THESIS

INFANT EXPLORATION AND CHILDHOOD ACTION PLANNING IN CHILDREN WITH DOWN SYNDROME

Submitted by

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ABSTRACT

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Children with Down syndrome (DS) are predisposed to delays across domains of development and there is a dearth of information on longitudinal associations across early childhood that would help to characterize skill acquisition. Executive functions (EFs) are the thinking and problem-solving skills that direct behavior to achieve goals. Planning is a subconstruct of EF that is an area of relative challenge for children with DS in middle childhood and adolescence. This investigation examined the foundations of planning in DS between infant exploration behavior and emerging childhood planning. METHODS: Forty-six children with DS and their parents participated in two waves of data collection. Infants' first visit was held between 9 and 17 months (M = 12.76 months; SD = 2.16) for Wave 1 and the second research visit was when children were 3 to 7 years old (M = 5.03 years; SD = 0.80) for Wave 2. RESULTS: No significant predictive link was found between infant exploration and early childhood planning. No significant findings emerged between biomedical status and childhood planning. CONCLUSION: Results of this investigation did not identify a predictive link between infancy and early childhood planning. The current study was among the first longitudinal analyses examining development in early childhood for children with DS. Future work should further characterize the heterogeneity observed in children with DS to tailor intervention supports to emerging planning skills.

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INTRODUCTION

Down syndrome (DS) is the most common neurogenetic syndrome associated with intellectual disability (Presson et al., 2013), occurring in approximately 1 of 700 births (Mai et al., 2019). Over the past several decades, research on DS has converged on a pattern of relative strengths and challenges, often called a “behavioral phenotype”. A behavioral phenotype is a probabilistic description of outcomes related to an individual's genotype; however, abilities vary among individuals (Daunhauer & Fidler, 2011). Previous research has characterized a pattern of relative strengths in visuospatial processing and social engagement and relative challenges in motor skills, working memory, and task persistence for children with DS (Daunhauer & Fidler, 2011; Edgin, 2013; Fidler, 2005). Although areas of strengths and challenges have been identified in cross-sectional investigations with individuals with DS, there is a gap in understanding the development of individual trajectories and the precursors that may indicate areas needing additional support. This gap is particularly apparent in early childhood for children with DS because there is limited previous research that has examined developing cognition longitudinally from infancy.

Early skill acquisition can have positive cascading effects throughout development. A developmental cascade occurs when early learning in one domain of development influences how other domains develop, creating long-term impacts on outcomes (Libertus et al., 2016). New insights into the early foundations of skill development in motor and cognition for infants with DS suggest these domains inform how the other domain develops (Cardoso & de Campos, 2015; de Campos et al., 2013; Fidler, Schworer, Prince, et al., 2019; Schworer et al., 2021, 2020). Early intervention to support motor skill building may reduce the time to acquire cognitive skills for children with DS (McConnell, 2000).

The present study will investigate the links between early features of infant cognition and early childhood outcomes related to the critical construct of planning. Planning is a cognitive dimension that is important for activities of daily living and goal achievement across the lifespan. Planning is the organization and sequencing of behavior to achieve goals, including the ability to update behavior when modifications are needed or when the environment around the goal has shifted (Willoughby & Hudson, 2021). There is minimal research on the precursors of planning from infancy in children with and without disabilities. A better understanding of the origins of planning will provide insights for intervention to target these precursors. One potential precursor to planning is exploratory behavior in infancy, which can provide important early opportunities to plan and coordinate behavior across a variety of sensory modalities and motor behavior. This investigation will examine whether the coordination of early exploratory behavior during infancy relates to later planning skills in children with DS.

REVIEW OF THE LITERATURE

Executive Function

Planning is a subconstruct of a group of cognitive skills called executive functions (EFs). EFs direct behavior and action to achieve goals and facilitate day-to-day activities (Carlson, 2005; Diamond, 2013). Using EFs is considered effortful because they require intentional thought and action that originate from top-down mental processes (Diamond, 2013). Top-down regulation refers to whether the frontal lobe has developed to reach and direct other brain regions, which provides greater control of prepotent actions and behavior (Müller & Kerns, 2015). Top-down regulation has been associated with an increased ability to inhibit behavior and flexibly shift between activities, allowing for increased reflective and intentional responses to the environment (Müller & Kerns, 2015). This top-down control matures from early forms in childhood to increasingly complex regulatory behavior that emerges in adolescence and adulthood. As such, EFs are associated with important long-term outcomes, including academic achievement, employment, and improved health, across the lifespan in children and adults with and without disabilities (Best & Miller, 2010; Michaelson & Munakata, 2020; Moffitt et al., 2011; Pellicano, 2012; Tomaszewski et al., 2018; Will et al., 2017).

Factor Structure of Executive Function

Several important issues arise in the study of EF, including an ongoing scientific discussion regarding EF subconstructs and their factor structure. A widely accepted factor model of EF includes three component processes: inhibitory control, cognitive flexibility, and working memory (Diamond, 2013; Miyake & Friedman, 2012; Miyake et al., 2000). These three subdomains of EF are sometimes considered the core EFs, from which higher-order EFs like problem-solving and planning are built (Diamond, 2013). When factor analysis is used to examine EF performance in children with typical development, several models have emerged for the subconstructs that comprise EF. For example, Laureys et al. (2022) identified a fourth factor of planning as another dissociable component of EF in children older than twelve years and a unitary structure of EF for children eleven years and younger. The unitary structure of EF in younger participants aligns with an analysis of preschoolers reported by Wiebe et al. (2011). However, research involving the dissociability of EF in children is divided with evidence for one-factor and two-factor solutions of EF in preschool-age children without disabilities (Karr et al., 2018; Miller et al., 2012; Wiebe et al., 2011).

It is challenging to reconcile the different accounts of EF composition, particularly during the early childhood and preschool years. Unsurprisingly, unitary and two-factor structures were the best fit when the protocol was limited in its measurement of only some of the subconstructs of EF and there were few direct-assessment tasks administered (Miller et al., 2012; Wiebe et al., 2011). The two-factor structure identified in Miller et al. (2012) was better positioned to find a more nuanced factor structure by including assessments of each of the three core subconstructs in the tasks administered, but it was still limited because of its exclusion of planning tasks.

Assessments to capture emerging planning skills are notably missing from each of these preschool-EF investigations. There is limited work to examine how preschooler planning fits into the EF composition at this age. Factor analyses will only be able to identify the EF subconstructs that have construct validity with the tasks administered. In addition to including tasks across EF subconstructs, it is also preferable to administer multiple assessments from the same subconstruct of EF (Willoughby & Hudson, 2021). Preschool assessments often need to avoid extended assessment batteries because they can be challenging for young children and their parents. To address this limitation, complementary measurement tools to direct-assessment tasks should be considered.

Planning in preschool-age children has been analyzed through proxy-report measures. Questionnaire measures provide the opportunity to gather information regarding EF subconstructs across several questions, which addresses some of the challenges associated with limited direct assessment time with preschool-aged children. Preschooler EF can be measured with an ecologically valid proxy report that uses a five-factor structure of EF. This questionnaire is more comprehensive, and planning/organization is included as a discrete subconstruct (Gioia et al., 2003). The use of this questionnaire provides more opportunity to explore evidence of a multicomponent EF developing in preschool.

Factor Structure of Executive Function in Clinical Populations. The research presented above has aimed to characterize the factor structure of EF in children without disabilities. In clinical populations, however, there is mounting evidence for the dissociability of the subconstructs of EF, even during childhood. Investigations using EF proxy reports with parents of children with autism spectrum disorder (ASD) and attention deficit hyperactivity disorder (ADHD) without intellectual disability described elevated challenges in specific subdomains of EF that correlated with the child's diagnosis (Granader et al., 2014; Otterman et al., 2019; Pellicano, 2012). Ozonoff and Jensen (1999) designed a laboratory-based study of EF in children with autism, ADHD, and Tourette's syndrome to examine differences in EF challenges across diagnostic groups. That investigation reported more significant challenges in the specific domains that aligned with the hypothesized difficulties in EF for each diagnostic group (Ozonoff & Jensen, 1999). Specifically, children with autism demonstrated greater difficulty with flexibility and planning, while the children with ADHD and Tourette's syndrome had greater challenges with the inhibition task (Ozonoff & Jensen, 1999). This investigation propelled future work into examining areas of relative strength and challenge within domains for different neurodevelopmental conditions, such as DS.

Research on EF in individuals with DS has largely focused on middle childhood, adolescence, and adulthood. Group-level findings suggest dissociability in EF because patterns of strength and weakness emerge across the subconstructs of EF. These investigations routinely comprise laboratory-based measures and proxy reports from teachers and parents. In school-age children with DS, challenges in working memory and planning/organizing skills are identified relative to strengths in cognitive flexibility and emotional control from caregiver ratings (Daunhauer et al., 2014; Lee et al., 2011; Loveall et al., 2017). Results from laboratory-based studies demonstrate similar areas of challenges with working memory and planning tasks but not shifting or inhibition compared to peers without disabilities at similar developmental levels (Daunhauer et al., 2017). However, only a limited number of investigations have used a comprehensive battery to directly assess EF in children with DS (Daunhauer et al., 2017). In adolescents with DS, cognitive flexibility emerges as an area vulnerable to challenge in addition to persistent difficulties with working memory and planning (Lanfranchi et al., 2010; Loveall et al., 2017). Further research is needed to understand the dissociability and developmental trajectory of EF in DS in early childhood.

Planning

Planning is a subconstruct of EF that is central to adaptive behavior and activities of daily living. It involves organizing thoughts or behavior in a sequence of steps, modifying actions in response to changing demands, and using other cognitive processes (e.g., inhibition and working memory) to support completing tasks and achieving goals (Willoughby & Hudson, 2021).

