American Journal on Intellectual and Developmental Disabilities Evaluation of a modified Corsi task to assess visuospatial short-term memory in young children with Down syndrome

--Manuscript Draft--

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Abstract

Short-term memory (STM) challenges are often observed in children with Down syndrome (DS), but existing early STM measures introduce measurement confounds in this population. To address the need for valid early STM measures for future DS interventions, this study evaluated the psychometric properties of a modified Corsi span task, administered to 110 children with DS. Results indicated that the modified Corsi task has feasibility in the age range of 5-8 years and is scalable across chronological and mental ages. Minimal practice effects and evidence of test-retest reliability and convergent validity were observed. Implications for using a modified Corsi task in studies of early STM and treatment trials for children with DS are discussed.

Evaluation of a modified Corsi task to assess visuospatial short-term memory in young children with Down syndrome

Down syndrome (DS), caused by trisomy 21, is the most common neurogenetic condition associated with intellectual disability (Antonarakis et al., 2020). Although heterogeneity of outcomes is observed across many domains in this population, individuals with DS are generally predisposed to patterns of relative strength and challenge across a range of cognitive and behavioral dimensions (Fidler, 2005; Grieco et al., 2015). Short-term memory (STM), or the ability to temporarily store information, is a dimension that is especially impacted in many individuals with DS (Jarrold & Baddeley, 2001; Purser & Jarrold, 2005). Both verbal and nonverbal STM have been the focus of DS research at various stages of the lifespan (Frenkel & Bourdin, 2009; Jarrold & Baddeley, 1997), with some studies demonstrating that individuals with DS show greater STM challenges than would be expected when compared to mental age (MA) matched controls (Conners et al., 2011; Godfrey & Lee, 2018; Tungate & Conners, 2021). While verbal STM performances largely drive these effects, various aspects of nonverbal STM abilities be impacted as well (Bennett et al., 2013; Tungate & Conners, 2021).

Characterizing and supporting the development of visuospatial STM is important because of its role in everyday cognitive processes. The ability to maintain visual and/or spatial information in temporary storage is necessary for remembering and following visually represented information like maps and calendars, completing sequences of actions required for activities of daily living, and organizing/maintaining personal spaces. In the general population, preschooler visuospatial memory predicts current and future math ability, and in school-age children it is associated with arithmetic calculation, reading fluency, and reading comprehension performances (Berg, 2008;

Bull et al., 2008; Swanson & Howell, 2003). Supporting the development of visuospatial STM in individuals with DS could lead to improved quality of life by enhancing these essential skills.

In contrast with working memory, another construct that has received research attention in DS, STM involves the basic temporary storage of information without any manipulation or resistance of interference (Godfrey & Lee, 2018). Visuospatial STM may be a particularly promising intervention target, as this dimension demonstrates modifiability in the general population (Jaeggi et al., 2011). Gade et al. (2017) investigated the modifiability of visuospatial STM in preschool-aged children in the general population and reported significant improvements after 9 or 12 training sessions on a visuospatial STM task. Notably, participants with lower pretest performance scores benefited most from the intervention, supporting a compensatory gain hypothesis. This has relevance for young children with DS, who often demonstrate emerging cognitive challenges from an early age (Fidler, 2005). Near-transfer effects have also been demonstrated in STM training studies with neurotypical adults, wherein visuospatial STM training yields improvements in untrained tasks within the same domain (Harrison et al., 2013).

These promising findings offer the possibility that treatment approaches may yield positive outcomes for visuospatial STM in children with DS. However, future treatment work will require performance-based measures that are sensitive to changes in STM processing efficiency and that can capture intervention effects in childhood – a period during which treatment may have the greatest downstream effects. At present, many STM measures are not suitable or have not been evaluated for use with young children with DS. As a result, little is known regarding early emerging STM skills in this population. Currently available early STM measures often present interpretational confounds as they may involve other developmental skill areas, like language and motor skills, that are known areas of challenge for those with DS (Daunhauer & Fidler, 2011; Martin et al., 2009; Tungate & Conners, 2021). Adapting and evaluating existing visuospatial STM tasks, like the Corsi Span block task, to minimize or remove those confounds can advance DS clinical science by both facilitating early STM developmental characterizations and providing psychometrically sound outcome measures for treatment studies for young children with DS.

