DISSERTATION

FOUNDATIONS OF EARLY PLANNING IN DOWN SYNDROME

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ABSTRACT

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Goal-directed behavior, or planning is critical for academic and daily outcomes, and an area of distinct challenge in Down syndrome. This study examined early foundations of object-related planning in toddlers (N=38) with Down syndrome. Motor abilities, visual attention, and motor cognition were tested as predictors of two planning outcomes in DS: object-related problem solving and functional object use. In addition, a potential developmental cascade from motor abilities to object-related problem solving was also tested. Results revealed that motor abilities are an important developmental foundation for both types of object-related planning outcomes. Results also revealed differences in the contribution of visual attention and motor cognition to object-related planning outcomes. Findings also provided support for a potential developmental cascade between motor abilities and planning outcomes. Collective results from this study contribute to the understanding of early development within Down syndrome, and therefore provide implications for the development of early, targeted intervention.

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CHAPTER ONE

Introduction

Down syndrome (DS) is the most common neurogenetic syndrome linked to intellectual disability, occurring approximately 1 in every 691 live births (Parker et al., 2010). There has been a 31% increase in the prevalence of DS between 1979 and 2002 (Shin et al., 2009), and a growing population of individuals with DS will continue to pose new challenges for the development of effective intervention, education, and services for this population. Importantly, gaps still remain in the fundamental scientific understanding of early development in DS, which currently limits the developmental impact of early intervention. Although researchers have characterized aspects of the DS behavioral phenotype in older individuals (see Daunhauer & Fidler, 2011 for review), including areas of relative developmental strength and challenge, critical gaps remain in our understanding of the early emergence of this profile and its contributing mechanisms.

Generally, the DS behavioral phenotype is characterized by relative strengths, or mental-age appropriate performance, in the areas of receptive vocabulary and socio-emotional functioning, and relative challenges in the areas of spatial memory, verbal working memory and verbal/auditory processing, expressive language, and aspects of motor development (Abbeduto, Warren, & Conners, 2007; Daunhauer & Fidler, 2011; Fidler, 2005). This profile is a result of a dynamic set of developmental processes that begin to emerge in early development (Dykens & Hodapp, 2001; Fidler, Lunkenheimer, & Hahn, 2011; Karmiloff-Smith, 2011). The phenotypic profile associated with DS is shaped probabilistically over time through the bidirectional influence of etiological constraints and the environment (Karmiloff-Smith, Casey, Massand, Tomalski, & Thomas, 2014). Understanding the nature of early developmental profiles in DS can provide potential insights into later, more complex areas of challenge.

Executive function (EF), in particular, is an area of significant challenge for individuals with DS, affecting the ability to engage in daily activities and execute goal-directed behavior (Fidler, Will, Daunhauer, Gerlach-McDonald, & Visootsak, 2014; Daunhauer & Fidler, 2011; Daunhauer, Fidler, Hahn, Will, Lee, & Hepburn, 2014). Although extant literature captures deficits in cognitive abilities specifically related to EF in DS, the early developmental foundations influencing the DS cognitive profile remain poorly understood. It is speculated that deficits in early motor development contribute to deficits in aspects of early EF (Fidler, Hepburn, & Osaki, 2011), but this has not been directly tested. Furthermore, the specific foundations through which motor development impacts early emergent EF have not been investigated.

The primary aim of the present study was to evaluate the impact of early atypical motor abilities on aspects of early EF (goal-directed behavior) in toddlers with DS (ages 11-45 months). The present study also aimed to examine the role that motor cognition and visual attention play as potential foundations that influence EF outcomes for toddlers with DS. A final aim of the present study was to test potential developmental cascades through which motor development impacts early EF outcomes in toddlers with DS. Collectively, results may provide early intervention targets in young children with DS.

CHAPTER TWO

Literature Review

Emergence of Behavioral Phenotypes

Phenotypes are the observed presentations of an underlying genotype (Nussbaum et al., 2001). The field of intellectual and developmental disability (IDD) research adopted the concept of “behavioral phenotypes” in order to describe the cognitive/behavioral profiles of specific neurogenetic syndromes associated with IDD (Hodapp & Dykens, 1994; Hodapp & Dykens, 2012). Neurogenetic syndromes, such as DS, can be characterized by phenotypic expression across a range of areas that include medical, physiological, and behavioral functioning. Increased research on behavioral profiles has contributed to our understanding of between syndrome differences, as well as the utility of phenotype-specific intervention planning (Fidler, Daunhauer, Will, Gerlach-McDonald, & Schworer, 2016; Fidler & Nadel, 2006; Fidler, Philofsky & Hepburn, 2007; Hodapp & Dykens, 2012; Will & Hepburn, 2015). A behavioral phenotype perspective also provides a foundation for characterizing the emergence of phenotypic vulnerabilities during early development, and as such, creates the potential to target cascading developmental trajectories via early intervention (Fidler, Lunkenheimer et al., 2011; Fidler & Nadel, 2007).

Behavioral phenotypes emerge through probabilistic epigenesis (Dykens, 1995; Dykens & Hodapp, 2001; Gottlieb, 1998). Probabilistic epigenesis describes the process of dynamic emergence of developmental profiles from bidirectional influences between genes and environment (Gottlieb, 1998; Gottlieb, 2007; Karmiloff-Smith, 1998). Within a probabilistic framework, individuals with a particular neurogenetic disorder are understood to demonstrate specific behavioral outcomes relative to those without the disorder. For example, both DS and

Williams syndrome (WS) are associated with intellectual disability (ID), yet individuals with DS and WS demonstrate different patterns of strength and weakness in aspects of social functioning, language abilities, and memory (Abbeduto et al., 2007; Fidler, et al., 2016; Klein & Mervis, 1999). By identifying phenotypic differences within early development, we can better understand the developmental complexities within each disorder.

The probabilistic nature of behavioral phenotypes also results in the potential overlap between syndromes, wherein certain behavioral outcomes are “partially specific” and observed in several disorders. For example, several neurogenetic syndromes, including Smith-Magenis, Prader-Willi, and DS, demonstrate greater propensity for repetitive and ritualistic behaviors, yet each syndrome demonstrates very different behavioral profiles in cognition, language, and other developmental domains (Dykens & Hodapp, 2001; Dykens, Hodapp, & Finucane, 2000; Levitas, Dykens, Finucane, & Kates, 2007). While similarities amongst neurogenetic disorders associated with ID provide a starting point for intervention, additional factors including between-syndrome differences and within syndrome variability must be considered for effective intervention.

Probabilistic emergence also gives rise to various degrees of within syndrome variability, where not every individual the same neurogenetic diagnosis manifests precisely the same phenotypic profile (Dykens, 1995; Hodapp & Dykens, 2012). For example, phenotypic complexities, such as the co-occurrence of developmental psychopathology, can manifest in some individuals with DS, but not others. Approximately 7-10% of children with DS have comorbid autism spectrum disorder (Diguiseppi et al., 2010) and up to 30% of individuals with DS have significant levels of inattention and aggression (Coe et al., 1999; Dykens, 2007). Despite increased risk for these comorbidities, not every individual with DS will carry a dual-diagnosis, and many will demonstrate varying levels of maladaptive symptomatology.

Dynamic systems theory

There is now growing recognition within behavioral phenotype research that patterns of developmental competence and challenge emerge and develop dynamically over time. As such, early phenotypic profiles can be understood as “starting states” that may shape later patterns of strength and challenge in a neurogenetic disorder such as DS (Fidler, Lunkenheimer et al., 2011). A dynamic systems perspective provides a useful framework for understanding early behavioral profiles through the application of the concepts of self-organization and developmental cascades (Fidler, Lunkenheimer et al., 2011; Karmiloff-Smith, 2011).

According to DST, interrelated aspects of development comprise a system, within which developmental patterns dynamically emerge and change over time (Granic, 2005; Karmiloff-Smith et al., 2012; Lewis, 2000). These early developmental pathways arise from early starting states, which are influenced by underlying genetic-etiology and neurodevelopment (Granic, 2005; Edgin, Clark, Massand, & Karmiloff-Smith, 2015; Lewis, 2000; Karmiloff-Smith, 1998; Smith & Thelen, 2003; Thelen & Smith, 1994). Thus, pronounced phenotypic patterns are preceded by the interaction between etiology, starting states, and the dynamic interplay between developmentally cascading systems (Granic, 2005; Karmiloff-Smith, 1998; Karmiloff-Smith et al., 2012; Karmiloff-Smith et al., 2014; Lewis, 2000; Smith & Thelen, 2003; Thelen & Smith, 1994).

In DS, genetic and neurodevelopmental constraints are placed upon the system, affecting early developmental processes (Edgin et al., 2015; Fidler, Lunkenheimer et al., 2011). Many aspects of early development are delayed within DS, including motor, language, and cognition (Abbedutto et al., 2007; Cicchetti & Ganiban, 1990; de Campos et al., 2013; Edgin et al., 2015). These compromised starting states shape specific developmental pathways and how these emergent pathways influence one another (Fidler, Lunkenheimer et al., 2011; Karmiloff-Smith et al., 2014; Thelen, 1994). Most importantly for this study, findings from infant science suggest that challenges with aspects of early motor abilities have the potential to influence outcomes related to other areas of developmental importance, such as early cognition- particularly goal-directed behavior (Gallese, Rochat, Sinigaglia, & Cossu, 2009; Piaget, 1952; Lockman, 2000; Needham & Libertus, 2011; Smith, 2005; Thelen, 1994). Evaluating the potential for this type of developmental cascade in a population with early motor delays, such as DS, can confirm this early developmental relationship.