Planning is used across the lifespan in activities of daily living and increasingly helps guide behavior with the acquisition of advanced skills in cognition, language, motor, and socialization.

Some childhood activities require less planning and preparation because they are structured with routines embedded into the context (e.g., a math lesson at school). Other childhood activities are more open-ended. Open-ended and unstructured contexts provide individuals the opportunity to direct their behavior from a range of possible actions (e.g., free play). Planning is particularly useful in guiding behavior toward a goal in novel, unstructured, and nonroutine situations (Banich, 2009). Individuals use planning in novel and unstructured environments to create goals and direct their behavior toward achieving them.

Planning is closely related to other core constructs of EF, including working memory, inhibition, and cognitive flexibility. Working memory facilitates the maintenance of goal-relevant information when devising and implementing a plan (Diamond, 2006). Inhibition involves stopping a prepotent action by ignoring extraneous stimuli and maintaining selective attention to a plan or goal (Diamond, 2006; Will et al., 2014). Inhibition is important for planning because, in pursuing goals, there is a need to forego short-term rewards in favor of longer-term goal attainment (e.g., earning a degree). The EF component of cognitive flexibility involves the ability to shift attention and behavior when task environments change (Diamond, 2013). Cognitive flexibility is also important for planning, which can require adjusting and updating plans as circumstances change. As a result of this interconnectedness, measures of planning tend to assess multiple aspects of EF.

Measuring Planning

Preschool laboratory-based measures of EF often involve game-based activities wherein children can demonstrate their early use of EF components. The measurement of planning includes different types of game contexts to examine sequenced behavior in the pursuit of goals. One type of planning task is forward planning, which involves organizing behavior to achieve a goal prior to starting the task. Forward planning in preschool-age children includes games such as the simplified Tower of Hanoi task, Truck Loading, or Kitten Delivery (Carlson et al., 2004). These planning games require children to listen to an examiner set up an imaginative play space with rules, and then children should follow those rules to complete the task. Children then respond by sequencing their behavior in orderly and directional ways while using working memory to hold the goal of their actions in mind. Other types of planning assessments may include fluency tests (Troyer et al., 1997) or standardized assessments of planning, such as the Woodcock-Johnson III – COG Planning subtest (Woodcock et al., 2001). Each of these planning tasks requires the consideration of an end goal to inform the selection of a sequence of behaviors. A third type of planning task can involve open-ended play opportunities for children to explore their generativity around novel objects (Fidler et al., 2014). The generativity-style task taps into children's planning in unstructured environments. Each of the planning tasks described above requires non-EF and other requisite EF skills to participate in the task.

An alternative approach to evaluating planning during early development can be through action-planning tasks. Action-planning tasks involve problem solving, inhibition, cause and effect thinking, and sequencing movement toward a goal (Fidler et al., 2005). Action planning tasks reduce demands on working memory, vocabulary, and children's generativity by setting up a simple play space that changes between trials. Action planning requires children to remember one goal, such as retrieving a toy, and continue to work toward that goal across trials. In the meantime, the examiner adjusts the task space, such as changing the direction the child will need to reach to obtain the toy. This type of task requires no expressive language and minimal working memory demands to measure planning skills. When selecting an assessment of planning, it is important to consider the other executive and non-executive processes that may be confounded in the measurement of the task.

Foundations of Early Planning Through Exploration

The central role of planning for adaptive outcomes throughout childhood makes it a critical target for intervention (Fidler et al., 2005; Will et al., 2014). Identifying the precursors of planning during the infant and toddler years may lead to interventions that support more adaptive outcomes in a number of clinical populations. Early exploration experiences offer important opportunities to develop a foundation for cognitive skills later in childhood (Barrett et al., 2008; Fidler, Schworer, Prince, et al., 2019). Through exploration, infants gain an understanding of the physical world and the properties of objects through varied early experiences with objects (Libertus et al., 2013; Rochat, 1989), which leads to sequencing behavior to achieve goals (Schworer et al., 2020). The downstream impact of exploratory behavior during infancy can be observed in studies where early engagement and efficiency with exploration during infancy are associated with better academic achievement in adolescence (Bornstein et al., 2013). Identifying whether there is a link between exploration in infancy and preschool planning in children with specific EF vulnerabilities may provide the opportunity to support children prior to identifying challenges in planning and EF in the toddler and school years.

To date, however, no longitudinal studies have examined the link between exploratory behavior and later planning in children with or without disabilities. Object exploration theoretically does involve early forms of planning in that it involves multimodal (e.g., visual, oral, and physical) sequenced actions on an object (Barrett et al., 2008; Fidler, Schworer, Prince, et al., 2019; Schworer et al., 2020). Multimodal exploration requires the coordination of different exploratory actions to successfully manipulate objects, which leads to an understanding of object properties and affordances (Fidler, Schworer, Prince, et al., 2019). This manipulation is similar to planning because it involves an infant producing intentional and varied movements on objects based on the object's distinct properties (Rochat, 1989).

Exploration and Early Planning.

Infants begin to use different exploration modalities in response to object properties as early as three months of age (Rochat, 1989). Shortly after infants demonstrate responsiveness to the unique features of objects, exploration continues to evolve based on efficiency with reaching and grasping. Infants 4 to 6 months without disabilities demonstrate differentiated exploratory behavior based on their reaching abilities (Libertus et al., 2013). Infants with less advanced reaching spend more time exploring large objects through visual and physical manipulation compared to peers with more advanced reaching who spend more time exploring smaller objects (Libertus et al., 2013). These results suggest that infants select objects in their environments that will be easier for them to obtain. When infants are able to reach independently, they select the objects they can physically manipulate based on their reaching ability (Libertus et al., 2013). This example highlights infants' anticipation of object properties as a way to plan their behavior in ways that enable their exploration of nearby objects. Efficiency with exploration is likely to develop from repeated opportunities to get to know their environments and nearby objects.

An informative approach to measuring infant exploration as an early form of planning is through the analysis of infant behavior in open-ended task contexts. These tasks often allow infants to play with a toy that is set directly in front of them, such as a teether, spoon, or series of balls, for a pre-specified amount of time (Rochat, 1989). Infant behavior can be analyzed for strategy production (actions on the object) and strategy efficiency (the duration/latency to complete actions).

Exploratory tasks often involve infant motor skills, like reaching and grasping, to direct behavior to the object. One approach to measuring early action planning involves presenting infants with objects of different textures, sizes, and rigidity (Barrett et al., 2008). One study by Barrett et al. (2008) aimed to identify how infants represented properties of the different types of balls and subsequently modified (i.e., planned) their exploratory behavior in response to the different object features. Infants between 5 and 15 months used visual information from an object to anticipate how to reach and grasp for objects in different ways (Barrett et al., 2008). Although Barrett et al. (2008) focused on grasping behavior in infants, the task is also an open-ended context to characterize infant exploration strategies and efficiency.

Infant exploration efficiency and strategy production may also emerge alongside developing handedness. Handedness likely develops in a dynamic way through the integration of sensory experience, environmental influences, and genetics (Corbetta et al., 2006). Handedness is the use of one hand in favor of the other for the completion of tasks. Handedness preference may vary toward the end of the first year for a child before establishing a more clearly preferred hand (Corbetta et al., 2006). In examining how children approach exploring objects, taking handedness into account may inform their strategy production.

Planning in Young Children with Down Syndrome

As noted above, a significant amount of research has described EF in childhood, adolescence, and young adulthood in individuals with DS. Relatively less is known about EF emergence during early childhood (Tungate & Conners, 2021; see also Daunhauer et al., 2014, 2017; Lanfranchi et al., 2010; Lee et al., 2015, 2011; Loveall et al., 2017). DS is unique relative to other diagnoses of developmental disability because it is identified prior to or very close to the birth of the child. This allows for prospective investigations of developing trajectories to better understand many aspects of development, particularly cognition. A better understanding of EF developmental trajectories during early childhood, particularly in the area of planning, can inform early intervention.

Very few studies have measured planning in early childhood for children with DS. Considering the importance of planning to adaptive behavior and daily living skills, identifying measures that capture emerging skills is critical to identifying intervention targets. One study exploring planning skills in toddler-age children with DS identified relative challenges in planning efficiency on an action-planning task (Fidler et al., 2005). The action-planning task required the use of motor planning in order to obtain a toy hidden under a three-sided clear plastic box (Fidler et al., 2005). This is one of the only investigations that examined developing planning skills for toddlers with DS and included a task without substantial non-EF requirements in the laboratory task. There is a lack of research that explores the importance of planning at this age for children with DS and the implications from early challenges. Interventions in this sensitive window of development may have the opportunity to bolster EF and support positive outcomes for individuals with DS throughout the lifespan. Longitudinal studies to explore the earliest underpinnings of planning will help identify which children are most vulnerable to challenges with planning.

Foundations of Planning in infants with Down Syndrome

Infants with DS tend to show relative challenges in aspects of action planning compared to infants with typical development (Schworer et al., 2021, 2020). In one study, when infants with DS completed an action-planning task, they were slower on average than the infants without disabilities (Schworer et al., 2020). There have not been any investigations to examine how these differences in infancy develop across early childhood. Understanding if there are longitudinal links in the development of action planning from infancy is necessary to understand what developmental profiles may have greater vulnerabilities to delays.

Exploration in Infants with Down syndrome. Infant exploration in DS has gained attention in recent research, but the implications for developmental outcomes from individual differences in exploration are poorly understood. Time spent exploring objects during infancy provides infants with increased opportunities for experiential learning and understanding the affordances of objects. Children with DS may demonstrate under-responsivity to their environment and low persistence, potentially restricting engagement in learning opportunities through trial and error (Fidler et al., 2005; Kasari & Freeman, 2001). Therefore, limitations in infant exploratory experiences may have lasting implications for developmental outcomes (Fidler, Schworer, Prince, et al., 2019).