Assessing Short-Term Memory in Children with Down Syndrome

The Corsi Span block task is a measure of visuospatial STM that has been frequently used in research on older children and adolescents with DS (see Yang et al., 2014 for a review; Conners et al., 2011; Godfrey & Lee, 2018). This task involves the sequential presentation and recall of visuospatial information. The original version of the task involved an examiner tapping a sequence of identical blocks, with the participant then asked to reproduce the sequence (Corsi, 1972). STM capacity was evaluated by gradually increasing the length of the tapping sequence until the participant could no longer accurately replicate it. Adaptations of the Corsi Span involve participants replicating sequences in different ways, for example, using their fingers or controlling a character in computerized versions of the task (e.g., Brock & Jarrold, 2005; Rasmussen & Bisanz, 2005). Existing STM research in DS using the Corsi Span and Corsi-type tasks suggests that individuals with DS demonstrate delays in alignment with their overall developmental status (Yang et al., 2014). However, previous work has primarily focused on older children and adolescents (on average, over 15 years old) and without DS-informed modifications (Yang et al., 2014). Therefore, little is known

regarding the early foundations that lead to later Corsi performances in older individuals with DS.

When evaluating STM in young children with DS, it is essential to consider specific phenotypic dimensions within an adapted Corsi-type task. Doing so may mitigate potential confounds to ensure a more accurate assessment of the cognitive construct of interest. Children with DS often experience motor planning challenges (Daunhauer & Fidler, 2011; Fidler, 2005), which must be considered when designing early measures of memory and cognition. Variations in Corsi block-tapping task materials, such as block size and color, may significantly impact performance (Kessels et al., 2000). For example, using small blocks that are uniform in color may pose challenges for participants with perceptual or motor difficulties. An adapted Corsi Span measure for younger children with DS should eliminate complex motor planning (such as reaching and grasping smaller objects) and use high-contrast colors for different tapping locations to reduce confounding factors. In addition, DS is associated with receptive and expressive language challenges (Fidler, 2005; Martin et al., 2009). Modifications to the Corsi task should minimize receptive and expressive language demands, for example, by reducing the complexity of verbally presented instructions and minimizing the need for a verbal response. Finally, when targeting younger age groups, selecting and designing tasks that are inherently motivating is essential. Design elements that enhance engagement and resemble play are helpful for maintaining attention and accurately capturing STM performances.

The present study aimed to evaluate the psychometric properties of a modified Corsi-type task in assessing visuospatial STM in young children with DS, aged 2.5 to 8 years. The design of the *Childhood Modified Corsi Span (CMCS)* task was informed by phenotypic features often observed in children with DS to minimize potential confounding effects from perceptual and motor challenges often observed in this population, including the use of colorful switches with 5.25-inch large activation surfaces for participants to press instead of tapping small blocks (Figure 1) and a reduced number of tapping targets. Additionally, the *CMCS* task was presented in a game-like manner, with reduced receptive language demands, and when appropriate, teaching strategies were used during teaching trials, such as hand-over-hand assistance. The adaptations made to the original Corsi task for the *CMCS* are summarized in Table 1. The present study evaluated task feasibility, floor effects, test-retest reliability, practice effects, developmental sensitivity, and convergent validity of the *CMCS*, thereby contributing insights for future research aimed at understanding or enhancing visuospatial skills among individuals in this population, and characterizing within-DS heterogeneity in performances along this dimension.

Methods

Participants

Participants were 110 children aged 2.50 to 8.75 years $(M = 5.26$ years; $SD = 1.52$) with a confirmed DS diagnosis and their caregiver. Child participants were required to meet the following inclusion criteria according to parent/caregiver report: previous genetic diagnosis of DS, no more than a mild documented hearing loss, and no uncorrected visual impairment. Participants with other co-occurring conditions, such as autism, were included to represent the overall population of individuals with DS. Inclusion criteria involved the ability to sit unsupported for at least one minute and reach for toys, and at least six weeks since any surgical procedures. Both child participants and their caregivers were required to understand instructions in English.