Developmental areas of interest in DS

The present study focuses on the dynamic interaction between early motor and cognitive development in young children with DS. In typically developing children, each of these developmental domains refines itself through experience with the environment, while also exacting an influence on the other (Gallese et al., 2009; Gibson, 1979; Gibson & Pick, 2000; Needham & Libertus, 2011; Piaget, 1952; Thelen, 1994; Thelen et al., 2001). Early motor abilities afford the opportunity for object exploration, which then facilitates early learning and cognition through action and experience with objects (Bornstein, Hahn, & Suwalsky, 2013; Corbetta & Thelen, 1996; Gallese et al., 2009; Gibson & Pick, 2000; Keen, 2011; Libertus & Needham, 2010; Lockman, 2000; Needham & Libertus, 2011; Needham et al., 2002; Thelen, 1993; Thelen & Corbetta, 1994; Thelen et al., 2001). This link between motor and cognition has important implications for DS, as motor delays may restrict early environmental learning, particularly in early object-related goal directed behavior (Fidler, Hepburn, et al., 2011; Fidler, Lunkenheimer et al., 2011).

Motor abilities in DS. The motor system provides a critical foundation for the development of early goal-directed behavior in typically developing children (Thelen, 1995; Thelen, Kelso, & Fogel, 1987; Thelen & Smith, 1994). However, most children with DS experience significant motor challenges in early development, and also demonstrate a much greater propensity for hypotonia, or low muscle tone, and hyper-flexibility (Block, 1991; Harris & Shea, 1991; Jobling, 1998). Collectively, these motor challenges have implications for both the emergence and proficiency of motor abilities, likely influencing early exploration and its downstream effects on cognitive development in DS.

In general, motor abilities emerge late and variably in children with DS relative to typically developing children (Pereira, Basso, Lindquist, da Silva, & Tudella, 2013). Infants with DS demonstrate delays of up to 4 months in the acquisition of reaching and other gross motor abilities relative to TD infants of the same chronological age (CA; de Campos, Rocha, & Savelsbergh, 2010; Jobling & Mon-Williams, 2000; Pereira et al., 2013). Along with general delays in the emergence of motor abilities, motor development in DS is also characterized by significant variability (de Campos et al., 2013; Looper, Wu, Barroso, Ulrich, & Ulrich, 2006; Ulrich, Ulrich, Collier, & Cole, 1995). For example, early pre-walking behaviors were found to emerge variably in infants with DS, with some 15 month-olds showing less proficiency than 8 month olds (Ulrich et al., 1995). In addition, onset of independent walking has also been found to range between 19 months and 34 months in toddlers with DS (Ulrich et al., 1995).

Once young children with DS acquire motor abilities, their skills remain error-prone and unrefined (de Campos et al., 2010; Looper et al., 2006; MacTurk et al., 1985). De Campos and colleagues (2010) examined the development of reaching and grasping abilities in infants with DS and TD infants between at 4, 5, and 6-months of age. Compared to CA-matched TD infants,

infants with DS demonstrated poorer reaching control, a higher frequency of reaching without successfully grasping an object, and less refinement in grasping between 4 and 6 months (de Campos et al., 2010). Continued errors and lack of refinement in motor abilities have implications for the development and proficiency of exploratory experience for young children with DS.

Motor emergence and practice are important for the development of early cognition in typical development (Thelen, 1995; Gibson, 1979; Gibson & Pick, 2000; Lobo & Galloway, 2013; Lockman, 2000; Needham et al., 2002). The motor system self-organizes, developing from accumulated practice and repetition which contributes to increased muscle development and learning of effective motor strategies (Thelen, 1995). The resulting refinement in motor abilities affords greater exploratory opportunity (Corbetta & Thelen, 1994; Lockman, 2000; Thelen, 1995). In turn, object manipulation and exploration through motor experience facilitates better cognition, independently of age (Lobo & Galloway, 2013). In DS however, delayed and variable emergence of motor abilities, along with a lack of proficiency following experience, may influence the emergence of early cognition, particularly executive function (Barrett et al., 2008; Bornstein et al., 2013; Sommerville et al., 2008).

Executive Function. Executive function (EF) encompasses a set of interrelated self-regulatory cognitive constructs essential to daily functioning and adaptive behavior (Carlson, 2005). EF includes working memory, the temporary storage and manipulation of information; inhibition, the ability to ignore competing stimuli in the environment to execute a goal; and cognitive flexibility, the ability to shift thought processes or refocus one's attention following a change in the environment (Baddeley & Jarrold, 2007; Diamond, 2006). Each domain of EF contributes to the higher-order process of executing goal-directed behavior, or planning

(Diamond, 2006; Miyake et al., 2000; Pennington & Ozonoff, 1996; Zelazo et al., 1997). EF is associated with a wide range of outcomes in TD children, including academic success and adaptive behavior (Blair & Razza, 2007; Bornstein et al., 2013; Will, Fidler, Daunhauer, & Gerlach-McDonald, 2016).

Individuals with DS demonstrate a unique profile of challenges in EF (Daunhauer et al., 2014; Lee et al., 2014; Lee et al., 2011; Rowe, Lavender & Turk, 2006). This profile includes greater difficulty in working memory than individuals with other neurogenetic disorders, and more pronounced deficits than would be expected based on developmental status (Baddeley & Jarrold, 2007; Costanzo et al., 2013; Lanfranchi et al., 2009; Lanfranchi et al., 2010; Lanfranchi et al., 2012; Lee et al., 2011). Challenges in EF also include difficulties with some aspects of inhibition (Lanfranchi et al. 2012; Rowe et al., 2006). Verbal inhibition is impaired in DS relative to matched groups of both individuals with Williams syndrome (Costanzo et al., 2013) and typically developing children (Lanfranchi et al., 2010), yet there is little evidence for differences in the area of motor inhibition (Carney et al., 2013; Costanzo et al., 2013; Rowe et al., 2006). In terms of shifting, both adolescents and adults with DS demonstrate poorer shifting abilities compared to individuals with other developmental disabilities (Carney et al., 2013; Costanzo et al., 2013; Rowe et al., 2006). Challenges across each of these EF domains likely contribute to the marked impairments associated with the critical construct of planning in DS.

Planning is conceptualized as a central, higher-order subcomponent of EF that recruits working memory, inhibition, and shifting, to support the execution of goal directed-behaviors (Zelazo et al., 1997). Two primary components of planning include: 1) the insight involved in mentally representing the problem or goal at hand, and 2) planning and executing the necessary steps to solve the problem or execute the means-end goal (Zelazo et al., 1997). Planning, or goal-directed behavior, enables individuals to effectively engage with their environment (Banich, 2009; Carlson, 2005), and is also critical for adaptive and academic outcomes (Bornstein et al., 2013; Fidler, Hepburn et al., 2005). It is therefore potentially useful to identify foundations of early vulnerabilities associated with planning difficulties in DS.

Early development of planning

Goal-directed actions on objects emerge in infancy and early toddlerhood as the earliest form of planning (Corbetta, Thelen, & Johnson, 2000; McCormack & Attance, 2011; Needham & Libertus, 2011; Thelen et al., 2001). This early form of planning manifests in two different contexts: *object-related problem solving* and the *functional object use*. *Object-related problem solving* in early development involves executing multi-step actions towards a goal. For example, infants demonstrate object-related problem solving by pulling a string or moving a barrier in order to obtain a desired toy (Willatts, 1999). The second context—*functional object use*—involves using objects in a particular way based on the physical features, in order to execute a goal-directed action (Barrett et al., 2008; Keen, 2011; McCarty, Clifton, & Collard, 2001; McCormack & Attance, 2001). Examples include linking rings together or using a toy hammer to bang a ball. Certain developmental foundations interact to shape the emergence of these early object-related planning.

Both object-related problem solving and functional object use emerge in typically developing infants through three interrelated processes: motor abilities, visual attention, and motor cognition (Corbetta et al., 2000; Gibson, 1979; Gibson & Pick, 2000; Lockman, 2000; Needham & Libertus, 2011; Thelen, 1995). Motor abilities include fine and gross motor milestones that, within typical development but not necessarily DS (Vicari, 2006; Spano et al., 1999), generally track with chronological age such that certain milestones are expected around a particular age. *Visual attention* involves allocating attention towards objects for processing information about an object. *Motor cognition* is the integration or mental representation of object information necessary for executing a goal-directed action (Lockman, 2000; Needham et al., 2002; Thelen, 1995). These interrelated constructs influence one another to inform the emergence of early object-related planning. Early motor abilities enable an infant's manual exploration and enhanced visual exploration of objects. Accumulated experiences with object exploration, in turn, facilitate the development of the critical construct of motor cognition, or the integration of perceived object information into an action plan. Gradually, ongoing experiences that involve motor abilities, visual attention, and motor cognition interact to facilitate the development of object-related planning (Gibson, 1979; Gibson & Pick, 2000; Lockman, 2000; Thelen, 1995; Thelen et al., 1988). A closer examination of how these constructs develop normatively can serve as an important foundation for identifying atypical developmental patterns in early development in DS.