A recent study illustrates the connection between different profiles of infant exploration and concurrent cognitive and motor development. This study used latent profile analysis to identify two exploration profiles for infants with DS at a mean chronological age of approximately 9 months (Fidler, Schworer, Prince, et al., 2019). These profiles reflected the presence of active and passive explorers within the sample of infants with DS, with probability of profile membership associated with cognition, motor, and communication performance than the passive explorers (Fidler, Schworer, Prince, et al., 2019). This study highlights that different initial exploration presentations exist among infants with DS. Encouraging active engagement with the infant's environment may promote positive cognitive control and motor skills outcomes.

The measurement of exploratory behavior in infancy may introduce a variety of early trajectories in infants with DS. The task by Barrett et al. (2008), which is described above, is one task that can be used with infants with DS to understand early exploration efficiency in children with DS. In children with DS, this task has been used to examine the latency of infants to reach for different objects, the frequency of reach attempts, and infant swatting behavior (Fidler et al., 2021). This task can also be used to operationalize infant exploration efficiency.

Specific Features that May Influence Planning for Children with DS

In recent years, researchers have applied a developmental cascades perspective to the study of development in individuals with DS. Early planning skills may be one outcome from infants using their motor control to explore their environment (Fidler, Needham, et al., 2019). A developmental cascades approach considers the influences of early learning experiences and how development across skill domains is shaped by opportunities to practice and initiate learning experiences (Fidler, Needham, et al., 2019; Needham et al., 2021). Studying areas of development together, such as cognition and motor skills, can inform how researchers interpret skill acquisition for simultaneously developing domains (Keen, 2011). Early foundations in areas such as motor function may be essential to facilitate the development of exploratory behavior.

Resulting from the interrelatedness of developing cognition and motor skills, delays in motor skills may hinder opportunities to develop skills in planning. Infants with DS tend to show delays in fine motor skill attainment relative to typically developing peers, which has been associated with delayed reach attempts and longer time to begin grasping (de Campos et al., 2013, 2010; Fidler et al., 2005; Frank & Esbensen, 2015; Schworer et al., 2021). This delay in infants' reaching stems from a delay in developing motor skills, but prevents an infant from having exploratory opportunities with the objects in their environment. This delayed engagement in exploratory behavior may cascade into further delays in developing cognition and EF, including planning (Fidler et al., 2011). Furthermore, given the delays in motor skill acquisition observed in many infants and young children with DS, researchers should carefully consider any motor demands involved in cognitive assessment tasks.

Vulnerabilities from Co-occurring Conditions in Down syndrome

Several biomedical risk factors frequently co-occur in children with DS, and these conditions may confer vulnerability to more pronounced developmental delays in executive function and motor skills. These co-occurring conditions may be a source of heterogeneity within this population, potentially explaining differences in planning skills for children with and without the most commonly occurring conditions. Congenital heart defects (CHD) and prematurity are two biomedical conditions frequently associated with developmental outcomes (Alsaied et al., 2016; Aoki et al., 2018; Fidler, Schworer, Will, et al., 2019; Onnivello et al., 2022; Visootsak et al., 2013). A CHD occurs in 44.0-55.9% of children with DS (Freeman et al., 1998; Torfs & Christianson, 1998). CHDs have been associated with language, cognition, and motor development delays relative to children with DS without CHD in early childhood (Alsaied et al., 2016; Visootsak et al., 2013). The long-term outcomes of CHDs on development in the general population are mixed, often with the early delays associated with the CHD no longer evident in cross-sectional analyses by school age (Alsaied et al., 2016; Rosser et al., 2018). Prematurity occurs in approximately 18% of children with DS (Aoki et al., 2018). One investigation found evidence of the negative association between prematurity and an infant's latency to shift attention (Fidler, Schworer, Will, et al., 2019). From infancy to early childhood, the ways that early biomedical conditions may confer risk for more delayed outcomes have not been fully explored.

There is very little research that examines developmental trajectories associated with co-occurring conditions in DS. One recent longitudinal analysis demonstrated an increased link between younger gestational age and ADHD in children with DS (del Hoyo Soriano et al., 2020). Another recent paper identified longer infant visual inspection and greater general sensory processing dysregulation as predictors of ADHD, ASD, and EF dysregulation symptomatology during early childhood (Fidler et al., 2023). Increasing the number of longitudinal studies in this area will provide a better-informed understanding of co-occurring conditions associated with developmental vulnerabilities. This will support interventions that can be targeted to children in need of greater support and tailor existing intervention supports to meet the individual trajectories of children with DS.

CURRENT STUDY

Early delays in exploratory behavior in infants with DS may lead to long-term challenges with EF, particularly with planning. The present study extended prior work by investigating the predictive associations between infant object exploration and early childhood planning. Participants were drawn from a study of early cognitive control in infants with DS 4 to 17 months (Fidler et al., 2021; Fidler, Schworer, Will, et al., 2019; Onnivello et al., 2022; Schworer et al., 2021, 2020). Children participated in two waves of data collection. Wave 1 included an early exploration task for infants 9 to 17 months old. Participants were reassessed with a motor-planning task at Wave 2 between the ages of 3 and 7 years. Additional analyses examined whether biomedical risk confers vulnerability for difficulties with early childhood action planning.

METHODS

Procedures

The current study examined the association between early exploratory behavior during infancy and performance on an early childhood motor planning task in young children with DS. This research received ethical approval from the Institutional Review Board at Colorado State University for all procedures. Informed consent was obtained before beginning any direct assessment procedures at the start of each wave. Caregivers completed several questionnaires regarding their child's adaptive skills, behavior, and demographics. Enrolled children participated in various games that measured early cognitive control and EF.

Study data were collected and managed using REDCap electronic data capture tools. REDCap (Research Electronic Data Capture) is a secure, web-based application designed to support data capture for research studies, providing: 1) an intuitive interface for validated data entry; 2) audit trails for tracking data manipulation and export procedures; 3) automated export procedures for seamless data downloads to common statistical packages; and 4) procedures for importing data from external sources (Harris et al., 2009).

Participants

Forty-six infants (M= 12.76 months, SD= 2.16) participated in a cognitive assessment at Wave 1. Infants and their families were recruited nationally through flyers and advertisements distributed by local DS clinics, associations, support groups, and community boards. Infants participated in evaluations between 4.0 and 17.0 months and participated in a second visit 6-months later. Wave 1 data were drawn from the research visit when infants ranged in age from 9 to 17 months old. At Wave 2, children were assessed between the ages of 3 and 7 years (M =

5.03 years, SD= 0.80). Approximately 54% of the participants were male. Most participants were White (90.2%) and non-Hispanic (84.6%).

The time between Wave 1 to Wave 2 varied across participants, with an average time between visits of 3.98 years (SD = 0.80 years, Range = 2.47 – 5.74 years). All Wave 1 visits were complete prior to the onset of the COVID-19 pandemic. During 62% (n = 27) of Wave 2 visits, examiners implemented safety protocols to prevent the spread of COVID-19. Safety procedures included prioritizing family comfort to have a research visit and the examiner wearing a combination of face masks, face shields, and scrubs. The combination of safety procedures used at each visit reflected concurrent institutional and municipal guidelines.
It is noted that a subset of the enrolled infants at Wave 1 (n = 13) participated in a brief, parent-mediated intervention to support early reaching behavior (Fidler et al., 2021). Intervention enrollment was evaluated as a potential covariate in analyses.

Table 1
Demographic Information

Child variable	% (n)
% Male	54.3 (25)
Child Chronological Age (CA)	
Wave 1(months; SD; n= 1 missing)	12.76 (2.16)
Wave 2 (months; SD)	60.39 (9.63)
Child Mental Age (MA)	
Wave 1 (months; SD)	9.45 (1.88)
Wave 2 (months; SD)	26.86 (5.85)
Race (n= 5 missing)	
Asian-American	2.4 (1)
Black/African American	2.4 (1)
White	90.2 (37)
More than one race	4.9 (2)
Ethnicity (n=7 missing)	
Hispanic	15.4 (6)
Not Hispanic	84.6 (33)
DS Type (n=8 missing)	

Trisomy 21	92.1 (35)
Mosaicism	5.3 (2)
Translocation	2.6 (1)
Premature Birth (% yes; n=6 missing)	32.5 (13)
Congenital Heart Defect (% yes; n=6 missing)	72.5 (29)
Caregiver variable	
Primary Caregiver Age (Mean/SD; n= 5 missing)	39.12 (5.45)
% Primary Caregiver Education at least 1 year of college/tech training (n; n=6 missing)	87.5 (39)
% Annual Income (n; n=5 missing)	
Below $50,000	4.9 (2)
$50,000-100,000	29.3 (12)
Above $100,000	65.9 (27)

Measures

Caregiver Report Questionnaires

Caregivers completed questionnaires about their child's development at each research visit. The focus of questionnaires included family demographics and reporting on child behavior.

Family Background Questionnaire (Wave 2). The family background questionnaire requests information about several demographic variables. This includes reporting the child's race and ethnicity, caregiver age and education, and family income.

Medical History Questionnaire (Wave 2). The medical history questionnaire includes information on potential diagnoses the child may have or had from birth to Wave 2, including vision impairment, hearing difficulty, gestational age, CHD, sleep problems, and other biomedical conditions (e.g., thyroid problems, gastrointestinal issues, Leukemia, head injuries, or seizures).

Developmental Status

Bayley Infant Scales of Development Third Edition (Bayley-3; Wave 1; Bayley, 2006)**.** At Wave 1, infants completed the Bayley-3, a measure of cognition, communication, and motor skills for children 1 to 42 months (Bayley, 2006). This measure has high internal consistency (.86-.93) and test-re-test reliability (.80-.87; Bayley, 2006). A trained graduate student administered the cognitive and motor scales of the Bayley-3 assessment. Raw scores were transformed into scaled scores that compare relative developmental status to children of the same chronological age.

