These measures are described below.

**Coding.** Two coders (undergraduate research assistants) who were unaware of the study’s research questions coded the data (see Appendix 1 for coding scheme). As in Study 1, Cohen’s *kappa* was calculated for 20% of the videos to establish interrater reliability (*kappa* = .84). The same coding processes for the intentionality task described in Study 1 were used in this study with the exception of coding for facial, vocal, and gestural behaviors.

**Oral and Written Language Scale.** The Oral and Written Language Scale (OWLS; Carrow-Woolfolk, 1996) is a standardized measure of expressive and receptive language. The OWLS has been normed on children and adolescents ages 3-21. Only the Listening Comprehension and Oral Expression scales were used in the present study. The Listening
Comprehension Scale is a receptive language measure that involves responding to prompts by indicating an answer with a nonverbal response (pointing to a picture on a page). The Oral Expression Scale involves answering a question, completing a sentence, or generating a sentence in response to a verbal or visual stimulus. The OWLS reports both high test-retest reliability (.73-.89) and high interrater reliability (.93-.99; Carrow-Woolfolk, 1995; 2008). It also has demonstrated both content and construct validity (Carrow-Woolfolk, 1995; 2008). The OWLS demonstrates high convergent and discriminant validity with other measures (i.e. Peabody Picture Vocabulary Test-Revised [Dunn & Dunn, 198], Test for Auditory Comprehension of Language Revised [Carrow-Woolfolk, 1985], Clinical Evaluation of Language Fundamentals-Revised [Semel, Wiig, & Secord, 1987], Kaufman Assessment Battery for Children [Kaufman & Kaufman, 1983], Wechsler Intelligence Scale for Children - Third Edition [Wechsler, 1991]; Carrow-Woolfolk, 1995; 2008).

**Early Social Communications Scale.** The Early Social Communication Scale (ESCS; Seibert, Hogan & Mundy, 1982), as described in Mundy, Sigman, and Kasari (1990), was designed to elicit examples of intentional communication (i.e., requests), joint attention behavior (e.g., following and initiating), and affect sharing. This consists of a series of social-communicative interactions with an experimenter and a set of toys in a semi-structured play interview. For the purpose of this study, only the joint attention (following joint attention and initiating joint attention) and affect sharing variables were used. Coding of behaviors was based on the frequency of occurrence of Requesting, Joint Attention, and Social Interaction behaviors, which involves classifying the function of the behavior, identifying who initiated the function, and identifying the behavior code (Mundy et al., 1990). Finally, the duration of the behaviors and the total length of time involved in each presentation were coded.
**Behavior Rating Inventory of Executive Function – Preschool Version.** The Behavior Rating Inventory of Executive Function – Preschool Version (BRIEF-P, Gioia, Espy, & Isquith, 2003) is a 63-item standardized rating scale assesses executive function in preschool aged children. The BRIEF-P assesses five different aspects of executive function (inhibit, shift, emotional control, working memory, and plan/organize) and can be used to calculate one composite score (Global Executive Composite) of overall executive function. Also, the five domains of executive function are combined to form three index scales: Inhibitory Self-Control, Flexibility, and Emergent Metacognition. The BRIEF-P’s normative sample includes reports from 460 parents and 302 teachers of children age 2 to 5 years. The BRIEF-P reports both high test-retest reliability for parent reports (.78-.90) and high internal consistency for (.80-.97; Gioia et al., 2003). It demonstrates convergent and discriminant validity with other measures of behavior and the authors used confirmatory factor analyses to validate their conceptual model of executive function.

For the present study, the BRIEF-P parent report form was used instead of the school-aged BRIEF parent report form. Recent research using the school-aged version of the BRIEF in children with DS suggests that not all the items were applicable to school-aged children with DS (Lee et al., 2011). However, the items on the BRIEF-P were found to be more appropriate for the developmental level of school-age children with DS (Lee et al., 2011). In order to calculate scores on the BRIEF-P, the child’s mental aged from the Leiter-R was used instead of their chronological age.

**Data Analysis**

As in Study 1, a proportion was made for the performance of target actions, imitations of the failed intention, and “other actions”. Spearman rho correlations were used to characterize the
magnitude of the relationship between task performance, joint attention, and affect sharing in young children with DS. Also, Spearman rho correlations were used to examine task performance and executive function abilities in older children with DS. Spearman rho was used because the proportion created for performance was treated as an ordinal variable. To examine if other domains of cognition predicted performance on the intentionality task in children with DS, multiple linear regression analyses were performed. For younger children with DS, the proportion of target actions on the failed intention administration and imitations of the failed intention were used as outcome variables and joint attention and affect sharing were used as predictor variables. For older children with DS, the same outcome variables were used, but the predictor variables were raw scores on the BRIEF-P.

To examine the age related differences of intentionality in children with DS, linear regression analyses were performed. For these regressions, the proportion of target actions on the failed intention administration and imitations of the failed intention were used as outcome variables, and developmental status (i.e. CA, VMA, and NVMA) was used as the predictor variable. In order to further describe performance on the intentionality task in children with DS, relative likelihood statistics and the confidence intervals associated with those statistics were used. This approach was used because it offers a simple interpretation of the relative differences in performance between the two groups (Fidler et al., 2007), which may be more informative than the magnitude of the difference when the proportions are close to zero (Agresti & Finlay, 2009). Also, use of this statistic allows for an easy interpretation of the relative differences between the two groups because a relative likelihood statistic of 2, for example, means that one group was twice as likely to perform an action than the other group (Fidler et al., 2007). Relative likelihood statistics, and the confidence intervals associated with those statistics, were also used
in this study to compare performance on the intentionality task between young children with DS and children with DD.
CHAPTER 8 – STUDY 2 RESULTS

Research Question 1 – Relationship between Performance and Other Domains of Cognition

For young children with DS, it was hypothesized that task performance on the intentionality task would be correlated with joint attention and affect sharing abilities. Table 8.1 presents the Spearman rho correlations for task performance, joint attention, and affect sharing. Performing an “other action” (e.g. any action the child performs other than the target action or imitation of the failed intention) on the failed intention administration was negatively correlated with both joint attention ($r_s(14) = - .69, p < .01$) and affect sharing ($r_s(14) = - .57, p < .05$). This suggests that for young children with DS, an increase in the performance of “other actions” during the intentionality task was related to lower levels of joint attention and affect sharing abilities and vice versa. Also, a marginally significant positive correlation was found between imitations of the failed intention and affect sharing ($r_s(14) = .49, p = .07$). This indicates that as affect sharing abilities increase, young children with DS perform more imitations of the intention and vice versa. No other statistically significant associations were found between task performance and joint attention or affect sharing.