Planning in typical development. The typical trajectory of early object planning involves a shift from more basic modalities of exploration (visual), to intermediate modalities (manual) to enhance object knowledge, then back to more advanced iterations of previous modalities (visual) when the intermediate are no longer required (Lockman, 2000; Needham & Libertus, 2011; Thelen, 1995). In the first 3 months, infants initially learn about objects through visual exploration (Baillargeon, 1987; Baillargeon & DeVos, 1991). Using these visual skills, infants begin to recognize important features of objects, and they develop expectations about objects based on visual information (Baillargeon & DeVos, 1991). However, once an infant's motor repertoire develops to include reaching and grasping, they begin to engage in a more sophisticated manual exploration (Corbetta et al., 2000; Gibson, 1979; Gibson & Pick, 2000; Thelen, 1995; Thelen et al., 1988).

Manual exploration is an important infant milestone because it specifically allows infants to perceive new and important information about object characteristics that they were unable to perceive through visual exploration alone (Bourgeois, Khawar, Neal, & Lockman, 2005; Clifton, et al., 1991; Gibson, 1979; Gibson & Pick, 2000; Thelen, 1995; Thelen et al., 1988). For example, as infants grasp and manually explore, they detect information about object texture (e.g. whether an object is squishy or hard), and then utilize this information to adjust their motor acts (e.g. change their grasp) to better accommodate the object characteristics (Gallese et al., 2009). This progression is demonstrated in studies where 4 to 6 months-old infants demonstrate grasp planning based on object characteristics, but only after acquiring information from manual contact, indicating that the experience of manual exploration provided critical information to inform object related planning (Corbetta et al., 2000; Lobo & Galloway, 2013; Newell et al., 1989; Newell et al., 1993).

Interestingly, after this developmental period in which motor abilities emerge and become refined, infants subsequently shift back the use of visual attention for object-related planning (Corbetta et al., 2000). As infants get older, their object-related planning progresses, and they display forms of planning with only the visual perception of object characteristics. Around 8-months of age, typically developing infants demonstrate reach and grasp planning based on object affordances prior to any physical contact with the object (Corbetta et al., 2000; Lockman et al., 1984; von Hofsten & Fazel-Zandy, 1984; von Hofsten & Ronnqvist, 1988). However, the intermediate manual exploration that occurs once motor abilities afford manual exploration is essential for the development of motor cognition (Corbetta et al., 2000). This intermediate stage facilitates more refined planning (Lobo & Galloway, 2013), and subsequently, infants can again rely only on visual attention (Corbetta et al., 2000). As development in motor abilities, visual attention, and motor cognition progress, these developmental constructs interact to shape the emergence of object-related planning.

Bidirectional influence of planning constructs. The developmental progression described above demonstrates that the foundational skills of motor abilities, visual attention, and motor cognition are interdependent, and infants rely on the interaction between all three for the development of effective planning (Bourgeois et al., 2005; Corbetta et al., 2000; Needham et al., 2014; Needham & Libertus, 2011; Thelen et al., 2001). These constructs operate in a bidirectional feedback loop, wherein each exacts influence on and enhances one another (Lockman, 2000; Needham & Libertus, 2011; Thelen, 1995). Motor abilities facilitate the opportunity to interact with objects, which enhances visual attention to objects and motor cognition (Needham et al., 2002). Enhanced visual exploration and motor cognition each contribute to a greater understanding of objects, which facilitates the further development of object-related planning (Lobo & Galloway, 2013; Needham et al., 2002). These bidirectional influences have been empirically demonstrated in a series of infant intervention studies, where infants were provided the opportunity for enhanced object exploration (Barrett et al., 2008; Libertus & Needham, 2010; Needham et al., 2002). This enriched set of exploratory opportunities resulted in accelerated motor development, enhanced visual attention to objects, and enhanced motor cognition (Libertus & Needham, 2010; Needham et al., 2002).

Taken together, the ongoing interactions between motor abilities, visual attention, and motor cognition serve as an important context for the emergence of early object-related planning (Barrett et al., 2008; Corbetta & Thelen, 1994, 1996; Corbetta et al., 2000; Gallese, Rochat, Cossu, & Sinigaglia, 2009; see also Keen, 2011 for review; Libertus & Needham, 2007; Needham & Libertus, 2011). Disruption in any of these foundational mechanisms, motor abilities in particular, has significant implications for the emergence of object-related planning. Because motor abilities are related to both visual attention and motor cognition in TD infants (Corbetta et al., 2000; Needham, 2002), abnormal motor abilities experienced by infants and toddlers with DS are likely to affect these constructs, and in turn, object-related planning. Early motor challenges in infants and toddlers with DS are therefore hypothesized to facilitate an atypical developmental cascade onto early object-related planning.

Planning in DS. Children with DS consistently demonstrate challenges in planning (Fidler, Hepburn et al., 2005; Fidler, Philofsky et al., 2005; Lee et al, 2011; Daunhauer et al., 2014). Although object-related problem solving and functional object use are different aspects of planning, both are diminished in DS (Fidler, Hepburn et al., 2005; Fidler Philofsky et al., 2005; Fidler et al., 2014). There are significant implications for compromised object-related planning in DS, and it is therefore important to understand its foundations within early development (Fidler, Hepburn et al., 2005; Fidler, Philofsky et al., 2005; Fidler et al., 2014; Yoder, Woynaroski, Fey, & Warren, 2014).

Object related problem solving. A child’s ability to adapt an approach in order to obtain a desired object is an indication of object-related problem solving. In classic object retrieval tasks, a child must first pull a cloth in order to bring an object into reach (Willatts, 1999), or modify a reach in order to obtain a desired object (Bojczk & Corbetta, 2004; Keen, 2011; Fidler, Hepburn et al., 2005; Fidler, Philofsky et al., 2005). Toddlers with DS demonstrate significantly diminished object-related problem solving compared to MA-matched TD children and children with other developmental disabilities (Fidler, Hepburn et al., 2005; Fidler, Philofsky et al., 2005).

Specifically, toddlers with DS produce significantly less efficient reaching strategies than developmentally matched children (Fidler, Hepburn et al., 2005; Fidler, Philofsky et al., 2005). Early disruption to developmental foundations including motor abilities, visual attention, and motor cognition may influence object-related problem solving outcomes.

Functional object use. In addition to object-related problem solving, functional object use is another critical aspect of early planning skills. Functional object use relies on perceiving object affordances to perform a functional action. For example, understanding that a string of beads is flexible and can fit into a cup enables a child to perform the *functional act* of putting the beads into the cup (Fidler, Hepburn et al., 2005). Young children with DS demonstrate diminished abilities in this area (Fidler, Hepburn et al., 2005; Fidler et al., 2014). In one study, when provided with a collection of toys with ambiguous play value, school aged children with DS performed significantly fewer functional acts on objects, and showed significantly reduced interest in objects relative to comparison groups (Fidler, et al., 2014).

Similar challenges observed in toddlers with DS indicate that challenges in the functional use of objects emerge early in the DS lifespan (Fidler, Hepburn et al., 2005). For example, when compared to MA-matched toddlers with other developmental disabilities, toddlers with DS performed significantly worse on functional object use planning task items, such as putting coins in a bank (Fidler, Hepburn et al., 2005). Although functional object use and object-related problem solving are distinct aspects of planning, a similar pattern of developmental foundations may contribute to functional object use outcomes for toddlers with DS.

Implications of diminished planning in DS. In typically developing children, early object-related experiences are linked to outcomes related to IQ and academic achievement (Bornstein et al., 2013), tool use (Claxton, Keen, & McCarty, 2003; Kahrs & Lockman, 2014), and greater overall understanding of one's environment (Libertus & Needham, 2010; Lockman, 2000). Preliminary findings suggest object-related planning challenges impact other areas of development in DS, including communication (Fidler, Philofsky et al., 2005), adaptive behavior (Fidler, Hepburn et al., 2005), and response to intervention (Yoder et al., 2014). Because of this potentially widespread impact, it is critical to understand the contribution of motor abilities, visual attention, and motor cognition as early foundations of object-related planning outcomes in toddlers with DS.