Stanford-Binet 5th edition Abbreviated IQ (SB5-ABIQ; Roid, 2003a). At Wave 2, child participants completed the SB5, a direct assessment measure of cognition that is standardized for individuals 2 to 85 years old. Participants completed two routing subtests, one for Nonverbal Processing Ability (Object series/Matrices) and a second for Verbal Knowledge (Vocabulary; Roid, 2003a). Raw scores are transformed into IQ estimates and age-equivalence scores. The SB5-ABIQ has high reliability with the other IQ scales of the SB5 (+.90; Roid, 2003b).

Planning Tasks

Infant Motor Planning Task (Wave 1; Barrett et al., 2008; Fidler et al., 2021). An early motor planning task was administered at Wave 1 that involved presenting infants with four balls with different textures, sizes, colors, and rigidity. The different characteristics of each ball changed the required reach and grasp that an infant could use to retrieve the ball. One ball had indentations (craters); one ball had protrusions (nubs); one ball had many soft ridges (squidgy); and one ball was comprised of many strings connected at the center (koosh). The examiner presented each ball to the infant, one ball per administration, for up to 30 seconds. Each ball was presented by the examiner with a flat hand to prevent the modeling of grasping behavior for each ball. Trained coders observed the amount of time between the presentation of each ball to the child's contact of the ball. This amount of time was designated as the child's "latency-to-contact" the ball. Coders also scored each time the infant used exploratory actions (e.g., swats, incidental touches, and grasping) with the behavioral coding procedures described in Appendix A. The interrater reliability between coders for this scheme was Kappa = .78.

The outcome variables from this task included dimensions of both strategy production and the efficiency of exploration. Wave 1 "Strategy Production" was defined as the average number of exploratory behaviors an infant produced across all four ball presentations. Strategy production behaviors that were coded included swatting, grasping, and touching of the object. The second variable of interest was Wave 1 "Exploration Efficiency," which was defined as the average latency (in seconds) to make initial contact with each of the balls. For 3 participants, at least one of the four trials was missing due interruption of administration by external factors and child fussiness. To address missingness, trial data were not imputed, rather, the denominator for the average calculation was adjusted to reflect the number of trials completed by each participant.

Object Retrieval Task (Wave 2; Fidler et al., 2005). At Wave 2, child participants completed an object retrieval task, which is a measure of action planning. This task involved placing a small toy animal inside a clear, three-sided box. Participants were encouraged to retrieve the toy from the box, and the opening was re-positioned for each trial. The task included 2 practice and 15 test trials. During the practice trials, children were provided with a demonstration of how to reach for the toy and directly retrieve it from the box if they did not spontaneously reach for the toy after the examiner gave an enthusiastic instruction, "Grab it!". Examiners used an encouraging affect and positive facial expressions to encourage task completion and participant motivation. Feedback to the child from the examiner was provided up to two times during each of the teaching trials. Tactile feedback may also have included a reminder to immediately remove the toy and not play with it in the box. Prior to administering this task in assessment visits, examiners were trained to a standardized task procedure. Examiners met a minimum of 90% agreement to the specified task administration procedure before assessment in research visits.

Trained coders evaluated child responses across each trial to measure the time to retrieve the toy, response strategies, and reach quality (see Appendix B for the complete coding scheme). Research assistants were naïve to the hypotheses of the study and conducted behavior coding via Noldus Observer XT software (Noldus Information Technology, 2013). Two raters overlapped to score at least 30% of the same videos. Coders had good interrater reliability for frequency (Kappa = 0.82) and duration codes (Kappa = 0.92).

There were two primary outcome variables at Wave 2 from the object retrieval task that captured strategy production and efficiency. Childhood "Strategy Production" was defined as the average reach quality score for each participant. Reach quality was operationalized on a scale from 1 to 4. A score of 1 indicated the participant reached through the top of the box before searching through a side, whereas a score of 4 meant the participants reached directly through the open side to retrieve the toy. Wave 2 "Planning Efficiency" was defined as the average length of time the participant took to retrieve the object from the box across trials. Eight participants (17.4%) could not participate in the object retrieval task. Examiners cited reasons for missingness as elopement (n = 1), opposition (n = 3), and missing administration due to time or participant fatigue (n = 4).

Handedness Observation (Wave 2). For the first trial of the Object Retrieval task, the placement of the box was always oriented with the opening facing the participant. Coders observed which hand was used to reach for the toy on the first trial. This reach choice was included in post hoc analyses as a likely indicator of hand dominance.

ANALYTIC APPROACH

Multiple regression analyses were used to estimate the longitudinal association between infant exploration and early childhood planning. The first regression analysis evaluated the effect of infant strategy production (average number of exploratory behaviors) on childhood strategy production (average quality of reach strategy). The second regression analysis examined the effect of infant exploration efficiency (average latency to contact) on childhood planning efficiency (average latency to retrieve). The association between intervention enrollment and the four primary outcome variables was examined.

A small sample size limited the ability to examine outcomes within a Null Hypothesis Statistical Testing (NHST) framework. To facilitate preliminary interpretations from this longitudinal sample, effect sizes were interpreted in addition to p-values (Cumming, 2013, 2014; Fidler et al., 2021). A small sample size affected the assumptions for linear regression. Assumptions of variable normality and distribution of the residuals were violated by the predictor and outcome variables. Analytic transformations for these variables were explored.

Two sets of t-tests were conducted to investigate the association between two biomedical conditions, prematurity and CHD, to action planning in childhood. The first tests compared childhood outcomes for participants born prematurely and those born at full term to Wave 2 Strategy Production and Wave 2 Planning Efficiency. The second tests compared children with and without a CHD on the Wave 2 outcome variables.

Post-hoc analyses were conducted to examine whether child handedness influenced the quality of reach strategy on the object retrieval task. A second set of post-hoc analyses examined the associations of overall development to these measures. To examine the association of developmental continuity, a t-test was used to compare strategy development with z-scored results from infancy and childhood strategy production. Second, a Spearman's ρ correlation matrix was used to examine associations between scores on the Bayley-3 at Wave 1, the SB5-ABIQ at Wave 2, and performances from the Infant Motor Planning task and Childhood Object Retrieval Task.

Complete missing data from Wave 1 and Wave 2 were left as missing due to the small sample size and dearth of information in this developmental window to guide a missing imputation procedure. Partial missingness (e.g., a missing trial) was addressed through adjusted denominators to calculate average strategy production and efficiency scores at Wave 1 and Wave 2 for each participant.

RESULTS

Descriptive Analysis of Task Performances

Wave 1 Infant Motor Planning Task

Thirty-seven infants completed the Infant Motor Planning task. Descriptive summaries of infant exploratory behavior performance are reported in Tables 2 and 3. These tables include the average frequency of exploratory behaviors (e.g., touches, swats, and grasps) and the average latencies to contact the four toy balls. Histograms for each of the exploratory behaviors included in the Wave 1 strategy production variable are shown in Figure 1. The average total number of swatting, touching, and grasping behaviors observed during the Infant Motor Planning task was 4.62 (SD = 1.69; Range = 2.00-8.50). All infants produced at least one exploratory behavior.

Table 2
Infant Early Motor Planning Task

Type of Ball	Mean Latency to Contact (seconds; SD)	Range (seconds)
Koosh	3.37 (3.58)	0.57 – 15.38
Orange Crater	3.00 (4.02)	0.43 – 19.55
Squidgy	2.16 (2.28)	0.40 – 11.64
Yellow Nubby	2.83 (3.25)	0.33 – 18.32

Table 3
Infant Early Motor Planning Task

Exploratory Behavior	Koosh Mean Occurrence (SD; Range)	Orange Crater Mean Occurrence (SD; Range)	Squidgy Mean Occurrence (SD; Range)	Yellow Nubby Mean Occurrence (SD; Range)
Mouthing	0.56 (0.97; 0-4)	1.08 (1.09; 0-4)	1.26 (1.37; 0-5)	0.84 (1.08; 0-4)
Swatting	0.18 (0.51; 0-2)	0.46 (0.93; 0-4)	0.33 (0.87; 0-4)	0.45 (0.92; 0-4)
Touching	1.49 (1.05; 1-6)	2.24 (1.50; 0-6)	1.62 (1.02; 1-5)	1.58 (0.98; 0-5)
Grasping	2.56 (1.67; 0-8)	2.65 (1.74; 0-8)	2.31 (1.32; 0-6)	2.29 (1.66; 0-7)

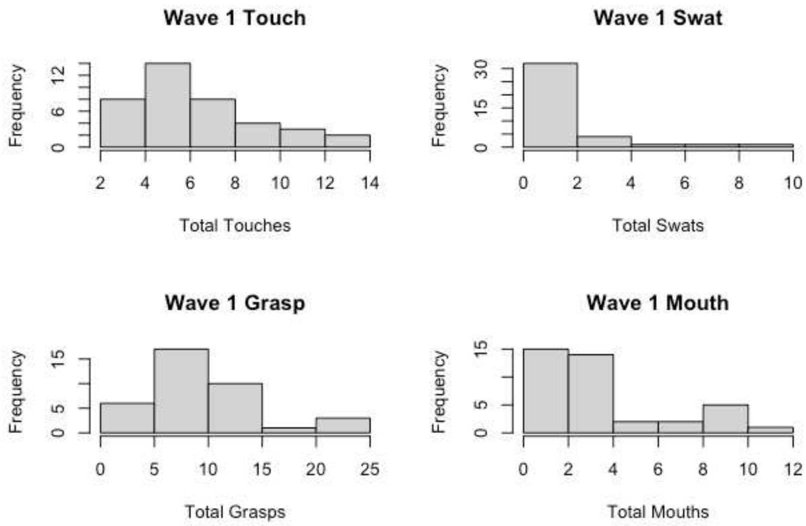

Figure 1: *Histograms of Exploratory Behaviors at Wave 1*

The average latency to contact toys during the four trials at Wave 1 was 2.78 seconds (SD = 2.21; Range = 0.68 - 10.82). However, there was a wide range of performances on this dimension, including latencies as short as 0.33 seconds and up to 19.55 seconds. No meaningful association was observed between Wave 1 average exploratory behaviors and Wave 1 latency to contact (*r* (35)= -.18; *p* = .29; see Figure 2).