One family that expressed interest in the study was ineligible due to English language requirements. See Table 2 for participant demographics.

Procedures

Data for this study were collected from two ongoing studies of cognition in DS under IRB approval at {*withheld for review*}. Recruitment took place through DS community organizations and clinics. Child participants in each study completed a battery of assessments measuring developmental status and cognitive functioning and caregivers completed questionnaires. Direct assessment measures were counterbalanced across participants *a priori* through an enrollment log. Four randomizations were used in the larger project, and two randomizations were used in the smaller study. A subset of participants $(n = 29)$ returned for a second visit two weeks later to examine test-retest reliability and practice effects.

In the larger study on cognition, participants were evaluated four times over the course of a year in laboratory spaces available to the research team. Research visits were scheduled in either two- or four-hour increments based on family preferences and availability. Each research visit in this larger project compensated families \$50. In the smaller study, participants were evaluated during one two-and-a-half-hour visit in a location convenient to the family, including laboratory spaces, families' homes, and community spaces (i.e., library study rooms). An incentive of \$40 was provided to all participating families upon consent to this second project.

Measures

Childhood Modified Corsi Span Task (CMCS). Visuospatial STM performance was evaluated via the *CMCS*, a modified Corsi-type task. The examiner gained the participant's attention and modeled tapping on an array of four large colorful button

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switches programmed to each produce a unique tone when pushed (Figure 1; Enabling Devices, 2019). The participant was given the opportunity to briefly engage with the buttons to capture interest and increase motivation. The four switches were presented on a white tray in the order blue, red, green, and purple from the participant's left to right. After demonstrating tapping for the trial at one tap per second, the examiner pushed the tray toward the child, without a delay, to allow an immediate imitation of the sequence presented. To ensure the participants understood the nature of the task, two teaching trials were administered before the scored trials. After each teaching trial, the examiner provided feedback, confirming whether the response was correct or teaching the correct response. Feedback was provided up to two times for each teaching item, and participants were given up to three attempts to reproduce the target sequence on each teaching trial. Feedback was provided verbally, but children could also receive handover-hand teaching to demonstrate the goal of the task. The scored trials were administered after the teaching trials regardless of the participant's performance during the practice phase.

The *CMCS* task design included starting point rules tailored to different CA groups. Starting items for children ages 2.5-4.9 years involved reproducing sequences initially with a single tap and increasing to up to eight taps. For children ages 5 years and older, administration began with two-tap sequence items. If children in the older CA group did not respond correctly to either teaching trial at the two-tap starting point, they were administered the single-tap teaching trials, including accompanying feedback. The task was discontinued when a child produced three consecutive incorrect responses or did not respond on three consecutive scored trials. In situations where children older than 5 years passed at least one teaching trial, but the first three scored trials incorrect,

the examiner dropped back to administer the single tap items. If a participant did not pass teaching trials and did not provide any correct responses before reaching the stop rule, they were assigned a score of 0. If a participant displayed ongoing interfering behaviors during teaching trial administration, the task was discontinued and the data were considered missing.

Task administration time was approximately five minutes. Single-tap trials were scored 1 point for correct reproduction or 0 points for incorrect reproduction, and ≥ 2 tap trials were scored 2 points for correct reproduction, 1 point for the correct locations in an incorrect sequence, and 0 points for incorrect location tapping or sequence number. Children in the older age group who successfully produced a response at their later start point were given credit for the four scored trials from the earlier start point. Trial scores were summed for a total task score. Each span length (1-span up to 8-span) included four scored trials, such that the highest possible total score was 60 points (4 single-location trials with the highest possible score of 1 and 28 items with the highest possible score of 2). *Task Behavior.* Examiners assessed the participant's attention and task opposition (dichotomously, yes/no) immediately after administering the *CMCS*. Participant attention was defined as attending to the examiner during instructions and displaying interest in the task by actively looking at the materials or examiner. Opposition was defined as demonstrating resistance to the task by refusing to follow task instructions, shaking of the head, looking away, pushing away task materials, and/or attempting to move away from the task.