Table 8.1

<table>
<thead>
<tr>
<th></th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Proportion of Target Act on Target Administration</td>
<td>-</td>
<td>.33</td>
<td>.34</td>
<td>-.35</td>
<td>-.17</td>
<td>.38</td>
<td>.30</td>
</tr>
<tr>
<td>2. Proportion of Target Acts on Failed Intention Administration</td>
<td>-</td>
<td>-</td>
<td>-.27</td>
<td>.12</td>
<td>.17</td>
<td>.32</td>
<td>.17</td>
</tr>
<tr>
<td>3. Proportions of Imitations of the Failed Intention</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>.18</td>
<td>-.63**</td>
<td>.38</td>
<td>.49#</td>
</tr>
<tr>
<td>4. Proportion of Other Acts on Target Administration</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-.16</td>
<td>.16</td>
<td>.42</td>
</tr>
<tr>
<td>5. Proportion of Other Acts on Failed Intention</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-.69**</td>
<td>-.57*</td>
</tr>
</tbody>
</table>
A multiple regression was conducted to examine if performance on the intentionality task (performance of the target action on the failed intention administration and imitations of the failed intention) could be predicted from joint attention and affect sharing abilities in young children with DS. The results of this analysis indicated that a significant model emerged for imitations of the failed intention, \( F(2,11) = 9.73, p < .01 \) (see Table 8.2). Approximately 64% of the variance in imitating the failed intention can be accounted for by the linear combination of joint attention and affect sharing. The regression equation for the prediction of imitating the failed intention is: 

\[
\text{Predicted Imitation of the Failed Intention} = -0.01 \times \text{Joint Attention} + 0.42 \times \text{Affect Sharing} - 0.11.
\]

Examination of the predictor variables showed a significant effect for affect sharing, such that young children with DS who had higher levels of affect sharing performed more imitations of the failed intention.

Table 8.2

Regression Analyses for Variables Predicting Performance on the Intentionality Task from Joint Attention and Affect Sharing (\( N=13 \))

<table>
<thead>
<tr>
<th>Variable</th>
<th>Target Actions</th>
<th>Imitations of the Failed Intention</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>( B )</td>
<td>( SE )</td>
</tr>
<tr>
<td>Joint Attention</td>
<td>.30</td>
<td>.19</td>
</tr>
<tr>
<td>Affect Sharing</td>
<td>-.24</td>
<td>.20</td>
</tr>
<tr>
<td>( R^2 )</td>
<td></td>
<td></td>
</tr>
<tr>
<td>( F(2,11) )</td>
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</tbody>
</table>

\#p < .10, *p < .05, **p < .01
For older children with DS, it was hypothesized that task performance on the intentionality task would be correlated with parent report of executive function abilities as measured by the BRIEF-P. BRIEF-P raw scores were used because using the raw scores provides a measure of actual EF skills, globally, not a measure of EF that is standardized for chronological or mental age. The only type of task performance that was associated with EF was performing “other actions” on the target administration (see Table 8.3). Specifically, there was a negative correlation between performing an action other than the target action on the target administration and both the Global Executive Composite \( (r, (19) = -.48, p < .05) \) and Emotional Control \( (r, (20) = -.45, p < .05) \). These findings suggest that children with DS who perform more “other actions” on the target administration have lower scores on the Global Executive Composite and Emotional Control as measured by the BRIEF-P and vice versa. Also, there were marginally significant negative correlations with performing any other action on the target administration and the Flexibility Index \( (r, (20) = -.43, p = .06) \), Inhibitory Self-Control \( (r, (20) = -.41, p = .07) \), and Emergent Metacognition \( (r, (19) = -.40, p = .09) \). This suggests that children with DS who perform more “other actions” on the target administration may have lower scores on the Flexibility Index, Inhibitory Self-Control, and Emergent Metacognition as measured by the BRIEF-P and vice versa.
Table 8.3

*Relationship between Performance and Executive Function (N =24)*

<table>
<thead>
<tr>
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<th>1</th>
<th>2</th>
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<th>4</th>
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<th>8</th>
<th>9</th>
<th>10</th>
<th>11</th>
<th>12</th>
<th>13</th>
<th>14</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Proportion of</td>
<td>-23</td>
<td>.45*</td>
<td>-.57**</td>
<td>-.31</td>
<td>.27</td>
<td>.34</td>
<td>.12</td>
<td>.11</td>
<td>.13</td>
<td>.27</td>
<td>-.08</td>
<td>.16</td>
<td>.14</td>
<td></td>
</tr>
<tr>
<td>Target Act on</td>
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<tr>
<td>Target</td>
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<td></td>
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<td></td>
</tr>
<tr>
<td>2. Proportion of</td>
<td>-</td>
<td>-08</td>
<td>-.23</td>
<td>-.57**</td>
<td>.11</td>
<td>.002</td>
<td>.10</td>
<td>.25</td>
<td>.11</td>
<td>-.11</td>
<td>.32</td>
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<td></td>
</tr>
<tr>
<td>Failed Intention</td>
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<td></td>
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<td>-.32</td>
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<td>6. Global</td>
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<td>.93**</td>
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<td>7. Emergent</td>
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<td>.67**</td>
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<td>-</td>
<td>.72**</td>
<td>.49*</td>
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<td>10. Inhibit</td>
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<td>.38</td>
<td>.49*</td>
<td>.09</td>
<td>.61**</td>
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<td>11. Shift</td>
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<td>.54*</td>
<td>.09</td>
<td>.70**</td>
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*p < .05, **p < .01
To further examine the relationship between EF and performance on the intentionality task, linear regressions were conducted. These analyses examined whether performance of target actions on the failed intention administration and imitations of the failed intention would be predicted by EF abilities as measured by the BRIEF-P. No statistically significant regression models were observed for the prediction of performance of target actions or imitation of the failed intention from EF abilities. Because statistically significant correlations were found between performance of other actions on the target actions and some of the domains of the BRIEF-P, linear regressions were performed to further explore the relationship between these skills and performance of other actions on the target administration. Statistically significant linear regression models did emerge for the prediction of performance of “other actions” on the target administration from the Global Executive Composite, Emotional Control, the Flexibility Index, Emergent Metacognition, and the Inhibitory Self-Control Index.