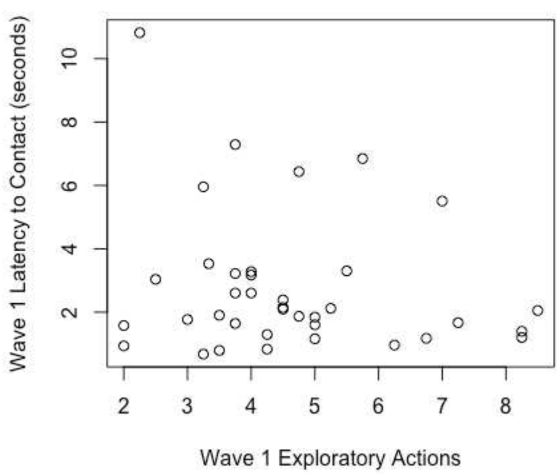

Figure 2: *Scatterplot Wave 1 Average Latency to Contact and Average Exploratory Behaviors Used*

Wave 2 Childhood Object Retrieval

Thirty-eight children completed the Object Retrieval task at Wave 2 and 90.1% of children successfully completed more than half of the trials. A majority of participants (60.5%; n = 23), had successful retrievals on all trials. Eleven participants (29.6%) successfully retrieved the toy on 60 to 90% of the trials completed. Only four participants (9.9%) retrieved the toy on fewer than 50% of trials.

Although rates of successful toy retrieval were relatively high in this sample, children used a variety of response and reaching strategies to complete the task. The response strategies included attempts to manipulate the box (e.g., trying to lift it) on at least one trial by 55.26% of participants (n=21), changing the hand the child was reaching with during a trial attempt by 68.42% of participants (n = 26), using their second hand to stabilize the box by 55.26% (n = 21) of participants, and all participants used a strategy that included contacting the wall of the box on at least one trial (100%; n = 38). A subset of the participants (47.37%; n= 18) also demonstrated other behaviors during the retrieval (e.g., playing with the toy in the box prior to removal).

The mean reach quality score was 2.14 out of 4 (SD = 0.78; Range = 0.33-3.41) across the sample. The average time for the participant to retrieve the object was 5.50 seconds (SD = 2.05; Range = 2.49-10.41). The childhood dimensions of average reach quality and latency to retrieve were significantly negatively correlated (r (38) = -.699, p <.001; see Figure 3). Therefore, children who took less time to retrieve the toy were likelier to use more parsimonious action plans than children who obtained the toy more slowly.

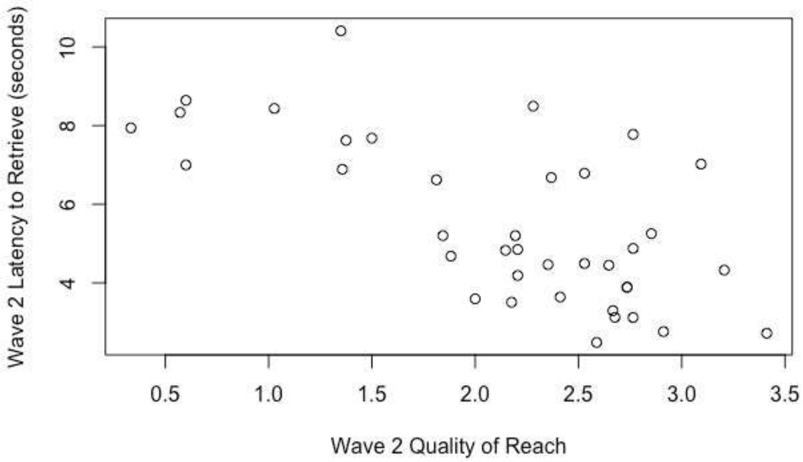

Figure 3: *Scatterplot of Wave 2 Average Latency to Retrieve and Average Reach Quality of Retrieval*

Nineteen children reached with their right hand (50%), and 19 children reached with their left hand (50%). None of the participants attempted to use both hands to bimanually grasp and retrieve the toy on the first trial.

Exploration during infancy as a predictor of childhood planning

From the overall sample of 46 children, 31 completed both the Wave 1 and Wave 2 tasks. This subgroup was included in longitudinal analyses. The average age of participants in the longitudinal analyses was 13.00 months (SD = 2.08) at Wave 1 and 5.02 years (SD = 0.67) at Wave 2. Multiple regression was used to estimate the effects of exploration during infancy on childhood planning strategy. Enrollment in the infant micro-intervention was not meaningfully or significantly related to the four primary variables of interest (*r*s =.04 -.12; *p*s = .3 - 1.0). Intervention enrollment was not used as a covariate in regression analyses. The assumption of normality for multiple regression was violated for both predictor and outcome variables. Attempts to transform these variables included polynomials, Box-Cox, and log functions. None of the transformations normalized the skewed variables. Several additional regression parameters were violated, including homogeneity of variance, linearity, and the normality of residuals as observations neared the ends of the distribution. Results should be interpreted in context of these challenges.

Strategy Production. The first regression model examined whether the average number of exploratory behaviors used during infancy was predictive of childhood average reach quality. No predictive association was observable in the regression model (F (1, 29) = 3.04, $p = .092$, $R^2 = .095$; see Table 4 and Figure 4). The unique effect of average exploratory behaviors was moderate and positive, but was not statistically significant (b = 0.31; $p = .09$).

Table 4
Regression Table of Wave 2 Strategy Production and Retrieval Efficiency

Model	Predictors	β	*p*	B	Standardized Confidence Interval		R^2	F	*P*
					Lower CI	Upper CI			
Wave 2 Strategy Production							0.095	3.04	0.092
	Intercept	0	<.001	1.55	-0.36	-0.36			
	Wave 1 Strategy Production	0.31	0.092	0.13	-0.05	-0.67			
Wave 2 Retrieval Efficiency							0.0016	0.045	0.83
	Intercept	0	<.001	5.43	-0.37	-0.37			
	Wave 1 Exploration Efficiency	0.04	0.83	0.04	-0.34	-0.42			

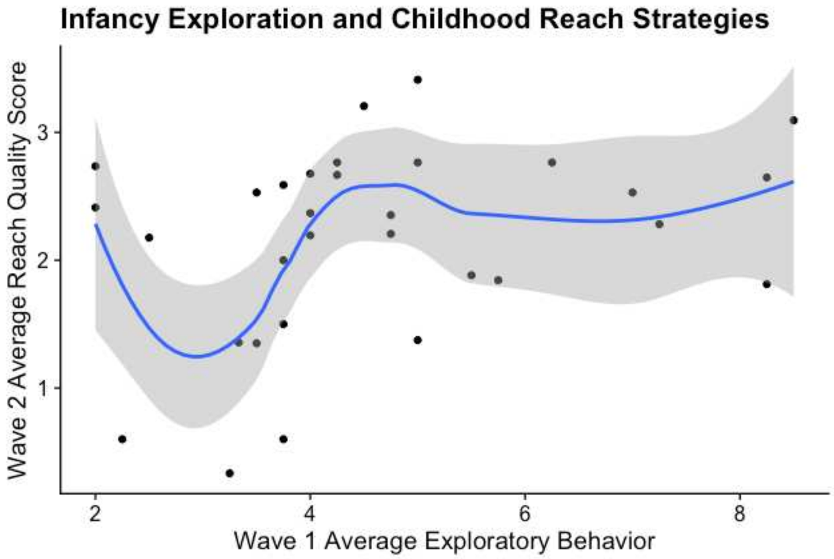

Figure 4: *Regression Plot of Wave 1 Exploratory Behavior and Wave 2 Strategy Production*

Efficiency. The second regression model examined whether average latency to contact a toy during infancy was predictive of the average childhood retrieval time. There was no observed predictive association from infancy to childhood in this regression model (F (1, 29) = 0.045, p = .83, R^2 = 0.0016; see Table 4 and Figure 5). The unique effect of the average latency to contact in infancy was negligible (b = 0.04; p = .83).

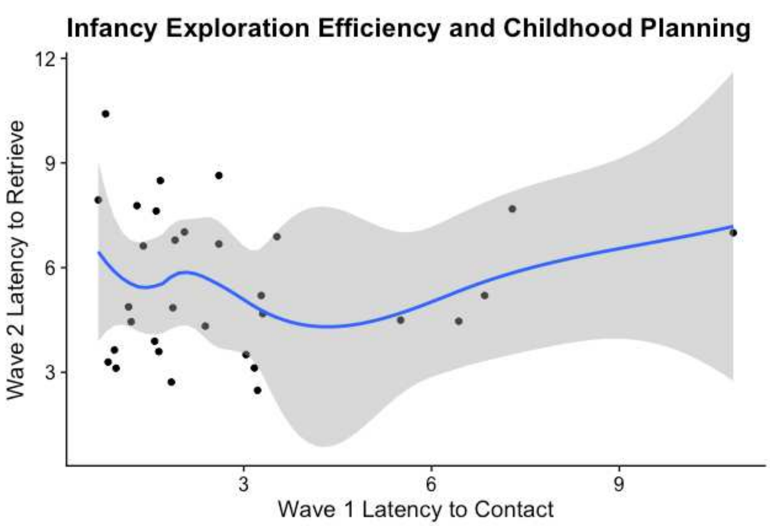

Figure 5: *Regression Plot of Wave 1 and Wave 2 Strategy Efficiency*

Prematurity and Early Childhood Retrieval Strategy and Efficiency

Prematurity status did not have a meaningful effect on the childhood outcomes of average reach quality or latency to retrieve. The comparison of mean childhood reach quality score by prematurity status was nonsignificant $t(31) = -.13$, $p = .90$. Similarly, the comparison of childhood retrieval latency by prematurity was nonsignificant $t(30.41) = 1.52$, $p = .14$.