Hypothesized early emergence of planning in DS

The emergence of object-related planning observed in typical development is a pathway that is likely to be disrupted in DS. Early object planning emerges from early motor foundations in typical development (Ballard et al., 2013; Bourgeois et al., 2005; Gallese et al., 2009; Lockman, 2000; Needham & Libertus, 2011; Thelen, 1995; Thelen et al., 1988). As a starting state, motor abilities afford enhanced perception of object affordances through visual and manual exploration (Bourgeois et al., 2005; Corbetta et al., 2000; Lobo & Galloway, 2013; Smith, 2005; Thelen et al., 2001). Throughout these processes, infants develop motor cognition, which, along with visual perception, enables them to execute goal-directed action on objects (Bourgeois et al., 2005; Corbetta et al., 2000; Lockman, 2000; Needham & Libertus, 2010; Thelen et al., 1995). Constraints at any juncture within these interrelated processes have implications for the emergence of object-related planning (Corbetta et al., 2000; Smith, 2005). The early DS behavioral phenotype includes several potential constraints to this early set of processes, including compromised integrity of the motor system, atypical visual attention, and diminished manual exploration (de Campos et al., 2010; de Campos et al., 2013; Fidler, Hepburn et al., 2011; Fidler, Lunkenheimer et al., 2011; MacTurk et al., 1985).

Children with DS have significant motor delays and challenges that extend well beyond the first year of life and affect early object-related experiences (de Campos et al., 2010; de Campos et al., 2013; MacTurk et al., 1985; Pereira et al., 2013). A diminished ability to manually explore objects due to early motor delays results in disproportionate levels of visual attention in infants with DS (de Campos et al., 2013; MacTurk, 1985). Infants with DS spend significantly more time visually attending to objects than TD infants (de Campos et al., 2013; MacTurk et al., 1985). Increased visual attention to objects may benefit toddlers with DS in the development of object-related planning in through increased visual processing of object characteristics. Alternatively, enhanced visual attention may compromise object-related planning if toddlers with DS fail to also manually explore. This is likely given that disproportionate visual attention persists after infants with DS have motor abilities affording manual exploration, indicating a lack of adaptation in exploratory patterns (MacTurk et al., 1985). Disproportionate visual attention, along with reduced manual exploration, may collectively compromise object-related planning outcomes.

As infants with DS spend significantly more time in visual exploration, they spend significantly less time in manual exploration (de Campos et al., 2010; de Campos et al., 2013; MacTurk et al., 1985). A lack of manual exploration of objects deprives infants and toddlers with DS of the opportunity for enhancing their detection of affordances, as well as the development of motor cognition (Bourgeois et al., 2005; Corbetta et al., 2000; Lobo & Galloway, 2013). With reduced manual exploration, infants and toddlers with DS are less able to gain new perceptual information and integrate it into their action planning repertoire, leading to compromised motor cognition (Corbetta & Thelen, 1994, 1996; Corbetta et al., 2000; Needham & Libertus, 2011). Exploration and experience that facilitates motor cognition directly leads to the development of object-related planning (Bourgeois et al., 2005; Corbetta et al., 2000; Thelen, 1995). Thus, diminished motor cognition is another potential foundation that potentially leads to object-related planning difficulties in DS.

Motor abilities, visual attention, and motor cognition are potential areas of developmental constraint in DS. The interaction between these foundational constructs likely influences the atypical emergence of object-related planning in DS. However, the precise influence of these foundations is unclear. Therefore, this study focuses on identifying the developmental impact of these foundations on object-related planning in toddlers with DS.

This Study

The aim of this study was to examine motor abilities, visual attention, and motor cognition as developmental foundations of object-related planning outcomes in toddlers with DS. Figure 1 presents the conceptual model of predictions of how motor abilities, visual attention, and motor cognition are associated with object-related problem solving and functional object use outcomes.

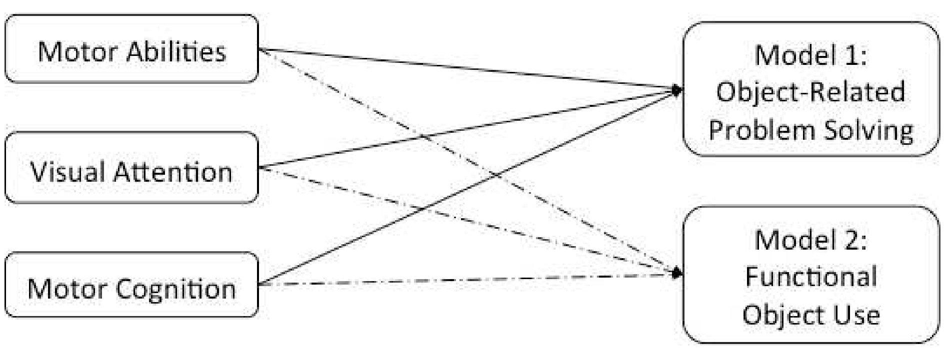

Figure 1
Hypothesized Planning Models

Within TD, these constructs develop in concordance with age normatively. Motor abilities in particular, are an early starting state of related constructs, and one that emerges expectedly within a certain, normative developmental window (Corbetta & Thelen, 1994;

Thelen, 1995). The utility of examining age as a reliable predictor of developmental outcomes has been questioned (Rutter, 1989; Wohlwill, 1970). Specifically, it has been argued that age is intertwined with development, greater utility results from deconstructing age into its componential parts- in this instance, motor abilities (Rutter, 1989). This has distinct relevance to DS because motor abilities emerge within a highly variable window (Periera et al., 2013; Vicari, 2006; Spano et al., 1999) that is not necessarily in concordance with CA. Toddlers with DS of the same CA may have very different motor repertoires, and therefore, very different accumulated object-related and exploratory experiences. Two approaches within this study were taken to address the developmental variability within DS. First, correlations among study constructs between age-subgroups within the sample were examined. Second, CA replaced motor abilities as a foundational construct of planning outcomes in a second set of analyses.

Finally, a cross-sectional mediation model was examined to test a potential developmental cascade between motor abilities and object-related planning outcomes through motor cognition and visual attention was also tested. Figure 2 depicts the conceptual model for a cross-sectional multiple mediation model for object-related problem solving outcomes.

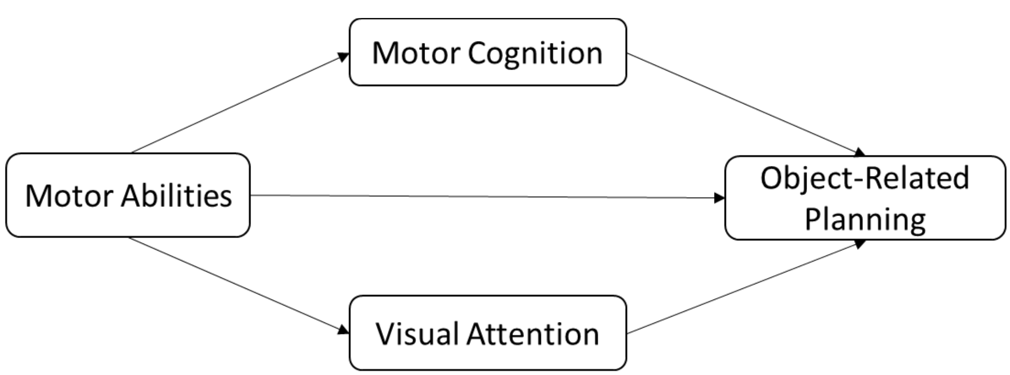

Figure 2
Cross-sectional Mediation Model

Hypotheses. The present study included the following hypotheses:

1. **Problem Solving Outcomes:** It is hypothesized that a) motor abilities, b) visual attention, and c) motor cognition will each be significantly associated with object-related problem solving when controlling for other predictors in the model (see model 1, Figure 1).
2. **Functional Object Use Outcomes:** It is hypothesized that a) motor abilities, b) motor cognition, and c) visual attention will each be significantly associated with functional object use, when controlling for the other predictors in the model (see model 2, Figure 1).
3. **Cross-sectional mediation:** It is hypothesized that the model depicted in Figure 2 will demonstrate effects consistent with mediation, such that a) motor abilities will be significantly associated with motor cognition, which will be significantly associated with object-related planning outcomes; and b) motor abilities will also be significantly associated with visual attention, and in turn visual attention will be significantly associated with object-related planning outcomes (see Figure 2).

Exploratory analyses. The utility of age as a predictor variable within early development (Rutter, 1989; Wohlwill, 1970) and DS in particular is unclear, yet may be a potential contributor to planning outcomes in toddlers with DS. Accordingly, regression analyses examining the association of chronological age to object related problem solving and functional object use outcomes when controlling other predictors in the model were exploratory.

CHAPTER THREE

Methods

Participants

Participants consisted of 38 toddlers with DS between the ages of 11 and 45 months [mean chronological age (CA) = 24.84 months (SD=9.91); mean nonverbal mental age (NVMA) = 15.54; SD=4.64]. The majority of the sample was Caucasian and male (see Table 1 for sample description). The majority of participants (89%) had DS caused by Trisomy 21, and 4 participants had Mosaic DS.