The statistically significant results of the linear regression predicting the performance of “other actions” from the Global Executive Composite are presented in Table 8.4, $F(1,18) = 7.17, p < .05$ (95% CI = -.02 to -.002). Global Executive Composite scores explained approximately 30% of the variance in performance of “other actions” on the target action. The regression equation for the prediction of performance of “other actions” on the target administration is:

$$Predicted \ Other \ Actions = -.01 \ Global \ Executive \ Composite + 1.66.$$  These findings suggest that older children with DS who have lower Global Executive Composite scores performed more “other actions” on the target administration.
Table 8.4

*Regression Analyses for Variables Predicting Performance on of other Action from the Global Executive Composite Task (N=19)*

<table>
<thead>
<tr>
<th>Variable</th>
<th>Other Action on Target Administration</th>
<th>B</th>
<th>SE B</th>
<th>β</th>
</tr>
</thead>
<tbody>
<tr>
<td>Global Executive</td>
<td></td>
<td>-.01</td>
<td>.01</td>
<td>-.55*</td>
</tr>
<tr>
<td>Composite R²</td>
<td></td>
<td>-.04</td>
<td>.01</td>
<td>-.61**</td>
</tr>
<tr>
<td>F(1,18)</td>
<td></td>
<td>7.17</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*p < .05 **p < .01

Similarly, the linear regression predicting the performance of “other actions” from Emotional Control was statistically significant, $F(1,18) = 10.55, p < .01$ (95% CI = -.07 to -.01; see Table 8.5). Emotional Control explained approximately 70% of the variance in performance of “other actions” on the target action. The regression equation for the prediction of performance of “other actions” on the target administration is: Predicted Other Actions = -.04 Emotional Control + 1.29. Results suggest that older children with DS who have lower levels of Emotional Control produced more “other actions” on the target administration.

Table 8.5

*Regression Analyses for Variables Predicting Performance on of Other Actions from Emotional Control (N=19)*

<table>
<thead>
<tr>
<th>Variable</th>
<th>Other Action on Target Administration</th>
<th>B</th>
<th>SE B</th>
<th>β</th>
</tr>
</thead>
<tbody>
<tr>
<td>Emotional Control</td>
<td></td>
<td>-.04</td>
<td>.01</td>
<td>-.61**</td>
</tr>
<tr>
<td>R²</td>
<td></td>
<td>.37</td>
<td></td>
<td></td>
</tr>
<tr>
<td>F(1,18)</td>
<td></td>
<td>10.55</td>
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</tr>
</tbody>
</table>

*p < .05 **p < .01
Table 8.6 presents the statistically significant linear regression predicting the performance of “other actions” from the Flexibility Index, $F(1,18) = 5.80, p < .01$ (95% CI = -.03 to -.002), which indicates that older children with DS who had lower scores on the Flexibility Index performed more “other actions” on the target administration. The Flexibility Index explained approximately 24% of the variance in performance of other actions on the target action. The regression equation for the prediction of performance of other actions on the target administration is: $\text{Predicted Other Actions} = -0.02 \times \text{Flexibility Index} + 1.23$.

Table 8.6

<table>
<thead>
<tr>
<th>Variable</th>
<th>Other Action on Target Administration</th>
<th>$B$</th>
<th>$SE$</th>
<th>$\beta$</th>
</tr>
</thead>
<tbody>
<tr>
<td>Flexibility Index</td>
<td></td>
<td>-.02</td>
<td>.01</td>
<td>-.49**</td>
</tr>
<tr>
<td>$R^2$</td>
<td></td>
<td>.24</td>
<td></td>
<td></td>
</tr>
<tr>
<td>$F(1,18)$</td>
<td></td>
<td>5.80</td>
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<td></td>
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</tbody>
</table>

* $p < .05$  ** $p < .01$

The linear regression predicting the performance of “other actions” from Emergent Metacognition was also statistically significant, $F(1,18) = 5.38, p < .01$ (95% CI = -.05 to -.002; see Table 8.7). Emergent Metacognition explained approximately 24% of the variance in performance of “other actions” on the target action. The regression equation for the prediction of performance of “other actions” on the target administration is: $\text{Predicted Other Actions} = -0.02 \times \text{Emergent Metacognition} + 1.90$. These findings indicated that older children with DS who have lower levels of Emergent Metacognition produced more “other actions” on the target administration.
Table 8.7

Regression Analyses for Variables Predicting Performance on of Other Actions from Emergent Metacognition (N=19)

<table>
<thead>
<tr>
<th>Variable</th>
<th>Other Action on Target Administration</th>
<th>B</th>
<th>SE B</th>
<th>β</th>
</tr>
</thead>
<tbody>
<tr>
<td>Emergent Metacognition</td>
<td></td>
<td>-.02</td>
<td>.01</td>
<td>-.49**</td>
</tr>
<tr>
<td>( R^2 )</td>
<td></td>
<td></td>
<td></td>
<td>.24</td>
</tr>
<tr>
<td>( F(1,18) )</td>
<td></td>
<td></td>
<td></td>
<td>5.38</td>
</tr>
</tbody>
</table>

*p < .05  **p < .01

Finally, the linear regression predicting the performance of other actions from the Inhibitory Self-Control Index was statistically significant, \( F(1,18) = 8.41, p < .01 \) (95% CI = -.03 to -.01, see Table 8.8). This suggests that older children with DS who have lower scores on the Inhibitory Self-Control Index performed more “other actions” on the target administration. The Inhibitory Self-Control Index explained approximately 32% of the variance in performance of “other actions” on the target action. The regression equation for the prediction of performance of “other actions” on the target administration is: \( \text{Predicted Other Actions} = -.02 \text{ Inhibitory Self-Control Index} + 1.41 \).