Congenital Heart Defects and Childhood Retrieval Strategy and Efficiency

There were no statistically significant differences between childhood outcomes of reach quality or average latency to retrieve the toys based on CHD diagnosis ($t\ (31) = -.27$, $p = .79$). Similarly, the comparison of childhood latency to retrieve the toy did not differ between groups with and without a CHD ($t\ (31) = -.17$, $p = .86$).

Post-hoc Analyses

Handedness. The hand the participant reached with in the first trial of the Object Retrieval task was used to examine the effect of hand dominancy on action planning performance. First trial hand preference during the childhood Object Retrieval task was not associated with the average reach quality that was observed in childhood ($t(26.86) = 1.110$, $p= .28$). A similar result was found such that children's preferred hand did not affect childhood average latency to retrieve the toy ($t\ (36) = -0.51$, $p = .62$).

Developmental Status. A paired t-test was conducted to examine the continuity in participant's performance rank relative to their peers from infancy to childhood. This analysis compared the z-scores of observed performances on both strategy production variables, Wave 1 Strategy Production and Wave 2 Strategy Production. The results of the t-test were nonsignificant $t(30) = -0.068$, $p = .95$.

Additional exploratory analyses to examine the potential impact of cognition were conducted via Spearman correlations. These correlations examined cognitive scores at each wave and chronological age at each wave with each target outcome variable at Waves 1 and 2. Bayley-3 Scaled Score Cognitive performance was associated with chronological age at Wave 1 (r= -.45; p= .04) and infant Exploration Efficiency (r= -.56; p= .01) at Wave 1. All other associations were nonsignificant (see Table 5).

Table 5

Spearman's Rho Correlations Table of Developmental Sensitivity and Strategy and Efficiency Outcomes

	1	2	3	4	5	6	7	8
1. Wave 2 CA	-							
2. Wave 1 CA	0.097	-						
3. W1 Exploratory Efficiency	-0.19	0.013	-					
4. W1 Exploratory Strategy	0.13	0.33	0.003	-				
5. W2 Reach Efficiency	-0.00099	-0.13	-0.002	0.036	-			
6. W2 Reach Strategy	0.31	-0.0049	-0.2	0.31	**-0.53***	-		
7. W1 Bayley-3 Cognitive	0.29	**-0.45***	**-0.56***	-0.083	0.022	0.1	-	
8. W2 SB5-ABIQ	-0.28	0.26	0.019	0.12	-0.25	0.27	-0.16	-

*Note = *p < .05; CA = Chronological Age*

DISCUSSION

This study was the first examination of potential continuity in early development related to planning in children with DS. This longitudinal study examined the link between infant manual exploratory behavior and childhood action planning in a sample of young children with DS. The two dimensions of interest were the strategies that participants produced to complete planning tasks and their efficiency in task completion. Planning is an essential skill for the development of goal-directed behavior, but it is a noted area of vulnerability for children with DS (Daunhauer et al., 2014; Fidler et al., 2005; Loveall et al., 2017). Overall, regression analyses did not provide evidence for a linear association between infant exploratory behavior and subsequent childhood action planning. Descriptive exploratory analyses identified meaningful developmental differences in children with DS that emerge as early as infancy.

Examining the association between infant and childhood performances

Strategy Production

A key finding of this study was the lack of association between infant exploration strategies and childhood reaching strategy production. The strategy production regression model did not meet the threshold for statistical significance in an NHST framework with alpha values below .05. However, it is notable that the magnitude of the unique effect in the regression model was medium. Despite the lack of statistical significance, a moderate effect size is suggestive of a potential link between strategy production in infancy and childhood. If replicated, this finding suggests that an increase in the average number of exploratory behaviors used during infancy could predict more competent reach quality in childhood action planning. The interpretation of the magnitude of this effect may demonstrate continuity in planning skill development. However, the present study cannot be conclusive on the possibility of continuity.

One key feature of this investigation was the longitudinal follow up for children with the neurogenetic condition of DS. Investigations more recently have found ways to increase sample sizes in their results through collaborations, but a frequent challenge in research with neurogenetic conditions is a smaller sample size. Smaller sample sizes pose challenges when using a NHST framework because several assumptions of normality are harder to attain with a smaller number of observations. The interpretation of the magnitude or effect size of strategy production in the regression model provides initial evidence for developmental continuity in infant exploration and childhood planning. Exploring alternate approaches to linear analytic frameworks could provide additional nuance to interpreting the developmental trajectories of the heterogenous population of children with DS.

A starting point for examining the hypotheses was the visualization of the variable distributions and the regression results. First, the examination of the distribution of Wave 1 strategy productions (from Figure 2) demonstrates that a smaller number of infants used either very few or many exploratory actions on average. A linear analytic approach may obscure some of the nuance in infant strategy production as a result of restricted range at the two ends of the distribution. The results from this first regression model should be examined with non-linear approaches to understand if there were more systematic differences between subgroups of children with DS.

It may be the case that early planning follows a non-linear trajectory of development. An alternate analytic strategy may be to consider a quadratic association between infant exploratory behavior and childhood action planning. Figure 4 demonstrates a plateau in the association between exploratory behaviors in infancy and more parsimonious reach quality in childhood. For infants who produced under 5 exploratory behaviors at Wave 1, there appears to be a positive association between the average number of infant exploratory behaviors and reach quality in childhood. In contrast, infants who produced 5 or more exploratory behaviors at Wave 1 did not see a linear increase in children's reach quality scores. A quadratic model may capture that a large proportion of this sample had attained developmental competency in early exploratory behaviors (e.g., incidental touch, swats, and grasps) and reach quality was similar across individuals meeting that threshold (5 or more exploratory behaviors). This visual interpretation of the data from Figure 4 suggests an optimal number of exploratory behaviors per object was 4 to 5 as a predictor of higher quality reach strategy at Wave 2. A quadratic analytic approach may indicate that there is a threshold of exploratory behavior that provides a foundation for better reach quality during childhood action planning.

The interpretation of a nonlinear pattern of results fits a majority of observations in this sample of children with DS. However, there were three infants with outlier performance on the exploration task. Each of these infants used only 2 to 3 exploratory behaviors per ball and had average reach quality scores at Wave 2. These advanced reaching scores in childhood were different from what would have been expected with so few exploratory actions. In further exploring these cases, it was determined these infants were not the youngest in the sample, nor did they have the greatest delays in developing cognition. It may be the case that this group was relatively more advanced at Wave 1 because each of these infants used at least one efficiently made grasp to reach for each object. Participants who efficiently grasped and explored the object with only 2 to 3 behaviors may have engaged in longer sustained play with the toy relative to an infant who has a less mature grasp and needed to contact and release the toy more frequently.

The exploration task outcome variables that have been selected for the majority of the sample, may not have been an appropriate measure of developing strategy production and efficiency for these infants with more mature grasping and cognitive skills.

In summary, linear multiple regression may not have adequately captured the association between infant and childhood strategy production. It is possible that exploration serves as a foundation for the development of planning skills in children with DS in a nonlinear fashion. The pattern of observed results may indicate an optimal range and maturity of grasping as predictors of planning skills in early childhood.

Exploration and Planning Efficiency

A second regression model examined the association between efficiency during infancy and childhood. Efficiency was operationalized as average infant latency to contact the toy and average childhood latency to retrieve the toys. Once again, no meaningful associations were observed between infant exploratory efficiency and childhood planning efficiency. The magnitude of effect in this regression model was negligible. This lack of association may indicate discontinuity in development between infant exploration efficiency and childhood reaching efficiency in developing action planning. A visual examination of the scatterplot of results, shown in Figure 5, does not reflect an identifiable pattern of continuity from infant exploration efficiency to childhood planning efficiency. Whereas there may not be group level patterns, similar to the multiple potential trajectories of strategy production, there may subgroups of children who demonstrate different patterns of efficiency in task completion.

Although no group level associations were observed, some insights can be gained from examining subgroups of participants in the scatterplots. For example, one subgroup of infants took more time to retrieve the toys at Wave 1, but a near average time to retrieve the toys at

Wave 2. This group might have been comprised of infants who had more pronounced motor delays and may have received more intensive physical and occupational therapy intervention from infancy to early childhood that addressed efficient reaching skills. Along these lines, Onnivello et al. (2022) reported that infants with DS with the most pronounced delays were more likely to participate in occupational therapy. Access to physical and occupational therapy interventions would have provided more opportunities to practice skills such as reaching and grasping. This extra practice may have indirectly supported developing cognitive planning skills as a result of the motor to cognition developmental cascade (Keen, 2011).

A second subset of participants demonstrated more infant exploration efficiency as observed with shorter latencies to contact the ball at Wave 1, but less development of childhood planning efficiency as observed with longer average latencies to retrieve the object during the Wave 2 Object Retrieval task. This subgroup of participants may have a developmental trajectory with a slower emergence of delays relative to peers with DS. This group could have benefitted from anticipatory intervention, however, these children may have just started receiving additional services from early intervention or school when their delays became more pronounced. The sample size of the current investigation does not allow for answering questions about the trajectories of these subgroups from infancy to early childhood. However, the exploratory observations may provide additional support for recent evidence of within-syndrome heterogeneity in infants with DS (Fidler, Schworer, Prince, et al., 2019; Onnivello et al., 2022).

Although hypothesizing about developmental trajectories from the subgroups in this sample may inform future directions, the results from this analysis are inconclusive. The null results in a NHST framework may have been the product of the small sample size, which is inadequate to parse subgroup heterogeneity in DS. It could also be the case that this is an area of development without a direct association to later childhood outcomes, or the measures inadequately captured these emerging cognitive skills. The pattern of results warrants further investigation of subgroup patterns of performance as it relates to developmental continuity from infancy to childhood planning.