Table 1
Participant Demographic Information

Variable	*M or n*	*(%) or SD*
Nonverbal Mental Age (NVMA) in Months	15.59	4.61
Chronological Age (CA) in Months	24.84	9.91
Gender (Male)		
Male	21	(55.3%)
Female	17	(44.7%)
Mothers' age in years (n=34)	38.03	4.71
Race/Ethnicity		
Caucasian	30	(78.9%)
American Indian/Alaska Native	2	(5.3%)
More than one race	2	(5.3%)
Unreported	4	(10.5%)
Hispanic or Latino	7	(18.4%)
Not Hispanic or Latino	24	(63.2%)
Unknown or not reported	7	(18.4%)

Procedure

Ethical approval was obtained from the Colorado State University Institutional Review Board in spring of 2015. Recruitment began in fall of 2015 and continued through early fall of 2016. Participants' parents were recruited through local DS organizations including Rocky Mountain Down Syndrome Association (RMDSA), the Sie Clinic for Down Syndrome at Children's Hospital, Colorado, the Colorado Springs Down Syndrome Association, and the Northern Colorado Down Syndrome Association; as well as international DS organizations, such as the International Mosaic Down Syndrome Association. The majority of participants were assessed in-home. A total of six participants were assessed out-of-state, requiring assessments to take place in hotel conference rooms.

Parents provided consent for their child's participation. Sessions lasted approximately 1.5 hours. The participants engaged in approximately 45 minutes of direct assessment, and parents completed questionnaires on their child's developmental history. Parents received a summary of their child's developmental assessment with therapeutic recommendations as compensation for participating.

Measures

Nonverbal Mental Age. The Mullen Scales of Early Learning (MSEL; Mullen, 1995) was administered to obtain an estimate of nonverbal mental age (NVMA). The MSEL is a comprehensive developmental assessment that measures gross motor, fine motor, visual reception (i.e. early cognition), receptive language, and expressive language. It is developmentally appropriate and normed for age birth through 5 years 8 months old. It has been standardized on a large sample (N=1,836). It has a high established internal consistency (0.75-0.83) and internal reliability (median=0.91). The test-retest reliability ranges from 0.71-0.96, and it also has high validity (see Mullen, 1995). It was specifically chosen because it can be administered in as little as 15 minutes, which is sensitive to the proposed ages of the participant sample. Age equivalence scores were derived for each subdomain using the MSEL scoring manual and guidelines. A NVMA was derived from averaging the age equivalence scores for visual reception and fine motor. NVMA was used to describe the sample in order to remove any language confounds from estimating developmental status (see Table 2 for descriptive statistics).

Motor abilities. Motor abilities were measured using the Gross Motor and the Fine Motor domains of the MSEL. Correlations between the Gross and Fine motor scale raw scores were tested to determine whether it was suitable to derive a motor composite variable from averaging these scores (see Table 3). The Fine and Gross motor scales were significantly and positively correlated with one another (r=0.67; p<0.001), so a composite variable was computed by averaging the summed raw scores of each of these scales.

Visual attention. Visual attention was measured in a 2-part task. In the first part of the task, participants were given colored wooden blocks to play with for up to 2 minutes. In the second part of the task, participants were given novel looking toys (i.e., Scrubbles) to play with for up to 2 minutes (see Figure 3).

Figure 3
Scrubbles

Behavior Coding. Total duration in seconds of visual exploration across the tasks was coded as a measure of object interest. Undergraduate research assistants were trained on behavior coding for this task. Research assistants trained to a reliability threshold of 0.70 kappa and reliability was met across approximately 30% of the sample with an average kappa of 0.74. Manual exploration was coded but not analyzed in order to avoid measurement confounds between motor abilities and object interest.

Motor Cognition. Motor cognition was measured using Barrett, Traupman, and Needham's, (2008) battery. In this task, participants were presented with 4 similar, but structurally unique balls (see Figure 4). Each ball provided differing affordances based on size, material, malleability, and physical structure. Each participant was presented with a single ball at a time and allowed to explore it for up to 30 seconds. After 30 seconds, the ball was removed, and the next ball was presented.

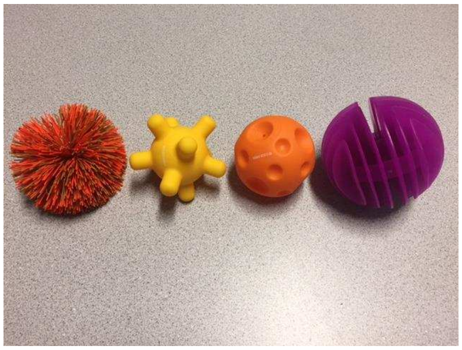

Figure 4
Motor Cognition Battery

Behavior Coding. This task was coded according to Barrett and colleagues (2008) original coding scheme. On each trial (i.e., ball), latency to *initial contact* was coded for. Type of contact was coded to distinguish between touching and swatting. Once contact was made, the latency between that contact and *initial grasp* was coded. After the initial contact and grasp were made (if at all), frequency of subsequent contact and grasping was coded. In addition, other exploratory behaviors, including mouthing and squeezing, were also coded. Graduate research assistants trained to a reliability threshold minimum of 0.70 Cohen's Kappa and coded approximately 30% of the sample for reliability purposes. A minimum reliability of 0.70 kappa was maintained and the average kappa across the reliability sample was high (average kappa = 0.78). The final variable used as an indicator for motor cognition was latency to initial grasp. The use of grasping as an optimal end-state of motor cognition is consistent with previous infant science research and conceptualization of this motor cognition task (Barrett et al., 2008; Corbetta & Thelen, 1994; Corbetta et al., 2000; Needham et al., 2002). Because latency to initial grasp is a measure of the time between when the participant obtained physical information about the object, and grasped the object, it reflects the child's time spent processing a motor approach based on physical affordances of the object.

Object-related problem solving. A graduated object retrieval battery was administered as a measure of object-related problem solving. A graduated battery was selected to account for developmental variability across the sample and included three different object retrieval tasks requiring a problem-solving element. The first and most developmentally simple task was an infant object retrieval task (Willatts, 1999), in which a desired toy is placed out of the child's reach, but on a cloth that is within the child's reach. This is an object-related problem solving task because it requires the child to pull the cloth in order to bring the toy within their reach and obtain it. In the second task, which was slightly more developmentally advanced, a desired toy was placed in a clear Plexiglas box with a hinged door on it. This task is an object-related problem solving task because it requires the child to open the door in order to obtain the toy. The third and most developmentally advanced task was an object retrieval task (Fidler, Hepburn et al., 2005). In this task, a desired toy was placed under a Plexiglas box with one open side, requiring the child to reach through the open side in order to obtain the toy. A total of 5 trials were administered and the open side was rotated on each trial. This final task is an object-related problem solving task because it requires the child to adapt his/her reaching strategy in order to obtain the toy. The task was discontinued if the child was unable to retrieve the toy on the first 3 trials or became visibly distraught (e.g., whining or fussing).

Behavior Coding. For the first task, whether the child pulled the cloth was coded (i.e., yes/no), and whether the child obtained the toy was coded (i.e., yes/no). For the second task, whether the child opened the door was coded, and whether the child obtained the toy was coded. For the third object retrieval task, successful instances of object retrieval were coded across each trial. Undergraduate coders were trained to a reliability threshold of .70 kappa. Approximately

30% of the sample across each task was coded for reliability. Average reliability was high (average kappa=0.85).

A “basal” and “ceiling” approach from traditional developmental assessment procedures was utilized in order to account for developmental variability across the sample. If a participant successfully passed the middle task, correct performance on the basic-level task was assumed. If a participant failed the middle task, a “ceiling” effect of null performance on the most advanced task was assumed. All “yes” responses were recoded as 1, and all “no” responses were coded as zero. A total summed score of successful retrieval was obtained by summing the points across each task for a minimum score of zero and a maximum score of 9. For example, if a participant successfully performed both actions on the first two tasks, but failed to retrieve a toy across any trial on the third task, their score would be a 4. A small number of younger participants obtained the toy on the first task without pulling the cloth. In this instance, these participants were given a score of 2 on this task because they still engaged in problem solving goal-directed behavior by moving closer to the toy (by scooting or kneeling) in order to reach the toy.

Functional object use. Functional object use was measured using an adapted Generativity task (Rutherford & Rogers, 2003; Fidler, et al., 2014). For this task, each participant was presented with a collection of toys, each of which consisted of different physical affordances (i.e., properties) and that collectively had an ambiguous play value. Examples of toys included Slinkys, accordion tubes, plastic rings, stretchy rubber toys, and pipe cleaners. This task was adapted from previously used versions (i.e., Fidler et al., 2014) to include toddler-appropriate toys. Toys were presented for 2 minutes and the examiner provided minimal attention to the participant during this time. If the participant tried to engage the examiner, the examiner nonverbally redirected the participant back to the set of toys using gestural prompts.

Behavior coding. In this task, interaction with objects was coded in a variety of ways across the 2-minute task, consistent with previous work (Fidler et al., 2014). Actions performed on objects that specifically utilized the physical affordances of the object or objects in conjunction with one another were considered functional. Examples of functional acts included but were not limited to linking rings together, grouping like objects, threading pipe cleaners through beads, and expanding Slinkys. Actions were coded as sensory if they only provided sensory input and did not utilize object affordances. Examples included but were not limited to swinging beads, spinning rings, and shaking a tube. First, objects were coded as the participant interacted with them, for whether the object was a new (i.e., one the participant had not yet interacted with) or familiar (i.e., one the participant had already interacted with during the 2-minute segment); whether the action performed on the object was sensory oriented- interacting in a way that only provided sensory input (e.g., shaking a slinky) or functional- that is based on the physical properties of the object (e.g., expanding and contracting an accordion tube); and whether the action was a new action (i.e., one they had not yet performed across the 2-minutes) or a familiar action (i.e., one they had already performed across the 2-minutes). Approximately 30% of the total sample was coded for reliability, which was also high (average kappa=0.77). Total number of functional acts on objects was used as an indicator of functional object use in regression analyses.