Medical History Questionnaire. Caregivers provided information about their child's sex, race, ethnicity, DS diagnosis type (Nondisjunction/Trisomy 21, Mosaic, Translocation, or Not Sure) and other co-occurring biomedical conditions.

Cognitive Status. Two standardized assessments were used to determine the cognitive status of participants. The Cognitive domain of the Bayley Scales of Infant and Toddler Development-4 (Bayley-4; Bayley & Aylward, 2019) was administered to children between the ages of 2.5 and 3 years, and the Stanford Binet-5 Abbreviated Battery IQ (SB-5 ABIQ; Roid, 2003b) was administered to children over 3 years. The Bayley-4 is a standardized assessment that measures cognition, communication, and motor skills in children 16 days to 3.5 years old. The Bayley-4 scales have high internal reliability (.93 to .95) and test-retest reliability (.81 to .84) in clinical and non-clinical populations (Bayley & Aylward, 2019). The SB-5 is a normed and standardized assessment frequently used in DS research. It has high internal reliability (.91) and testretest reliability (0.85; Roid, 2003a).

Participant MA was estimated via age equivalent (AE) scores from the Bayley-4 and SB-5 ABIQ. There were concerns with using the SB-5 ABIQ only due to potential floor effects within this sample, as the lowest AE score on the SB-5 is <2 years. To address this concern, if time allowed, children with a CA of 3 to 4.99 years were also administered the Cognitive domain of the Bayley-4 ($n = 14$). For all participants where a Bayley-4 Cognitive AE score was available, their Bayley-4 AE scores were used for analyses in place of the SB5-ABIQ. We note that although the Bayley-4 was designed for use in children up to 3.5 years of age in the general population, the measure was within the appropriate developmental range for children with DS ages 2.5-4.99, and AE scores were within range in this sample. Eight participants (7.3%) did not have an AE score because cognitive testing was not administered due to behavioral refusal (5), fatigue (1), administration error (1), or time constraints (1).

In addition to the use of AE scores, change-sensitive scores (CSS) from the SB-5 ABIQ were calculated. CSSs use item response theory to convert raw scores into criterion-referenced scores that measure absolute levels of ability. These person ability scores are designed to assess change within an individual, and they tend to be sensitive to small intraindividual changes while also preserving variability at the extreme lower ends of scoring (Farmer et al., 2020). Person ability scores, like CSS, may be useful in research with individuals with neurodevelopmental disorders because they measure change in ability rather than relative standing based on population norms (Farmer et al., 2020). Person ability scores cannot be compared across different assessment measures like the Bayley-4 and the SB-5, thus CSS from the SB-5 were used for those participants who had scores available $(n = 96)$, while MA was retained in analyses to compare across the two cognitive measures utilized for this study.

Convergent Validity: Working Memory. To evaluate convergent validity, performances on the *CMCS* task were compared with those on the *Garage Game,* a working memory task previously evaluated for use with young children with DS (see Pinks et al., 2023 for administration details). The *Garage Game* was adapted from the original *Three Boxes* task, a self-ordered pointing task used to measure working memory (Devine et al., 2019; Diamond et al., 1997; Petrides, 1995). The child is presented with a toy car garage with color-matched cars and garage doors and asked to locate the toy cars in up to three sets of trials. A repetitive search rate was calculated by dividing the total number of repeated search locations by the total number of cars the child searched for, with higher rates indicating greater working memory challenges.

Data Analysis

All analyses were conducted within the R statistical computing environment (R Core Team, 2022). Participants who were able to comprehend the task requirements enough to successfully engage with a task trial demonstrated feasibility. Feasibility of the *CMCS* was calculated as the percentage of participants who replicated at least one trial, including teaching trials. Some participants met feasibility criteria by correctly responding during a teaching trial but then did not produce any correct responses during scored trials, thus scoring at the floor of the task. A benchmark of 80% was established *a priori* for feasibility, in alignment with the threshold in prior work evaluating cognitive measures for use in children with DS and other conditions associated with intellectual disability (e.g., Hessl et al., 2016; Schworer et al., 2022).