Biomedical Factors and Developing Action Planning

In the current study, no differences were observed in childhood strategy production or efficiency for children born prematurely or with a CHD relative to their peers who were not born prematurely or with a CHD. Findings in the literature related to the developmental impacts of prematurity and CHD are complex and mixed. Part of this complexity stems from the operationalization of these biomedical conditions, where they are narrowly defined in some investigations and more broadly defined in others. For example, in one investigation with children with DS, prematurity was assessed based on the gestational age of the child, which was associated with a higher likelihood of elevated ADHD symptomatology (del Hoyo Soriano et al., 2020). Prematurity, used as a dichotomous variable, has been negatively associated with infant attention skills (Fidler, Schworer, Will, et al., 2019), but not related to emerging cognitive control on an infant strategy production task (Schworer et al., 2020). The current findings did not identify a link between prematurity and childhood action planning performance. It is notable that the results of this study may have overrepresented premature births in children with DS, limiting the opportunity to compare to children with DS born at term. This sample included a higher proportion of children born prematurely than the previously reported sizable minority in children with DS (Aoki et al., 2018), however, precise estimates are difficult to find (Fidler, Schworer, Will, et al., 2019). Additional work is needed to examine the impacts of prematurity on development in children with DS, particularly with nuanced methods that can capture idiographic differences across domains of development.

Similarly, a null result was found for children with and without CHD on the childhood outcomes for strategy production and efficiency. This non-significant result is in contrast to some previous research suggesting CHDs are related to greater language and cognitive development challenges during infancy and toddlerhood in children with DS (Alsaied et al., 2016; Aoki et al., 2018). However, the study findings align with previous research that did not identify greater challenges to performance on measures of infant cognitive control for infants with CHD relative to peers without a CHD (Schworer et al., 2020). This sample may overrepresent the prevalence of CHDs for children with DS, potentially as a result of the broad recruitment and opportunity to participate in a novel micro-intervention. The results should be considered in light of this limitation where almost three quarters of the sample had a CHD. Next steps for understanding the mixed findings for CHD in children with DS involve a need to better understand the developmental implications of CHD, including the characterization of CHD type, severity, and treatment complexity on developmental outcomes.

Post-Hoc Analyses

Handedness. Children's preferred reaching hand for the first trial of the Wave 2 Object Retrieval task was not related to better performance of strategy production or reaching efficiency. This finding is indicative of no differences in planning skills between children with different hand dominance, as may be expected. The Wave 2 object retrieval task could have side preferences depending on the side of the box that is open, however planning performance was not impacted by the hand dominance of each of the participants.

Notably, this sample was evenly split among children using their right and left hands, which is notably different from the laterality preferences observed in individuals without disabilities (Scharoun & Bryden, 2014). One potential explanation for the equal rates of hand preference in this sample is that preferred laterality may not be established yet for young children with DS. Hand laterality preference in children with DS has not received significant attention, and therefore its relationship to other aspects of development is unknown (Needham et al., 2021).

This preliminary finding for children's hand dominance should be interpreted in context to the study's operationalization, which could be less reliable than using parent reports or other *in vivo* assessments. Parent-reported handedness or other assessments could account for child preference across activities and environments. Future work should examine the role of handedness in developing action planning for children with DS.

Developmental Status. Additional post-hoc analyses explored possible correlates that could aid interpretations of associations between exploration efficiency and strategy production across infancy and early childhood. Examination of the developmental continuity of children's strategy between Wave 1 and Wave 2 was found to be nonsignificant. A second analysis examined associations with overall development Wave 1 and Wave 2. There were few significant associations between the infant exploration measures and early childhood action planning with standardized cognition scores or chronological age. First, chronological age was moderately negatively correlated with Bayley 3 cognition scores. This association aligns with previous findings for the growing disparity between chronological age and developmental status for children with DS, where skill acquisition is slowed relative to peers without disabilities (Fidler, Needham, et al., 2019; Hodapp et al., 1999). The second negative moderate association was between Bayley 3 cognitive scores and Wave 1 Exploration Efficiency. The direction of the correlation indicated that children who took longer to contact the toys at Wave 1 had lower Bayley 3 cognitive scores. This finding is matched with expectations that children with higher reaching efficiency have developed more competent cognitive skills. Significant correlations were only observed during infancy and not replicated at the childhood data wave.

Importantly, each of the outcome variables explored in this investigation showed large ranges of score variability. Heterogeneity amongst individuals with DS has been well-documented. Recent investigations have highlighted different subgroups within areas that have been well-observed at the group level (Fidler et al., 2019, 2022; Onnivello et al., 2022; Van Deusen et al., 2022). These findings open the door to further investigation of the trajectories of individuals with DS. Longitudinal study designs may facilitate answering these important questions regarding development in individuals with DS. The null associations are complex to interpret, and these associations might serve as evidence for the dissociability of EF, IQ, and age. An alternate interpretation of the null results may be that these skills do not have predictable continuity between each other. This gap in continuity may be a result of the operationalization of the constructs tested with the measurement tools used, or it may demonstrate that foundational skills in planning are not yet moldable in predictive ways from infancy to early childhood.

Task Feasibility

The tasks used in this study to assess object exploration and action planning were developmentally appropriate for children with DS, particularly because of the reduced task confounds that could impact interpretations of performance. The motor planning exploration task at Wave 1 included the use of toys that reduced requisite fine motor skills for participation. The task also required no expressive language and minimal receptive language demands. The toys had variable textures, rigidity, and appearance to increase child engagement and provide opportunities for exploration. Additionally, the age range of 9-17 months provided a higher likelihood that infants with DS could contact and use exploratory behaviors with the objects. All infants in the sample used at least one exploratory behavior.

The object retrieval task at Wave 2 was an action-planning task that required no expressive language for participation and reduced receptive language demands. This task was appropriate for this age range as it had 90.1% feasibility in this sample of young children with DS. Analyses with this task demonstrated an expected pattern between latency to retrieve and the reach quality a participant used to retrieve the object. Children who used a higher quality reach strategy were also more likely to retrieve the toys faster ($r = -.53$, $p < .05$).

There was a high level of completed retrievals within this sample of children with DS, but there was also a large amount of variety in reach strategy, time to retrieve, and response strategies used by the participants. The task appears to be a reliable way to measure action planning in children with DS, and this investigation extends the age range that it is developmentally appropriate for beyond the investigation from Fidler et al. (2005). This investigation is novel in its longitudinal approach and focus on EF at these young ages. Further investigations should seek to validate the outcome metrics for infant and early childhood planning measures.

Implications

This study examined the potential developmental continuity in planning skills from infancy to early childhood. This sample size is notable for the field and, for that reason, has the potential to inform the scientific understanding of the early trajectory for developing planning skills in DS. Examining exploration behavior as a foundation for developing skills in childhood action planning yielded overall null preliminary findings within an NHST framework. The possible preliminary link based on the magnitude of the effect between infant and childhood strategy production may suggest continuity in early development for these foundational planning skills. In contrast, the null result of the efficiency analysis suggested either discontinuity in developing planning skills or heterogeneity within the sample for subgroups of participants following different developmental trajectories. Overall, the regression findings could not conclude that there was evidence of developmental continuity in planning skills.

Task performance was also examined with exploratory descriptions of participant performance. The breadth of responses that were used by participants across tasks suggests that descriptive analytic approaches provide an opportunity to understand early development in children with DS. Utilizing exploratory descriptive analyses may launch new lines of inquiry and support researchers to design studies that are better positioned to identify effects of development in this population. Moving forward with complementary approaches in confirmatory analyses and exploratory small-sample approaches may best position the field to examine continuity in skills, such as planning, early in development.

Planning as an executive function

This investigation was focused on the EF subconstruct of planning. Planning is a top-down regulatory skill that draws upon several EF subconstructs, including working memory, inhibition, and shifting. Planning frequently requires integrating these skills to achieve goals and complete tasks. However, the action-planning task used in this work aimed to reduce the confounds associated with other requisite skills of EF and areas of development (e.g., language). This task required participants to update their approach to retrieving the toy as the opening on the three-sided box moved around and aimed to capture planning skills independent of the other constructs of EF.

Subsequent examinations of developing planning skills may benefit from characterizing EFs in context to one another. Planning in daily life is a more integrated EF, drawing upon the ability to remember, inhibit, and flexibly adjust to environments to achieve goals. Throughout childhood, adolescence, and adulthood, there is increasing complexity to complete activities of daily living. Examining the underpinnings of behavior from the earliest stages of the lifespan can be instructive for identifying areas needing support earlier in the lifespan.

Future Directions

Future investigations should consider non-linear analytic approaches to developmental growth in DS. Children in this study demonstrated a wide range performance across each metric of efficiency, production strategy, and response strategy at Wave 1 and Wave 2. Future work should explore novel analytic approaches and other methodologies to better understand this within-syndrome heterogeneity in development for individuals with DS (Prince & Fidler, 2021).

An additional direction for future work would be to more comprehensively characterize developing EF skills of young children with DS. Considering the dissociability of EF, there may be a core EF (e.g., working memory, inhibition, or shifting; Diamond, 2013) that is limiting the development of planning skills. Planning integrates aspects of each of the core EFs, so having a limited skillset in one core subconstruct may hinder the individual from employing planning skills in novel environments, including laboratory-based measures.

A more comprehensive characterization of skill development could also include physiological data collection tools, such as eye tracking and heart rate variability during the completion of tasks. These tools have been used primarily in research with children with and without disabilities to examine underlying differences in attention and processing speed during infancy (Kulke et al., 2017; Roberts et al., 2012). These measures have been used less frequently in children with DS, in part due to challenges related to calibrating eye trackers with co-occurring vision conditions (Fidler et al., 2023; Schworer et al., 2022). Additional exploration of biobehavioral approaches may further elucidate our understanding of developing cognitive skills in children with DS.