Table 8.8

Regression Analyses for Variables Predicting Performance on of Other Actions from Inhibitory Self-Control Index (N=19)

<table>
<thead>
<tr>
<th>Variable</th>
<th>Other Action on Target Administration</th>
<th>B</th>
<th>SE B</th>
<th>β</th>
</tr>
</thead>
<tbody>
<tr>
<td>Inhibitory Control Index</td>
<td></td>
<td>-.02</td>
<td>.01</td>
<td>-.56**</td>
</tr>
<tr>
<td>( R^2 )</td>
<td></td>
<td></td>
<td></td>
<td>.32</td>
</tr>
<tr>
<td>( F(1,18) )</td>
<td></td>
<td></td>
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<td>8.41</td>
</tr>
</tbody>
</table>

*p < .05  **p < .01
Research Question 2 – Trajectory of the Understanding Intentionality

To examine whether the understanding of intentionality in children with DS improves with developmental status (CA, VMA, and NVMA), multiple regressions were performed. Table 8.9 presents the results of these analyses. These analyses included developmental status as predictors and the proportion of target actions performed on the failed intention administration and imitations of the failed intention as outcome variables.

Table 8.9

<table>
<thead>
<tr>
<th>Variable</th>
<th>Target Actions on the Failed Intention administration</th>
<th>Imitations of the Failed Intention</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>B</td>
<td>SE B</td>
</tr>
<tr>
<td>Chronological Age</td>
<td>-.007</td>
<td>.003</td>
</tr>
<tr>
<td>Verbal Mental Age</td>
<td>-.002</td>
<td>.005</td>
</tr>
<tr>
<td>Nonverbal Mental Age</td>
<td>.02</td>
<td>.008</td>
</tr>
<tr>
<td>$R^2$</td>
<td>.22</td>
<td></td>
</tr>
<tr>
<td>$F(3, 33)$</td>
<td>3.15*</td>
<td></td>
</tr>
</tbody>
</table>

*p < .05 **p < .01

Of the two regression performed, a significant model emerged only for performing the target action on the failed intention administration, $F(3,33) = 3.15, p < .05$ (95% CI = -.03-.55). Approximately 22% of the variance of performance on target actions on the failed intention administration can be accounted for by the linear combination of developmental status. The regression equation for the prediction of performance of target actions on the failed intention administration is: $\text{Predicted Performance of Target Actions} = -.007 \text{ CA} - .002 \text{ VMA} + .02 \text{ NVMA} + .26$. Examination of the predictor variables showed a significant effect for NVMA, such that children with DS who had a higher NVMA performed more target actions on the failed intention administration.
administration. Also, a significant effect for CA was found, such that children with DS who were older, chronologically, performed less target actions on the failed intention administration.

To further examine the relative difference in performance on the intentionality task between young and older children with DS, relative likelihood ratios were calculated for each group. Results indicate that while both groups performed the target action often on both the target and failed intention administration (see Table 8.10), older children with DS imitated the failed intention more than young children with DS (relative likelihood = 1.33, 95% CI = .48 to 3.70).

Table 8.10

*Proportion of Performance of Children with DS during the Intentionality Task*

<table>
<thead>
<tr>
<th>Action Performed</th>
<th>Group</th>
<th>N</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Proportion of Target Act on Target Administration</td>
<td>Young Children with DS</td>
<td>15</td>
<td>93.8</td>
</tr>
<tr>
<td>Proportion of Target Acts on Failed Intention Administration</td>
<td></td>
<td>13</td>
<td>81.2</td>
</tr>
<tr>
<td>Proportions of Imitations of the Failed Intention</td>
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<td>4</td>
<td>25</td>
</tr>
<tr>
<td>Proportion of Other Acts on Target Administration</td>
<td></td>
<td>15</td>
<td>93.8</td>
</tr>
<tr>
<td>Proportion of Other Acts on Failed Intention Administration</td>
<td></td>
<td>16</td>
<td>100</td>
</tr>
<tr>
<td>Proportion of Target Act on Target Administration</td>
<td>Older Children with DS</td>
<td>22</td>
<td>91.7</td>
</tr>
<tr>
<td>Proportion of Target Acts on Failed Intention Administration</td>
<td></td>
<td>20</td>
<td>83.3</td>
</tr>
<tr>
<td>Proportions of Imitations of the Failed Intention</td>
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<td>8</td>
<td>33.3</td>
</tr>
<tr>
<td>Proportion of Other Acts on Target Administration</td>
<td></td>
<td>21</td>
<td>87.5</td>
</tr>
<tr>
<td>Proportion of Other Acts on Failed Intention Administration</td>
<td></td>
<td>24</td>
<td>100</td>
</tr>
</tbody>
</table>

It was further hypothesized that performance on the intentionality task would be similar between young children with DS and young children with DD. With regards to performance of
the target action on the target administration, almost all young children with DS and all of the children with DD performed the target action on the target administration. However, 81.2% of young children with DS performed the target action during the failed intention administration, compared to 68.8% of young children with DD (see Table 8.11). Therefore, young children with DS were 18% more likely to produce the target action on the failed intention administration (95% CI 0.79 to 1.77) than children with DD. Young children with DD, on the other hand, were 50% more likely to imitate the failed intention (relative likelihood = 1.5, 95% CI 0.52 to 4.32) than young children with DS.

Table 8.11

*Proportion of Performance of Children with DS and Children with DD during the Intentionality Task*

<table>
<thead>
<tr>
<th>Group</th>
<th>Action Performed</th>
<th>N</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Young Children with DS</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Target Act on Target Administration</td>
<td>15</td>
<td>93.8</td>
<td></td>
</tr>
<tr>
<td>Proportion of Target Acts on Failed Intention Administration</td>
<td>13</td>
<td>81.2</td>
<td></td>
</tr>
<tr>
<td>Proportions of Imitations of the Failed Intention</td>
<td>4</td>
<td>25</td>
<td></td>
</tr>
<tr>
<td>Proportion of Other Acts on Target Administration</td>
<td>15</td>
<td>93.8</td>
<td></td>
</tr>
<tr>
<td>Proportion of Other Acts on Failed Intention Administration</td>
<td>16</td>
<td>100</td>
<td></td>
</tr>
<tr>
<td><strong>Young Children with DD</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Proportion of Target Act on Target Administration</td>
<td>16</td>
<td>100</td>
<td></td>
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CHAPTER 9 – STUDY 2 DISCUSSION

The first goal of Study 2 was to examine the relationship between the understanding of intentionality and other cognitive abilities in children with DS (i.e. joint attention and affect sharing in young children with DS, and EF skills in older children with DS). With the exception of marginally significant relationship between imitating the failed intention and affect sharing in young children with DS, there were no statistically significant associations between these skills and performance of target actions on the failed intention administration or imitations of the failed intention in either age group. However, an interesting pattern of performance emerged for both age groups for performance of “other actions”.