Table 2
Descriptive Statistics

Variable	M	SD	Min – Max
Motor Composite	17.25	5.25	8.50 – 31.00
Visual Attention (in seconds)	174.11	58.00	58.00 – 240.00
Motor Cognition (in seconds)	8.56	15.74	0.30 – 92.59
Object Retrieval	6.74	2.25	2.00 – 9.00
Functional Object Use	5.38	4.71	0.00 – 19.00

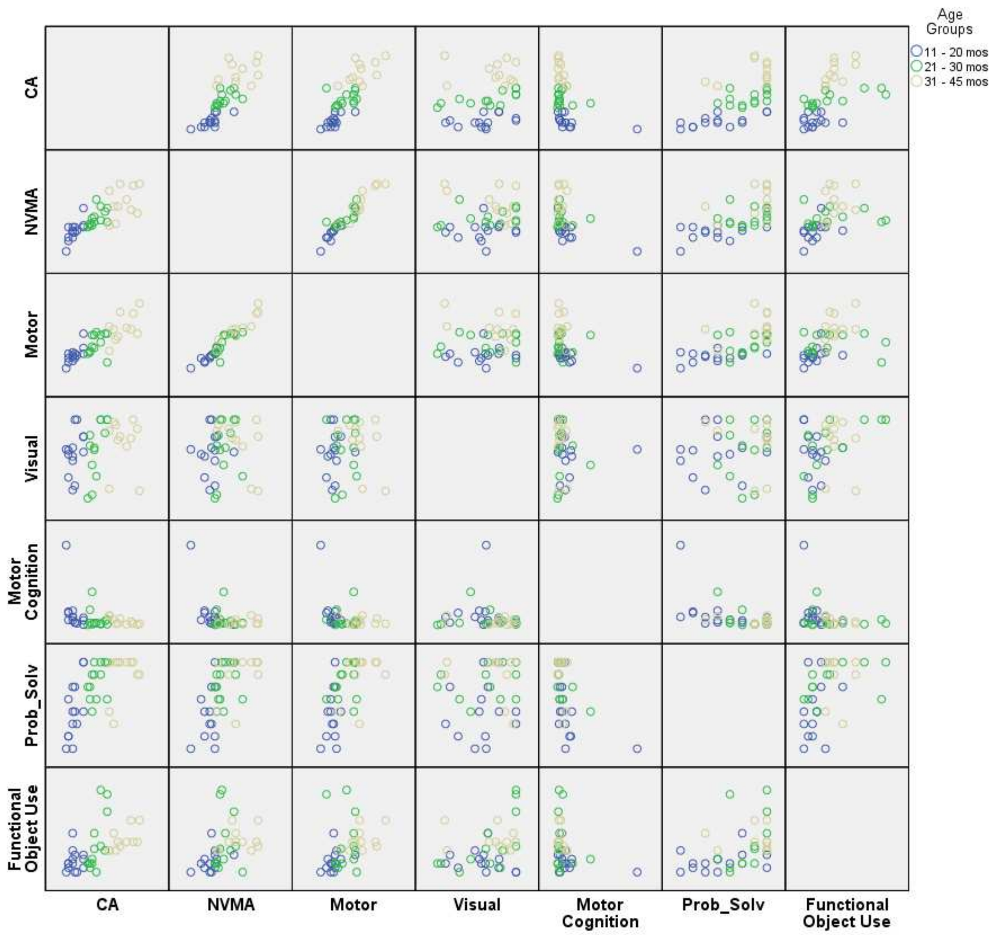

Figure 5
Age-grouped scatterplot matrix

CHAPTER FOUR

Results

Analytic approach

Descriptive statistics were calculated for all variables (see Table 2) and normality of distributions were examined. Figure 5 presents a scatterplot matrix of variables color coded for age subgroups. Non-normal distributions of residuals in the first regression model required motor cognition to be log transformed. Mean replacement was used to handle missing functional object use data for two participants. For the purpose of addressing potential age-based variability within the sample, frequency distributions for CA were calculated and identified three age-based subgroups within the sample: 1) 11 – 20 month olds (n=14); 2) 21 – 30 month olds (n=13); and 3) 31 – 45 month olds (n=11). Associations among variables were examined using bivariate correlations and included within the identified age subgroups as well as the overall sample (see Table 3). Multiple linear regressions and analyses were performed to test hypotheses. To test the first hypothesis, contributing foundations to object-related problem solving were examined by regressing object-related problem solving on motor abilities, visual attention, and motor cognition. To test the second hypothesis, contributing mechanisms of functional object use were examined by regressing functional object use on motor abilities, visual attention, and motor cognition. Two subsequent multiple regression models to test the potential contribution of age rather than motor abilities on object related planning outcomes were also estimated. Finally, to test cross-sectional mediating effects, a multiple mediation model was also tested.

Table 3

Correlation Coefficients between Key Variables

	1	2	3	4	5	6	7	8	9
	Entire Sample (N=38)								
CA	-								
NVMA	.80**	-							
Gross Motor	.83**	.89**	-						
Fine Motor	.64**	.83**	.67**	-					
Motor Composite	.82**	.95**	.93**	.89**	-				
Visual Attention	.26	.06	.11	-.20	.06	-			
Motor Cognition	-.34*	-.41*	-.27	-.35*	-.33*	-0.10	-		
Problem Solving	.63**	.61**	.61**	.50**	.61**	0.14	-.50**	-	
Functional Use	.55**	.4*	.57**	.07	.38*	0.34*	-.28	.43**	-
	11 - 20 month olds (n=14)								
CA	-								
NVMA	.74**	-							
Gross Motor	.40	.74**	-						
Fine Motor	.86**	.90**	.49	-					
Motor Composite	.74**	.96**	.85**	.87**	-				
Visual Attention	.36	.06	-.15	.30	.01	-			
Motor Cognition	-.49	-.67**	-.31	-.67**	-.57*	-.30	-		
Problem Solving	.67**	.56*	.50	.49	.57*	.11	-.48	-	
Functional Use	.10	.37	.41	.23	.37	-.25	-.32	.31	-
	21 - 30 month olds (n=13)								
CA	-								
NVMA	.52*	-							
Gross Motor	.72**	.64*	-						
Fine Motor	-.27	.51	.07	-					
Motor Composite	.24	.78**	.68*	.79**	-				
Visual Attention	.70**	.26	.38	-.21	.08	-			
Motor Cognition	-.24	-.14	.07	.06	-.08	-.10	-		
Problem Solving	.38	.14	.30	.21	.34	.31	-.57*	-	
Functional Use	.72**	.02	.49	-.55	-.11	.67**	-.20	.40	-
	31 - 45 month olds (n=11)								
CA	-								
NVMA	.35	-							
Gross Motor	.65*	.86**	-						
Fine Motor	.39	.93**	.83**	-					
Motor Composite	.57	.92**	.97**	.93**	-				
Visual Attention	-.08	-.50	-.34	-.52	-.43	-			
Motor Cognition	-.73*	.01	-.35	.04	-.21	-.23	-		
Problem Solving	.36	.38	.40	.24	.35	-.26	-.38	-	
Functional Use	.63*	.25	.49	.38	.47	-.24	-.35	-.04	-

*Note: *p<0.05; **p<0.01*

Table 4

Primary Regression Models

	Model 1: Object-Related Problem Solving						Model 2: Functional Object Use					
	b	*SE(b)*	*B*	*p*	95% CI		*b*	*SE(b)*	*B*	*p*	95% CI	
					Lower	Upper					Lower	Upper
Intercept	7.83	0.42		0.000	6.97	8.68	5.38	0.69		0.000	4.06	6.70
Motor Abilities	0.19	0.05	0.45	0.001	0.09	0.30	0.28	0.14	0.31	0.055	-0.01	0.56
Visual Attention	0.004	0.005	0.09	0.449	-0.01	0.01	0.03	0.01	0.31	0.045	0.00	0.05
Motor Cognition	-0.79	0.24	-0.42	0.002	-1.27	-0.30	-0.04	0.05	-0.15	0.359	-0.14	0.52
	R^2=0.54; p<0.001						R^2=0.26; p<0.015					

Note: Motor cognition was log transformed in model 1; all predictors were centered at the mean

Table 5

Age as predictor of planning outcomes

	Model 3: Object-Related Problem Solving						Model 4: Functional Object Use					
	b	*SE(b)*	*B*	*p*	95% CI		*b*	*SE(b)*	*B*	*p*	95% CI	
					Lower	Upper					Lower	Upper
Intercept	7.76	0.43			6.89	8.64	5.38	0.64			4.08	6.68
CA	0.12	0.03	0.47	0.001	0.04	0.17	0.22	0.07	0.46	0.004	0.08	0.37
Visual Attention	0.00	0.01	-0.01	0.970	-0.01	0.01	0.12	0.01	0.21	0.153	-0.01	0.04
Motor Cognition	-0.74	0.25	-0.39	0.005	-1.24	-0.24	-0.03	0.04	-0.10	0.495	-0.12	0.06
	R^2=0.52; p<0.001						R^2=0.36; p<0.003					

Note: Motor cognition was log transformed in model 3; all predictors were centered at the mean

Age-grouped bivariate correlations

Age-grouped bivariate correlation results indicated a variable pattern of association amongst variables across different age groups (see Table 3). In the younger age group (11 – 20 months), there was a strong and significant correlation between CA and fine motor, but not CA and gross motor. In addition, CA was also strongly associated with motor cognition in this age group. In the middle age group (21 – 30 months) however, CA as significantly associated with both fine and gross motor, as well as visual attention, but not motor cognition. In the oldest age group (31 – 45 months), CA was significantly associated with both gross motor and fine motor, as well as motor cognition. In both of the older groups, but not the younger group, CA was significantly associated with functional object use. Collectively these results demonstrate the variability in developmental areas in relation to CA within DS.