Group-level performances were evaluated for distributional issues. Floor effects were assessed by evaluating the number of participants for whom the task was feasible who obtained the task's lowest score; i.e., no correct responses on scored task trials. The threshold for acceptable floor effects was set at <20% in accordance with previous studies involving individuals with DS (Pinks et al., 2023; Schworer et al., 2022). Based on visual inspection of Corsi task performance distributions by CA, additional analyses were conducted to characterize the psychometric properties of the task in a subsample of participants who were 5 to 8 years old.

Test-retest reliability was assessed using an intraclass correlation coefficient (ICC) for the subsample of children who completed a second visit two weeks later. Criterion for reliability was set *a priori* as a value > 0.75 indicating good reliability, and a value between .50 and .75 indicating moderate reliability (Koo & Li, 2016). Paired samples *t-*tests were conducted to evaluate the presence of practice effects between

visits 1 and 2. Convergent validity was evaluated by comparing *CMCS* scores to those of the *Garage Game* via Bivariate Pearson correlations.

To evaluate developmental sensitivity, associations between developmental domains (CA, MA, and CSS) and task scores were tested using Quasi-Poisson regression models, which accounted for overdispersion in the outcome variable (Ver Hoef & Boveng, 2007; Wedderburn, 1974). Results of the models provided estimates of the average difference in *CMCS* scores for each 1-unit difference in age based on a crosssectional sample.

Predictor variables in regression models were continuous CA and MA values measured in years and continuous CSS values. For characterizing feasibility and distributional dimensions across MA and CA, categorical variables were created by rounding down to the nearest whole number to create one-year bins. For the binned MA variable, individuals with an MA of 24 months were categorized within the 1-year MA bin, whereas those with an MA ranging from 25 to 35 months were included in the 2 year MA bin.

Results

Feasibility. Of the 110 participants, 82 generated a correct response on a trial of the *CMCS* task, demonstrating a feasibility level of 74.5% for the entire age range represented in the sample. Out of the 28 who did not demonstrate feasibility, 20 (71.4%) children attempted the task and were not able to produce a correct response on a teaching item or test item. For the remaining 8 children, the task was initiated and then discontinued early due to behavioral/verbal refusal (5), non-understanding (2), or fatigue (1) according to examiner report. The CA of children who did not meet feasibility criteria ranged from 2 to 7 years. Nineteen children who did not demonstrate task

feasibility had an MA of 1 year and three children had an MA of 2 years. The remaining six children were missing an MA equivalent. The majority (95 of 110; 86.4%) of participants demonstrated attention to the task. Opposition was demonstrated by 25 (22.7%) participants. Of the 82 participants who met feasibility criteria, 14 (17.1%) demonstrated opposition. *Restricted CA subsample.* When the participant CA range was restricted to 5 to 8 years, task feasibility increased to 84.7% (50 out of 59 participants).

Performance Distributions. Of those participants who were administered scored task trials, the minimum score was 0, the maximum score was 28, and the mean score was 4.91 (*SD* = 6.80). The total score skew was 1.56, and the kurtosis was 1.79. Floor effects were defined as participants who passed a teaching trial but received the lowest score of 0. Out of the 82 participants for whom the task was feasible, 22 children (26.8%) performed at the floor of the task. Two additional children (2.4%) did not continue to the scored trials due to refusal (1) or non-understanding (1) according to examiner report. The CA of participants demonstrating floor effects ranged from 3 to 7 years. Of the 22 participants at the floor, 15 (68.2%) had an MA of 1 year, and six (27.3%) had an MA of 2 years. The remaining participant who displayed floor effects was missing an MA equivalent. *Restricted CA subsample.* When the CA range was restricted to 5 to 8 years, the total score skew was 0.87 and the kurtosis was -0.10. Only nine of 50 participants (18.0%) in this age range who met feasibility criteria performed at the floor of the task.

Test-Retest Reliability and Practice Effects. Test-retest reliability was investigated within a sub-sample of participants ($n = 29$; CA range = 2.53-8.71, mean CA = 5.20 years, *SD* = 1.82; mean MA = 2.16 years, *SD* = 0.92) who returned for a second visit two weeks after the first assessment. *T*-tests demonstrated that CA and MA were

not statistically different among the subset of participants who returned for a second visit compared to those who did not, $t(108) = 0.22$ and $t(100) = 1.49$, respectively, *p*values > .05. Test-retest reliability was moderate at two weeks, with an ICC of 0.62. No meaningful practice effects were observed from visit 1 (mean = 4.38) to visit 2 (mean = 3.79 , $t(28) = 0.54$, $p = .60$. The effect size for the practice effects, measured by Cohen's d, was $d = 0.10$.