In young children with DS, lower levels of joint attention and affect sharing were related to the production of more “other actions” on the failed intention administration. Past research on joint attention has produced two contrasting views of this ability. One line of research indicates that children with DS have difficulties with joint attention (Cebula & Wishart, 2008; Legerstee & Weintraub, 1997). The other line of research indicates that children with DS have appropriate levels of joint attention (Fidler et al., 2005; Kasari et al., 1995; Mundy et al., 1988; Sigman & Ruskin, 1999). Taken together, it is possible that some young children with DS have difficulties with joint attention, while other children with DS have developmentally appropriate levels of joint attention. If this were the case, it would explain the findings in the current study. In order to interpret the target action during the failed intention administration, children needed to use joint attention skills to gain the necessary information to successfully perform the action. Children with DS who have lower levels of joint attention may have missed the information needed to successfully perform the target action, which led them to perform “other actions” with the toys.
For older children with DS, performance of “other actions” on the target administration was associated with poorer performance on the Global Executive Composite, the Flexibility Index, the Inhibitory Self-Control Index, Emergent Metacognition, and Emotional Control. The Global Executive Composite is a summary measure of the child’s overall executive function ability. The other domains focus on the child’s ability to self-regulate, to self-manage tasks, to apply information from working memory to guide their actions, and the ability to shift, or move, between actions, behaviors, emotions, and response (Gioia et al., 2003). When examining the design of the intentionality task, it appears that all of these skills are necessary to successfully interpret the failed intentional information and perform the target action. As a result, older children with DS may have produce “other actions” because they knew they were supposed to do something with the toys, but did not have the skill set (i.e. planning, working, memory, inhibition, and shifting) to produce them. These findings add support to research by Zelazo and colleagues (1996), indicating that EF abilities influence theory of mind in individuals with DS.

The present study also examined the age-related changes in the understanding of intentionality in children with DS. The current findings suggest that the understanding of intentionality improves with developmental status in young children with DS. This finding is in line with past research on the understanding of intentionality in typically developing children (Bellagamba & Tomasello, 1999, Bellagamba et al., 2006; Huang et al., 2005). Older children with DS imitated the failed intention more than younger children with DS. This finding is particularly interesting considering that young children with DS also performed the target action more on the failed intention administration than MA-matched children with DD. There are two possible explanations for this finding. First, past research has suggested that cognitive development in children with DS is qualitatively different than developmentally matched peers,
in that it tends to slow and show greater variability in ability over the course of development (Carr, 1995; Dunst, 1990; Hodapp & Zigler, 1990; Hodapp, Evans, & Gray, 1999; Wishart, 1993; Wishart & Duffy, 1990). Therefore, these findings may provide suggestive evidence that children with DS have a better understanding of intentionality in early childhood than they do in middle childhood. However, it is also possible that the intentionality task used in this study was not developmentally appropriate for children with in the older DS group. The understanding of intentionality as not been examined in children over the age of 4 years. Therefore, it is possible that the behavioral reenactment procedure is not an effective measure of intentionality in older children. Future longitudinal research is needed to examine the development of intentionality in order to further characterize this pattern of performance. Also, future research should examine the appropriateness of the behavioral reenactment procedure in measuring intentionality in older children. This research may add insight into research on theory of mind in individuals with DS because the understanding of intentionality is an important precursor for theory of mind (Meltzoff, 2007; 1995). Nonetheless, the current study has taken an important first step in understanding the development of intentionality in children with DS.
CHAPTER 10 – GENERAL DISCUSSION

The purpose of this dissertation was to examine the understanding of intentionality in children with two neurogenetic disorders, WS and DS. Neurogenetic disorders can influence development by predisposing an individual to particular patterns of behaviors and abilities (i.e. directly; Dykens, 1995; Hodapp, 2004). In addition, the characteristics of an individual with a neurogenetic disorder may elicit specific reactions and responses from others that in turn lead to certain patterns of behaviors and abilities (i.e. indirectly; Hodapp, 2004; Hodapp & DesJardin, 2002; Scarr & McCartney, 1983).

To investigate the complex issues of how neurogenetic disorders influence behavior, researchers have focused on characterizing the behavioral phenotypes associated with neurogenetic disorders in order to identify areas of strength and challenge (Dykens, 1995; Hodapp, 2004; Hodapp & DesJardin, 2002). This dissertation examined whether WS and DS predispose children to specific areas of strength and challenge in the understanding of intentionality. Overall, the findings of this dissertation indicate that the understanding of intentionality improves with developmental status for both children with WS and DS, a finding that has also been observed in typically developing children (Bellagamba & Tomasello, 1999, Bellagamba et al., 2006; Huang et al., 2005). However, within each group, specific patterns of performance emerged on the intentionality task.

The findings from study 1 suggest that during perspective taking tasks, young children with WS are more likely to share in the emotional experience with their social partner than to try to interpret their social partner’s intentions. As such, this dissertation highlights the potential confounding influence of emotional displays when examining social cognitive abilities in children with WS (Fidler et al., 2007; Hepburn et al., 2011; Laing et al., 2002). Children with
WS who observed emotional displays during the administration of the behavioral reenactment procedure attended to the emotional information provided during the task rather than the social cognitive information they were presented with. Children with WS who did not receive the administration of affective cues, on the other hand, were more successful at interpreting a failed attempted using the social cognitive information they observed.