Motor abilities as a planning foundation

Foundations of object-related problem solving. Motor abilities, visual attention, and motor cognition were tested as predictors of object-related problem solving using a multiple regression model. Collectively, motor abilities, motor cognition, and visual attention accounted for approximately 54% of the variance in object-related problem solving (R^2=0.54; see Table 4, model 1). Results also indicated that when holding motor cognition and visual attention constant, motor abilities significantly predicted object-related problem solving (b=0.19; p=0.001). This effect was such that for an increase of 2 standard deviations in motor abilities, problem solving scores were predicted to increase by approximately 1-point (B=0.45). In addition, when holding motor abilities and visual attention constant, the natural log of motor cognition was also a significant predictor of object-related problem solving (b=-0.79; p=0.002). This effect was such that doubling the average motor cognition score to 17 seconds, problem solving would decrease by approximately 1-point. Likewise, increasing log motor cognition by 2 standard deviations was associated with a predicted 1-point decrease in problem solving scores (*B*=-0.42). Visual attention was not significantly associated with object-related problem solving outcomes.

Foundations of functional object use. A second multiple regression model was tested to identify predictive foundations of functional object use. Functional object use was regressed on motor abilities, visual attention, and motor cognition. Collectively, motor abilities, motor cognition, and visual attention accounted for approximately 26% of the variance in functional object use (R^2=0.26; see Table 4, model 2). When holding visual attention and motor cognition constant, motor abilities were approaching significance in association with the functional object use outcomes (*b*=0.28; *p*=0.055). This effect was such that an increase of 3 standard deviations in motor abilities was associated with approximately 1 additional functional act (*B*=0.31). In addition, when holding motor abilities and motor cognition constant, visual attention was also significantly associated with functional object use (*b*=0.03; *p*=0.045). This effect was such that, an increase of 3 standard deviations in visual attention was associated with a predicted increase of one additional functional act (*B*=0.31), which was a significant effect. Motor cognition was not significantly associated with the functional object use outcomes.

Age as a foundation of planning

Motor abilities tend to progress concurrently with CA, and although this progression is highly variable within DS, CA is a potential proxy for accumulated motor experience. Accordingly, two subsequent regression models estimating chronological age rather than motor abilities as a predictor of planning outcomes were tested. In the third model, object-related problem solving was regressed on CA, visual attention, and motor cognition. Findings from this model were similar to that of model 1 (see Table 5, model 3), with motor abilities as a predictor of object-related problem solving. Collectively, a similar amount of the variance was accounted for (R^2=0.52; p<0.001) and patterns of significance were also similar to model 1. Specifically, CA (b=0.12; p=0.001) and motor cognition (b=-0.74; p=0.001) were significantly associated with problem solving when controlling for other predictors in the model. The effect for CA was such that an increase of 2 standard deviations in CA was associated with a predicted increase of approximately 1-point in problem solving scores (B=0.47). The effect for motor cognition was such that an increase of approximately 2 standard deviations was also associated with an increase of approximately 1-point in problem solving scores (B=-0.39).

In the fourth model, functional object use was regressed on CA, visual attention, and motor cognition. This model accounted for slightly more variance in functional object use than model 2 (R^2=0.36; p=0.002) and patterns of significance also varied (see Table 5, model 4). Holding visual attention and motor cognition constant, CA was significantly associated with functional object use (b=0.22; p=0.004). This effect was such that an increase of approximately 2 standard deviations in CA was associated with a predicted increase of approximately 1 addition functional act (B=0.46), which was a significant effect. Visual attention was no longer significantly associated with functional object use when controlling for CA and motor cognition. Motor cognition was also not significantly associated with functional object use.

Mediation model

A cross-sectional mediation model was estimated to test potential developmental patterns consistent with mediation using the PROCESS package for SPSS (Hayes, 2013). Motor cognition and visual attention were examined as potential mediators of the relationship between motor abilities and object-related problem solving in a multiple mediator model (see Figure 6). Results from the model partially supported hypotheses. Motor abilities were found to significantly predict the natural log of motor cognition (*b*=-0.08; *p*=0.023), and the natural log of motor cognition was found to significantly predict object retrieval outcomes (*b*=-0.79; *p*<0.002), indicating that the model was consistent with mediation. Motor abilities were significantly associated with object-related problem solving both before (*b*=0.26; *p*<0.001) and after (*b*=0.19; *p*=0.001) the inclusion of the mediators, supporting results consistent with partial mediation. Results indicated that the indirect effect of motor abilities on object retrieval mediated by the natural log of motor cognition was estimated to be 0.07. A bootstrap sample of 5000 was drawn to determine if this effect was significantly different than zero, and the 95% bias-corrected confidence intervals indicated that it was (95% CI 0.01, 0.16). Results from the cross-sectional multiple mediator model also indicated that motor abilities did not significantly predict visual attention, and thus, the visual attention component of the model was not consistent with mediation. Because the path from motor cognition to functional object use was not significant, a cross-sectional mediation model estimating potential mediating effects on functional use of objects was not estimated.

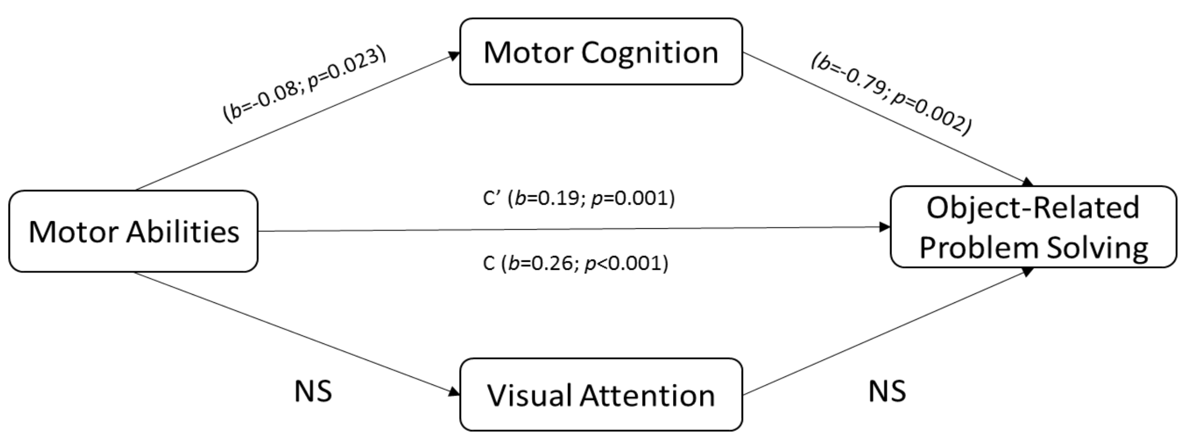

Figure 6
Cross-sectional mediation model

CHAPTER FIVE

Discussion

Summary of findings

This study examined the early developmental foundations associated with object-related planning in toddlers with DS. Two multiple regression models were estimated to test the contributions of motor abilities, visual attention, and motor cognition to object-related problem solving and functional object use. Motor abilities and motor cognition were found to be significantly associated with object-related problem solving, whereas motor abilities and visual attention were found to be significantly associated with functional object use. Cross-sectional mediation analyses indicated support for a pattern of developmental associations between motor abilities and object-related problem solving outcomes. The foundations of early object-related planning identified in this study have potential implications for early intervention in DS.

Object-related problem solving

This study identified specific foundations of object-related problem solving. As hypothesized, motor abilities were significantly associated with object-related problem solving outcomes. In partial support of hypotheses, motor cognition, but not visual attention, was also significantly associated with object-related problem solving. Finally, results identified a potential developmental association between motor abilities and object-related problem solving. Collectively, these findings suggest a pattern of developmental foundations of motor abilities and motor cognition for early EF for toddlers with DS.