Study Limitations

This investigation provided a novel examination of early development in children with DS, and the study is not without limitations. Data collection for this study was initiated a few weeks before the onset of the COVID-19 pandemic. The pandemic may have impacted performance not captured through the questionnaires and performances at Wave 2. As a result of pandemic protocols, children and families were recruited over a more extended period than initially planned to prevent the spread of the COVID-19 virus. The pandemic resulted in a broader age range of participants in the sample and more variability in the time between Wave 1 and Wave 2.

The study was limited by having only two-time points to observe developmental associations. A third data wave would have provided the opportunity to look at developmental trajectories and growth across participants. Capturing a third data wave would have also increased within-participant power, which could have supported nuanced statistical techniques to examine growth. An idiographic confirmatory approach may provide more opportunity to understand if exploration provides a foundation for developing planning skills. This investigation was not able to answer that question conclusively.

The sample size was too modest to analyze more than preliminary associations between waves of data collection. Sample size limited the power to detect effects, include other potentially relevant covariates, or explore non-linear methodologies that require more statistical power. The novelty of two-time points in early childhood in children with DS is a critical starting point, and the sample size is strong relative to other investigations with this population. Descriptive analyses provide a foundation to build lines of inquiry to further the understanding of developing planning skills for this population.

The results may have been impacted by the age ranges at each time point. The Wave 1 window included 9-17-month-olds, which is a period of significant development in infancy. Although 9-17 months is a relatively specific developmental window, some children with DS may just be starting to reach and grasp for objects more, particularly with infants showing higher levels of delay. On the other hand, some infants may have been more advanced than the coded behaviors observed in this exploration task. Consider the swat behaviors in Figure 1, which show a positive skew. Some infants may have been developmentally past using this as an object exploration technique, relying more on efficiently made grasps and more sustained play. Variations in skillsets could be hypothesized from recent work characterizing heterogeneity (Fidler et al., 2019, 2022; Onnivello et al., 2023; Van Deusen et al., 2022). However, this investigation was not powered to parse age and skill differences.

At the Wave 2 visit, similar difficulties may have emerged from the wide age range of the participants seen. The Wave 2 age range has a larger developmental window than was initially proposed (3-7 years versus 4-5 years). Although developmental status analyses did not indicate that chronological age was significantly related to the outcomes of interest, the wide range may have occluded patterns from a more targeted window (i.e., preschool). Additionally, developmental testing requires the child participant to sit and attend tasks similar to attending school. With the sample for this study, a subset of participants at Wave 2 attended school in different capacities, and school attendance was not accounted for in these results. Although school was not accounted for in these results, there were no associations that emerged as a confound to developmental status.

Each of the tasks used in this study were selected to be feasible for each developmental window of children with DS. However, the tasks may have lacked construct validity or convergent validity because the investigation was limited to one measurement of planning at each Wave. A clearer picture of planning may have emerged if more than one assessment of planning was administered at each time point. This would have provided greater evidence for examining planning as a distinct construct from the other EF subconstructs (Willoughby & Hudson, 2021). Additionally, more direct assessments could have provided the opportunity to look at multimodal use of exploration behaviors in infancy. This investigation was restricted to physical exploration of the objects due to the limited number of children using oral exploration. More comprehensive assessment of relative exploration strategies, including oral and visual exploration, may have shed clearer light on infant exploratory planning, which could have provided a clearer picture of continuity to early childhood action planning.

Conclusion

This investigation is the first examination of developmental trajectories for individuals with DS from infancy to early childhood. The null findings create some complexity for interpretation; however, they also illuminate important opportunities to understand the trajectories of development in children with DS. The pattern of results presented in this investigation opens the door to reducing the focus on continuity in development from infancy to early childhood. Instead, findings suggest malleability in trajectories and the potential to impact skill development throughout early childhood. An important future direction will be to examine different trajectories that result in similar skill competence in planning during early childhood. A more nuanced approach of this nature can inform opportunities for intervention that strengthens planning skills in children with DS from the earliest stages of the lifespan.

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APPENDICES

Appendix A: Motor Planning Task Coding Scheme

<u>Behavior Groups:</u>

a. **Motor Cognition**

    i. **Latency to initial contact (duration behavior):**

        1. This code was started when each ball was presented and stopped when the infant made contact with the toy.

            a. Modifiers:

                i. Specified the ball used in the trial (e.g., yellow nubby, orange crater, squidgy, koosh, or other)

    ii. **Latency to initial grasp (duration behavior):**

        1. This code started when the infant made initial contact and stopped when the infant made a complete grasp.

            a. Modifiers: Specified the ball used in the trial (e.g., yellow nubby, orange crater, squidgy, koosh, or other)

    iii. **Squeezing (frequency behavior):**

        1. Coded this behavior when the infant squeezed the ball.

        2. If the infant took 3 seconds or more as a break from squeezing and squeezed the ball again, a new squeeze code was scored.

            a. This code was used less frequently with the koosh ball because of its object properties.

iv. **Mouthing (frequency behavior):**

1. Coded this behavior when infant explored the ball by mouthing.
2. If the infant took 3 seconds or more as a break from mouthing and mouthed the ball again, a new mouthing code was scored.

v. **Contact (frequency behavior)**:

1. Coded when any type of contact was made with the ball.
2. Coded with initial latency to contact, immediately following the stop of the duration code.
    a. Modifiers:
        i. Specified which type of contact (e.g., a swat, or a touch that is NOT a full/clean grasp)
        ii. Ball type

vi. **Grasp (frequency behavior):**

1. Coded when the infant's hand was fully enclosed around one of the objects or the infant made full contact with their hand.
2. Could have been scored as one hand enclosing around an object OR both hands coming together to enclose around a larger object (e.g., too large to grasp with one hand)
    a. Modifiers:
        i. Ball Type
        ii. Efficiency of grasp: Coders considered, "Does the infant grasp in a way that is unique to the physical

properties of the ball?" These are all examples of efficient grasps:

1. Orange "crater" ball: Grasped with fingers in craters.
2. Nubby yellow ball: Grasped by nubs.
3. Squidgy: Grasped by one of the folds of the ball.
4. Koosh: Grasped with pincer (thumb and pointer) or refined grasp
5. If "other" ball:
    a. Large ball: Grasped with both hands if it was too large to grasp with one hand.

iii. Other types of grasps were considered inefficient (e.g., trying to grab a large ball with one hand when both hands were necessary)

**Any movement/guiding of child's hand from caregiver was not coded, unless the infant made additional actions on toy after the caregiver's hand left.

Appendix B: Object Retrieval Task Coding Scheme

Start: The observation for coding was started when the toy was placed on the table for first trial by the examiner.

<u>Coded Behaviors:</u>

A. Examiner Codes:

   a. **Redirections (frequency behavior):**

      i. Modifiers: Redirection type.

         1. Physical – The examiner adjusted the materials in response to off task behaviors and repositioned them to support the participants engagement.
         2. Verbal – Examiner repetitions of the instructions are considered redirections. Score this code when the participant is guided back to focus on the game and respond to the trial (e.g., "First, then", "Now we are playing this game").

B. Child Codes:

   a. **On-task Attention (duration behavior).**

   b. **Off-task Attention (duration behavior):** Children's off-task attention interrupted the flow of trials and diverted the examiner away from the task and task materials to respond to the child's off-task cues.

      i. Off Task Modifiers

         1. Refusal – For example, child's physical movement of materials away, "No", "I don't want to", complete a couple trials "All done".
         2. Crying

3. Elopement
4. Social – Child started conversation to stop/distract the examiner from completing the task.
5. Fatigue – Child yawning, turning themselves away from the materials.
6. Other Off Task Behavior: Commented to understand additional avoidance behaviors.

C. Trial Response

a. **Latency to respond (each trial – duration behavior):** Started when the examiner finished set-up for the trial by placing the clear box on top of the animal and/or prompting the child to "Grab it." (Coders monitored to start the latency to respond time when the participant initiated movement toward the box to respond.) The code stopped when the toy was removed from the box.

Coder notes:

-Contact with the clear box is not latency to respond. The code was kept on until the toy was removed from the box.

-If the examiner retrieved the toy to remove it from the box – Do not code this behavior.

i. Modifier Group: Accuracy
   1. Success
   2. Failure to retrieve object from under the box

ii. Modifier Group: Hand(s) Used
   1. One Hand

2. Both Hands (simultaneously)
3. Switched Hands During Trial

iii. Modifier Group: Opening of box

1. Child's Right
2. Child's Left
3. Away from child (Facing directly in front of examiner)
4. Facing Child (Facing away from examiner)

b. **Response strategy (each trial – frequency trial):** Coded immediately following the end of the previous latency to respond code. *Select all that apply*

i. Immediate retrieval
ii. Contact with wall of box
iii. Attempt to manipulate box: Participant tries to lift/move/shove/push the box
iv. Changing hands during search
v. Moving toy around in box, but not removing toy from box
vi. Using second hand to stabilize box
vii. Other (If you select other – please describe the other behavior in the spreadsheet tracking your coding.)

c. **Reach Score (each trial – frequency behavior):** Code for reach score and then assign reach value 1 - 4.

i. 1 = Child reached through top of the box first.
ii. 1.5 = Child reached through one or multiple sides of the box in effort to obtain the toy.

iii. 2 = Child leaned over to look through opening and reached for the toy while looking through opening.

iv. 3 = Child leaned over to look through opening, straightened their posture, and reached through while looking through top of the box.

v. 4 = Child reached through opening while looking at the box.

➔ Observation is stopped when the examiner removed the clear box and/or animals from the play surface. The observation was also complete if the examiner and participant played with the animals and the box was set aside, or if the child begins to manipulate the box.