Difficulties in the understanding of intentionality may influence the subsequent social development in WS. The frequently reported challenges in the area of complex social cognitive skills in WS, such as perspective taking, attributing mental states, theory of mind, and social decision making (Davies et al., 1998; Dykens & Rosner, 1999; Jarvinen-Pasley et al., 2008; Tager-Flusberg & Sullivan, 2000; Tager-Flusberg et al., 1998) may be related to difficulties in understanding intentionality because viewing others as intentional beings is fundamental to these skills (Tager-Flusberg, 2005). Furthermore, it is possible that early deficits in understanding intentionality may explain part of why individuals with WS struggle with making and maintaining friends because friendships are supported by our ability to understand the motivations and intentions of a social partner (Davies et al., 1998; Enifeld et al., 1997; Karmiloff-Smith, Klima, Bellugi, Grant, & Baron-Cohen, 1995; Mervis & Kline-Tasman, 2000; Udwin & Yule, 1991).

It is also possible that the social phenotype associated with WS in early childhood and infancy has a cascading, or indirect, effect on the understanding of intentionality (Tager-Flusberg, 2005). As suggested by Fidler, Lunkenheimer, and Hahn (2011), there is a hypothesized model of cascading effects associated with the WS social profile, such that the hypersocial profile that emerges in childhood and adolescence (Jarvinen-Pasley et al., 2008), is related to intersubjective constraints during infancy. In the context of the present study, these
early intersubjective constraints may have a cascading effect on the understanding of intentionality. However, to date, there is no research that longitudinally explores the emergence of the behavioral phenotype associated with WS. In order to fully examine the potential cascading effects proposed in the present study, longitudinal studies are needed.

The influence of affective cues on performance on the intentionality task adds more information to the claims of Huang and colleagues (2002; 2005) that there are other processes influencing children’s performance on the behavioral reenactment procedure (Meltzoff, 1995). Huang and colleagues (2002; 2005) suggest that intentional imitation, emulation, and mimicry are active processes in performance on the intentionality task, which leads children to use imitation rather than intention reading to interpret the failed intentional actions of the examiner. However, imitation can either be purposeful in that it facilitates perspective taking during social cognitive situations (Meltzoff & Moore, 1998), or it can simply serve the purposes of emotion sharing (Trevarthen & Aitken, 2001). Therefore, it is possible that imitation could be used to support the understanding of intentionality.

The present study attempted to examine the influence of imitation in understanding intentionality by administering the intentionality task with and without affective cues to children with WS. Because of the social phenotype associated with WS and the tendency for imitation, if imitation was influencing performance on the intentionality task (Huang et al., 2002; 2005), then children with WS, especially those who received affective cues, should have produced more target actions. However, children with WS in both groups performed the target action on the failed intention with the same frequency. From the findings of this study, a more appropriate, and perhaps more important, question that arises about the role of imitation in the behavioral reenactment procedure is how imitation supports the understanding of others’ intentions.
Therefore, the present study does not support or reject the claims made by Huang and colleagues (2002; 2005), but adds another dimension of imitation to consider when using the behavioral reenactment procedure to investigate the understanding of intentionality.

Similarly, study 2 of this dissertation examines how other cognitive abilities influence the understanding of intentionality as measured by the behavioral reenactment procedure. In the present study, older children with DS who had poorer EF skills performed more “other actions” on the behavioral reenactment procedure. These findings add support to Huang and colleagues’ (2002; 2005) claims that there are other skills influencing the performance of children on the behavioral reenactment procedure. These findings further suggest that the phenotypic characteristics of children with DS may have a cascading effect on the understanding of intentionality. Specifically, it is possible that difficulties in joint attention (Cebula & Wishart, 2008; Legerstee & Weintraub, 1997) and EF (Lee et al., 2011) lead children with DS to miss the target relevant information during the behavioral reenactment procedure leading them to perform more “other actions”. However, more longitudinal research is needed to examine the influence of these skills on the understanding of intentionality.

Although the understanding of intentionality (as measured by the behavioral reenactment procedure) did improve with developmental status for children with DS, older children with DS produced more imitations of the failed intention than young children with DS. These findings may indicate that, like other cognitive abilities in DS, the development of the understanding of intentionality may be qualitatively different in older children with DS as compared to young children with DS (Carr, 1995; Dunst, 1990; Hodapp & Zigler, 1990; Hodapp et al., 1999; Wishart, 1993; Wishart & Duffy, 1990). However, it is also possible that these findings suggest that the behavioral reenactment procedure is not developmentally appropriate for older children.
It is notable, however, that in young children with WS who did not receive affective cues and young children with DS, correctly interpreting the target action during the failed intention administration was similar to the performance of MA-matched children with DD. Therefore, in children with WS and DS, the ability to view a social partner’s actions as intentional and correctly interpret their failed intention, does not appear to be an area of particular strength or weakness.

**Limitations**

There are some important limitations to the present study. The sample sizes in the present dissertation were small, and only the parameters of interests could only be estimated from these samples. Therefore, the findings presented in this dissertation should be interpreted with caution. Also, both of these samples were non-random samples, which means that inference is not entirely valid. For this reason, the present dissertation examined group differences using percentages and likelihood ratios. Furthermore, confidence intervals were included because they provide the range of possible values of the likelihood ratios. In addition, both of the current studies were cross-sectional; therefore, it was not possible to examine the trajectory of development of intentionality. In order to truly characterize the development of intentionality in WS and DS, longitudinal studies with larger sample sizes are necessary.

In the present dissertation, a comparison group of MA-matched children with DD was available only for the young DS and the WS no affect groups. In order to fully understand the influence of affective cues on the understanding of intentionality in WS, future studies need to incorporate a comparison group of MA-matched children with DD who received affective cues. Also, although the present study suggests that the understanding of intentionality in older children with DS, like other cognitive skills (Carr, 1995; Dunst, 1990; Hodapp & Zigler, 1990;
Hodapp, Evans, & Gray, 1999; Wishart, 1993; Wishart & Duffy, 1990), tends to slow and show
greater variability in ability over the course of development, it is not possible to know if this is
actually the case without a comparison group of MA-matched children with DD. Furthermore,
for both studies, the addition of a typically developing MA-matched comparison group may
provide insight into how the understanding of intentionality in WS and DS is similar or different
from typically developing children.