Convergent Validity. Of the 110 participants in this study, 87 had scores available on the *CMCS* task and *Garage Game* working memory task. This includes participants who did not meet feasibility criteria on the *CMCS* and/or scored at the task's floor. Total scores on the *Childhood Modified Corsi* task were correlated with *Garage Game* repetitive search rates $(r(85) = -0.35, p = .001)$, demonstrating adequate convergent validity. A negative correlation indicates convergence, as higher *Garage Game* repetitive search rates represent greater challenges in working memory.

Developmental Sensitivity. CMCS scores were related to CA and MA in separate models, $exp(β) = 1.83$, $SE = 0.07$, $p < .001$ (95% CI: 1.59, 2.12) and $exp(β) = 1.94$, $SE =$ 0.09, $p < .001$ (95% CI: 1.63, 2.30), respectively. Based on these models, it was estimated that a 1-year difference in CA was associated with an 83% higher task score, and a 1-year difference in MA was associated with a 94% higher task score. See Figures 2 and 3 for visualizations of the relationship between Corsi scores and CA and MA. When including CA and MA in the same model, task scores remained significantly associated with both predictors, $exp(\beta) = 1.62$, $SE = 0.07$, $p < .001$ (95% CI: 1.41, 1.87) and $exp(\beta) =$ 1.51, SE = 0.08, *p* < .001 (95% CI: 1.29, 1.77), respectively. Holding MA constant, a 1 year difference in CA was associated with a 62% higher score on the Corsi task. Holding CA constant, a 1-year difference in MA was associated with a 51% higher score on the

Corsi task. From the present sample, the average Corsi scores found in Tables 3a and 3b were predicted based on CA and MA in children with DS.

To address limitations that are inherent in norm-referenced AE scores, the association between *CMCS* scores and SB-5 CSS values was modeled for the participants who completed the SB-5 ABIQ. *CMCS* scores were related to SB-5 CSS, $\exp(\beta) = 1.05$, SE = 0.01, *p* < .001. A model with CA and SB-5 CSS values confirmed that both CA and cognitive status remained significant predictors of *CMCS* performance when holding the other constant, $exp(β) = 1.63$, $SE = 0.08$, $p < .001$ (95% CI: 1.40, 1.92) and $exp(β) =$ 1.03, SE = 0.01, *p* < .001 (95% CI: 1.02, 1.04), respectively.

Discussion

The present study examined the psychometric properties of an adapted visuospatial STM measure administered to young children with DS. A modified version of the Corsi Span task was administered to children with DS between the ages of 2.5 and 8 years. Results demonstrated feasibility of administration in the 5-to-8-year CA range, and positive associations were observed between task performance and CA, MA, and CSS. Floor effects were observed for children with CAs younger than 5 and MAs younger than 2. Evidence of test-retest reliability was also demonstrated, with minimal practice effects, as well as developmental sensitivity. Based on these findings, the *CMCS* task appears appropriate for use in DS developmental and treatment research, particularly for children ages 5 to 8 years. It may also be a useful measure for interventions with CAs younger than 5 years who perform at the floor of the task, as a means to quantify treatment effects when STM gains are made.

Psychometric Properties and CA. Although overall psychometric properties were acceptable, there are important considerations to note within the observed performance

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patterns. In terms of feasibility, although the majority of children in the 2.5-8-year age range were able to correctly replicate a sequence, approximately 26% of children were not. Therefore, the *a priori* feasibility criterion set at 80% was not met in the full sample of participants. However, the CA range for this study was intentionally wide to generate as much descriptive information as possible about the measure's utility. When considering a narrower CA range of 5-to-8 years, the task exceeded the *a priori* criterion for feasibility and met criteria regarding floor effects. Additionally, in analyzing the total score outcome of the *CMCS* task, there were indications of skewness and a slight concern for kurtosis, but within the narrower age range of 5-to-8 years, these distributional problems were reduced.