Motor abilities. Consistent with previous findings (Fidler, Hepburn et al., 2005), motor abilities significantly predicted object-related problem solving, which suggests that motor development is a critical foundation for this outcome in DS. Although the object retrieval battery requires a basic motor response, it places greater emphasis on successful object retrieval, rather than on motor execution (Fidler, Hepburn et al., 2005). The lack of complexity in motor demands in this task underscores supports the unique contribution of motor foundations to object-related problem solving.

Motor cognition. This study also identified motor cognition as a significant foundation to object-related problem solving. Interestingly, motor cognition predicted object-related planning when controlling for motor abilities. This suggests that the ability to integrate object characteristics into an action plan uniquely impacts object-related problem solving for toddlers with DS. Furthermore, visual attention was not associated with object-related problem solving. Collectively, these findings indicate that, similar to young TD infants, motor cognition has a greater association to object-related problem solving than visual attention for toddlers with DS (Corbetta et al., 2000).

Cross-sectional mediation. The cross-sectional mediation model provides additional support for the importance and unique contribution of motor abilities and motor cognition to object-related problem solving in DS. Consistent with a mediating effect, results indicated that the influence of motor abilities on object-related problem solving partially occurred through motor cognition, which is the pattern of emergence observed within TD infants (Corbetta et al., 2000; Thelen et al., 1995). Specifically, motor abilities were significantly associated with both motor cognition and object-related problem solving, while motor cognition was also associated with object-related problem solving. Thus, despite early motor delays, children with DS come to demonstrate the emergent process observed in TD children at a later age (Fidler, Hepburn et al., 2005; Fidler, Hepburn et al., 2011; Fidler, Lunkenheimer et al., 2011). Results from this model underscore the importance of motor abilities and motor cognition as planning foundations within atypical development. Furthermore, these results offer the opportunity for early and targeted intervention to potentially shape the trajectory of object-related problem solving in DS.

Functional object use

This study also identified different foundational patterns for functional object use, which partially supported hypotheses. Similar to object-related problem solving, motor abilities were associated with functional object use, though this effect was only approaching significance. In addition, visual attention was also significantly associated with functional object use, whereas motor cognition was not. This pattern reflects similar processes within typical development and also has several implications for early intervention in DS.

Motor abilities. Motor abilities were also associated with functional object use, which is consistent with findings from typical development (Lockman, 2000; Keen, 2011). Motor abilities shape functional use of objects through facilitating actions on objects, learning the outcomes of actions, and refining actions accordingly (Corbetta et al., 2000; Libertus & Needham, 2010; Lockman, 2000; Keen, 2011; Needham et al., 2002; Thelen, 1995). TD infants learn about objects and their capabilities for action from direct exploration and practice (Lockman, 2000). Findings from this study indicate that motor abilities serve as a potentially critical foundation for functional object use for toddlers with DS as well, though future work with a larger sample is necessary.

Visual attention. Findings otherwise indicate different patterns in functional object use relative to TD infants, which also has implications for intervention. Specifically, visual attention, but not motor cognition, was significantly associated functional object use. This is somewhat contrary to patterns observed in typical development, which indicate that manipulating objects contributes to motor cognition and bolsters functional object use (Lobo & Galloway, 2013;

Lockman, 2000). Despite demonstrating adequate motor abilities and a significant pathway between motor abilities and motor cognition, toddlers with DS still relied on visual attention over motor cognition for functional object use. Two explanations potentially account for this: a specific developmental stage, or preference for visual processing.

Developmental stage. First, this specific finding may replicate findings from TD infants, which indicate that developmental shifts in visual exploration correspond with sophistication of object-related planning (Baillargeon & DeVos, 1991; Corbetta et al., 2000; Newell et al., 1993). Infants visually explore objects at very young ages, prior to the emergence of motor abilities when they lack sophisticated planning abilities (Baillargeon & DeVos, 1991). Infants then return to visual exploration at an older age (i.e., 8 months), after motor cognition has developed and infants can engage in sophisticated goal-directed planning with less reliance on manual exploration (Corbetta et al., 2000). The findings from the present study demonstrated that increased visual attention predicted increased functional object use in toddlers with DS. Thus, it is more likely toddlers with DS reflect the later developmental shift towards utilizing visual attention for sophisticated object-related planning (Corbetta et al., 2000; Newell, 1993).

Visual preference. A second possible explanation for this pattern is that toddlers with DS compensate for diminished manual exploration with disproportionate visual attention. Infants with DS have been shown to prefer visual exploration over manual exploration, even with adequate motor abilities, which may indicate necessity for greater processing time to acquire the same object information (MacTurk et al., 1985). Toddlers with DS in this study may have relied on visual attention over motor cognition because this is a preferred mechanism for detecting object characteristics required for functional object use. Although additional work is necessary to further disentangle foundations of functional object use, these findings have implications for intervention.

Age as foundation of planning

Results from two additional regression models tested whether CA was an appropriate alternative stronger predictor of planning outcomes than motor abilities and results were mixed. In terms of object-related problem solving, relatively no additional variance in problem solving outcomes was accounted for when substituting motor abilities for CA in the regression model. Patterns of significance also remained the same, suggesting that motor abilities were roughly equated with CA within this model. In terms of functional object use however, findings were much different when motor abilities were substituted for CA in the model. These results indicated that CA was the only significant foundation for functional object use outcomes within this model.

Taken together, these results have potential implications for considering age as a variable within early development in DS. One perspective may be that CA may serve as a proxy for developmental experience to some extent for toddlers with DS. This is supported by findings that motor abilities and CA were roughly equivalent as predictors of object-related problem solving outcomes. This notion is further supported in that CA was the only significant predictor of functional object use. While this provides evidence that development tracks somewhat concurrently for toddlers with DS as it does typically developing toddlers, it negates the additional and nuanced underlying foundations that occur within development (Rutter, 1989; Wohlwill, 1970).

A second perspective is that other developmental foundations may contribute more substantively to evaluating certain outcomes within early development than CA. CA has been referred to as an ambiguous variable with which to study developmental processes (Rutter, 1989), and results here support that notion. Specifically, considering the contribution of motor abilities further delineates what developmental processes beyond simply age and maturation contribute to early cognitive outcomes. Findings from the first two regression models indicate that additional foundations of motor abilities and visual attention are important for planning outcomes. These findings have greater and more specific relevance in terms of implications for intervention than attributing planning foundations primarily to CA. Furthermore, results from age-based correlations also indicate that strength of certain associations fluctuate rather than simply increase with CA in toddlers with DS. Although sub-samples were small, this suggests that there are greater nuances underlying developmental processes than CA can account for.

Implications

Findings from this study have several implications for promoting the early development of planning in DS. Motor abilities, visual attention to objects, and motor cognition were all identified as critical foundations for aspects of object-related planning. In addition, a potential developmental cascade from motor abilities to object-related planning through motor cognition was identified. Collectively, these findings offer the potential for targeted intervention within early development in DS.

Infants with DS generally receive some form of early intervention services that aim to address motor challenges, yet motor delays persist into the second year of life (Periera et al., 2013). Thus, children with DS may benefit from either a higher therapy dosage, or innovations to current therapeutic practices in order to effectively target motor delays. Current findings also demonstrate that targeting early motor development may influence the development of motor cognition along with EF outcomes. Finally, results from this study also indicate that targeted intervention should focus on visual attention to objects along with motor cognition and early motor abilities in order to exact developmental changes to EF.

Limitations

There are limitations to this study that must be considered in the interpretation of findings. First, the design was cross-sectional, yet aimed to investigate the relationship among developmental foundations and mechanisms. Although findings provide preliminary information regarding the pattern and contribution of developmental foundations to planning, longitudinal work is necessary to identify a specific developmental trajectory from motor abilities to planning outcomes. In addition, the sample size was relatively small which may have compromised statistical power in determining the contribution of motor abilities to functional object use. This study also lacked a comparison group, which limits interpretation of findings. Although toddlers with DS in this study were found to demonstrate developmental relationships similar to that of TD infants, the specific nature of delay cannot be determined without direct comparison.

Finally, another limitation to this study that must be considered is the lack of validation for the object-related problem solving battery. The developmental variability observed in DS poses specific challenges to measurement. The age range included in this study (11 to 45 months) was considerably small, yet participants were highly variable developmentally, presenting a challenge for continuity in problem-solving measures across developmental ability. Although the basic and advanced measures in the problem-solving battery had been previously validated in TD infants (Willatts et al., 1999) and toddlers with DS (Fidler, Hepburn et al., 2005; Fidler, Philofsky et al., 2005), the intermediate task was specifically developed for this study. In addition, “basal” and “ceiling” rules were applied from traditional developmental assessment approaches to approximate developmental appropriateness. Future work should attempt to validate this measurement approach and further address these limitations.

Future directions and conclusion

The emergence of object related planning is a critical developmental process in early infancy and toddlerhood. Children with DS are at increased risk for atypical outcomes as a result of motor delays and compromised exploratory opportunity. Early motor abilities, the integration of perceptual information, and visual attention to objects are all critical foundations to early object-related planning outcomes. Although future work is necessary to elucidate the full extent of developmental cascades associated with object-related planning outcomes in DS, preliminary findings identify motor abilities, motor cognition, and visual attention as early intervention targets to improve planning outcomes in DS.

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