Conclusions

This dissertation is the first study to examine the development of intentionality in WS and
DS. Examining the understanding of intentionality in these populations is important to better
understand the development of intersubjectivity, theory of mind, and social cognition in WS and
DS. Study 1 adds further evidence to past research on perspective taking and theory of mind in
WS, indicating that there may be a dissociation between social-perceptual and social-cognitive
skills in this population during early social-emotional development. Specifically, it seems that
the presence of emotional cues during intersubjective tasks leads to an emotional response
instead of a response based on social cognition. Future studies should continue to explore this
dissociation in order to examine the potential cascading effect of these abilities on later
development. Study 2 provides suggestive evidence that the understanding of intentionality may
follow a qualitatively different trajectory in children with DS. In addition, overall findings from
this study suggest that, the understanding of intentionality improves with developmental status in
children with WS and DS. However, in order to characterize the development of intentionality
longitudinal research is needed.

Both of the studies in this dissertation highlight the potential cascading effects of
behavioral phenotypes on development in WS and DS. For children with WS, the unique social
profile (Gosch & Pankau, 1997; Mervis & Klein-Tasman, 2000) leads children with WS to focus on more on the social and emotional aspects of the intentionality task, especially when affect is present, rather than the social cognitive information they need to attend to in order to interpret the intentional actions of a social partner. For young children with DS, having appropriate levels of joint attention (Fidler et al., 2005; Kasari et al., 1995; Mundy et al., 1988; Sigman & Ruskin, 1999) leads children with DS to be more successful at understanding the intentions of others. It is possible that young children with DS who experience deficits in joint attention (Cebula & Wishart, 2008; Legerstee & Weintraub, 1997) miss the intentional information needed to interpret the intentions of others. In addition, recent research on EF in children with DS has provided evidence of deficits in all domains of EF (Lee et al., 2011) and that EF skills influence the understanding of theory of mind (Zelazo et al., 1996). Therefore, it seems that EF skills also influence the understanding of intentionality, which is supported by research EF and theory of mind in typically developing children (Carlson et al., 2004a; Carlson et al., 2004b; Hughes, & Ensor, 2007; Rakoczy, 2010). More research is needed to examine the relationship between EF and theory of mind in DS, including early theory of mind abilities such as intentionality and perspective taking. Further characterizing social cognition in WS and DS will help to identify areas for targeted intervention to prevent the possible cascading effects of difficulties in social cognition on other aspects of development.
REFERENCES


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Intentionality Task Coding Scheme

Adapted from Meltzoff (1995)

**Conditions**

**Target condition:** the experimenter models the entire target action with the toy successfully.

**Failed Intention condition:** the experimenter tries to do the target action, but is unsuccessful. They fail to perform the action.

**Directions** –
1. Circle the objects being used
2. Make sure the child does not produce the target act during play time – if so make a note under the trial and whether or not the made sure to do a different action
3. Circle whether it was the target act or failed intention
4. Code (see definitions below)
   a. **Target** = target action, no action, other action
      i. For some children performing the target action was difficult because they are not strong enough to perform the action or they have poor motor skills. **Trying** is defined as actively trying to complete the action. This means that the child would have completed the task if they were stronger or had better motor skills
   b. **Failed Intention** = target action, no action, other action, imitate failed intention
      i. What the first action was, and if it was the target act or failed intention
      ii. If the target act or failed intention was completed in 20 seconds
   c. **No action** = they do nothing with the objects (just holding them is no action)
   d. **Other action** = any other action then the ones listed, this has to be an action that they do with the items!
   e. **Child Affective Displays**
      i. **Facial**
         1. Concentration/trying - Furrows brow
         2. surprise – eyes wide, eye brows up, mouth open (maybe smile)
         3. happy/joy – smile
         4. frown—corner of the lips turned downward
         5. purse or bite lips
         6. Other –please indicate
      ii. **Vocal**
         1. Look what I can do, can I have a turn, let me try, any variation
         2. Ta da
         3. Yay
         4. Ugh
         5. Shoot
         6. Motor sound
         7. Other – please indicate
      iii. **Gestural**
         1. Clap
2. Hands out and open
3. Hangs head
4. clenches fist
5. other

5. for actions in 20 sec circle all that you see in that time, once trail is done circle if they produced the target act or failed attempt at any time during the admin even after the 20 sec response time

6. circle all child behaviors seen during the admin, make sure that when you circle other you write what it is.

7. Definitions

   a. Cylinder, beads and stick
      i. Target Action (for examiner and child) –
         1. gather up beads and puts in cylinder, it is ok if they hit the side of the cup on the way in
         2. **Alternate 1**: Hold the beads so they hang above the table and hit them with the stick so they swing like a pendulum.
         3. **Alternate 2**: Place cup upside down on the table and then place the stick across the top forming a “T” shape
      ii. Failed attempt (for examiner) -
         1. *Attempt to deposit the necklace in the container, but miss. Make three attempts, dropping the beads on three separate sides of the container.
         2. **Alternate 1**: Attempt to hit the beads with the stick but miss – missing in front (too short), under, and over.
         3. **Alternate 2**: Place the cup upside down on the table. Attempt to place the stick across the top of the cup to form a “T’” but have the stick roll of the top.
      iii. Imitation by child
         1. gather up the beads and dangle them like the examiner so that they hit the rim and fall down the side of the cylinder
         2. **Alternate 1**: miss hitting the beads in the same manner and order as the examiner
         3. **Alternate 2**: Place the cup upside down on the table. Attempt to place the stick across the top of the cup to form a “T”, push the stick roll of the top
      iv. Failure to produce target act by child
         1. Performing an act other than the failed attempt or target action

   b. Prong and loops
      i. Target Action (for examiner and child)
         1. raise the nylon loop up to the prong and drape it over it.
         2. **Alternate 1**: Drape the loop over your hand like a bracelet.
         3. **Alternate 2**: Drape the loop over your right ear.
      ii. Failed attempt (for examiner) -
         1. try but fail to place the loop over the prong, each time releasing the loop so that it falls to the table. First, the loop is released too far to
the left, then too far to the right and finally too low, so that if falls
to the table directly below the prong.

2. **Alternate 1:** Attempt to drape the loop over your hand like a
bracelet. This should be done three times – missing to the right,
left, and under.

3. **Alternate 2:** Attempt to drape the loop over your right ear but miss
and let the loop fall.

iii. Imitation by child

1. try to place the loop over the prong, each time they release it
similarly to the examiner so that it falls to the table. First, the loop
is released too far to the left, then too far to the right and finally too
low, so that if falls to the table directly below the prong.

2. **Alternate 1:** Attempt to drape the loop over their hand like a
bracelet. missing to the right, left, and under in the same way and
order of the examiner.

3. **Alternate 2:** Attempt to drape the loop over their right ear but miss
and let the loop fall. Either the same ear as the examiner or as a
mirror image of the examiner

iv. Failure to produce target act by child

1. Performing an act other than the failed attempt or target action
c. Ring, ball, and post

i. **Target Action** (for examiner and child)

1. Pick up the plastic ring and put it over the post

2. **Alternate 1:** Place the stand on its side and place the ring over the
post.

3. **Alternate 2:** Pull the ball part.

ii. **Failed attempt** (for examiner)-

1. Pick up the plastic ring and attempt to put it on the dowel. It should
not align properly. The first attempt should undershoot the dowel
and remain on the left, the second time it overshoots the dowel to
the right, and the third time the hole is spatially in front of the
dowel.

2. **Alternate 1:** Place the stand on its side and attempt to place the
ring on the post. The ring should fall to the side of the post. Make
three attempts.

3. **Alternate 2:** Attempt to pull the ball apart but be unable to do so.
Your hand should slip off both sides of the ball.

iii. Imitation by child

1. put the ring to the same location that the examiner did and miss the
post

2. **Alternate 1:** place stand on its side either on the same side as the
examiner or as a mirror image then make the ring fall to the side of
the post

3. **Alternate 2:** hold the ball and act like they are pulling hard letting
their hands slip off both sides

iv. Failure to produce target act by child
1. Performing an act other than the failed attempt or target action
d. Nesting cups
   i. Target Action (for examiner and child)
      1. Place small cup on top of larger cup.
      2. Alternate 1: Nest the cups.
      3. Alternate 2: Place the smaller cup on the table upside-down. then place the larger cup on top of the smaller one right side up.
   ii. Failed attempt (for examiner)
      1. Attempt to place the smaller cup on top of the larger cup but each time it should slide off the edge of the cup. Three tries should be given.
      2. Alternate 1: Attempt to nest the cups but each attempt should fail because the cup falls off to one side. Three attempts should be made.
      3. Alternate 2: Place the smaller cup on the table upside down. Attempt to place the larger cup, right-side-up, on top of the smaller cup but it should slip off.
   iii. Imitation by child
      1. Place the smaller cup on top of the larger cup in a similar manner to the examiner, make it fall off the edge of the cup.
      2. Alternate 1: Start to nest the cups but purposefully make the cup fall off to one side.
      3. Alternate 2: Place the smaller cup on the table upside down. Starts to place the larger cup, right-side-up, on top of the smaller cup but make it slip off.
   iv. Failure to produce target act by child
      1. Performing an act other than the failed attempt or target action
e. Beads and string
   i. Target Action (for examiner and child)
      1. String beads on the string.
      2. Alternate 1: Stack beads so that they form an inverted “T.”
      3. Alternate 2: Place one block on its side. Place the second block, also in its side, on top of the first block.
   ii. Failed attempt (for examiner)
      1. Attempt to string one of the beads but have the rope miss and hit to the side of the hole. Three attempts should be given each one missing to a different side of the hole.
      2. Alternate 1: Attempt to form an inverted “T” but have difficulty getting the stem of the “T” to remain standing on the base.
      3. Alternate 2: Place one block on its side and then attempt to place the other block, also on its side on top of the first block. It should fall off.
   iii. Imitation by child
      1. Start to string one of the beads but force the rope to miss and hit to the side of the hole or direct the rope to the side of the hole instead of into the hole.
2. Alternate 1: form an inverted “T” but do not place the stem of the “T” the right way to remain standing on the base.

3. Alternate 2: Place one block on its side and then attempt to place the other block, also on its side on top of the first block and force or push it so it falls off

iv. Failure to produce target act by child

4. Performing an act other than the failed attempt or target action

f. Dumbbell

i. Target Action (for examiner and child)
   1. Pull outward with a very definite movement so that the toy comes apart into two halves.
   2. Alternate 1: Place the two halves on top of each other.
   3. Alternate 2: Stand the dumbbell on one of its ends.

ii. Failed attempt (for examiner)
   1. Attempt to pull the ends outward but the experimenter’s hand should slip off the end of the dumbbell. Alternate side from left to right to left.
   2. Alternate 1: Attempt to stack the two halves of the dumbbell but fail to do so having one block slip off of the other. Do this three times.
   3. Alternate 2: Attempt to slide the dumbbell on one end but it should fall over.

iii. Imitation by child
   1. Pull the ends outward but take hands off so they look like they slip off the end of the dumbbell. Do hands in the same order as examiner or as mirror image of examiner
   2. Alternate 1: start to stack the two halves of the dumbbell but force or push one block to slip off of the other.
   3. Alternate 2: to slide the dumbbell on one end but it should fall over. Child should set it up like the examiner or the mirror image of the examiner

iv. Failure to produce target act by child
   1. Performing an act other than the failed attempt or target action

h. Grasshopper and nets

i. Target Action (for examiner and child)
1. Place the grasshopper in one of the nets and move him back and forth between the 2 nets in the air

ii. Failed attempt (for examiner)
   1. Place the grasshopper in one of the nets and move him back and forth between the 2 nets in the air, but miss when moving from one net to the other

iii. Imitation by child
   1. takes the net with the grasshopper and deliberately tries to miss putting the grasshopper into the other net; throws grasshopper on the ground completely

iv. Failure to produce target act by child
   1. Performing an act other than the failed attempt or target action

i. Pooh and chair
   i. Target Action (for examiner and child)
      1. Sit Winnie the Pooh in the chair
      2. Alternative 1: Stand Winnie the Pooh on the chair
   ii. Failed attempt (for examiner)-
      1. Try to sit Pooh on the chair but he falls off
      2. Alternative 1: Try to stand Pooh on the chair but he falls off
      3. Alternative 2: Hook chair on Pooh’s ear, it falls off
   iii. Imitation by child
      1. Try to sit Pooh on the chair but push or take him off
      2. Alternative 1: Try to stand Pooh on the chair but push or take him off
      3. Alternative 2: Hook chair on Pooh’s ear, push or take it off
   iv. Failure to produce target act by child
      1. Performing an act other than the failed attempt or target action
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