Despite these distributional issues, the *CMCS* may still have utility for children with DS under the age of 5 years who perform at the floor of the task, when considered in the context of STM treatment studies. The *CMCS* may be useful for demonstrating baseline performances, even if they are at the floor of the measure, to capture subsequent gains at intervention exit. This potential use is bolstered by the noted developmental sensitivity of the task. Thus, even with feasibility concerns in children younger than 5 years, the *CMCS* may have measurement utility in the evaluation of intervention or treatment effects in young children with DS.

It is also noteworthy that approximately 7% of participants did not complete the task due to interfering behavior or examiner decision to end based on child comprehension of task instructions. Therefore, while the vast majority of participants demonstrated some meaningful performance on the *CMCS* task, a subgroup of participants was unable to do so, even at the older end of the CA range of this study. This aligns with a growing understanding of heterogeneity in cognitive presentation

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within the DS population (Määttä et al., 2006; Onnivello et al., 2022), and has two implications. First, understanding and describing heterogeneity is important for developing a more personalized approach to treatments and interventions. The CA and MA models generated in this study can begin to inform an understanding of normative DS performances by age, which can help identify when a child with DS may need more intensive interventions or tailored accommodations than other children with DS. A second implication of this study is the need to identify novel approaches to further expanding the lower bounds of STM span tasks to include an even broader range of performances within this population (Esbensen et al., 2017). Future work should identify additional ways to capture early performances on the dimension of STM in this population to better characterize this subgroup of individuals with DS.

Predicting Performances in Children with DS. As described above, the *CMCS* task demonstrated developmental sensitivity in the present sample, as observed in the Quasi-Poisson models. Although a substantially larger sample of participants would be necessary for establishing DS-related performance norms on this task across CAs, the Quasi-Poisson analyses and visualizations make it possible to preliminarily predict performance based on the cross-sectional distributions observed in the present sample. This type of modeling of adapted evaluation measures, like the *CMCS*, is a useful initial approach to studying individual and group-level performances in a population such as children with DS. Initial predictions of anticipated scores at the group level make it possible to contextualize individual performances on this task for a child with DS relative to other children with DS, which can be helpful information for caregivers, clinicians, and educators.

Convergent Validity. The *CMCS* task was also found to demonstrate modest convergence with an adapted working memory task previously evaluated for use with young children with DS. Working memory involves the temporary storage of information (as in STM), and an additional manipulation and processing element, and is considered a fundamental cognitive regulatory skill that contributes to executive function. The modest association observed in the present study suggests that the two tasks measure related constructs, but they do not capture identical underlying processes. This aligns with accounts of executive function skill acquisition, wherein more complex executive processes are thought to stem from more fundamental earlier skills, such as information processing, attention processes, and simpler forms of executive function skills, like retaining information briefly and delaying responses (Garon et al., 2008). These fundamental skills eventually integrate into higher-order cognitive processes such as actively manipulating information held in mind. The modest association observed between STM and working memory performance is aligned with a developmental emergence account of these dimensions.

Current versus Previous Findings. Because versions of the Corsi span task have been administered across a number of DS studies, an additional dimension worthy of consideration is the degree to which *CMCS* performances in younger children with DS are similar to those reported in previous studies of older individuals. The nature of different Corsi-type task administrations, however, makes it difficult to make a direct comparison. For example, tablet-based tasks like the CANTAB spatial span capture response times, and may show more variability than tabletop games, which have fewer trials and outcome metrics (Lanfranchi et al., 2004; Pennington et al., 2003). Studies using Corsi-type tasks with individuals with DS report variability in the outcome metrics of interest ranging from 0.83 SD to 17 SD (Frenkel & Bourdin, 2009; Pennington et al., 2003), with widely fluctuating maximum scores across tasks. Furthermore, few studies include participants younger than seven years old, making a comparison to other crosssectional DS investigations at this developmental stage difficult. Although the present adaptation of the Corsi task preserves the original cognitive targets, it is likely that the performances observed with the adapted measure differ somewhat from previous studies, given the emphasis on minimizing floor effects and reducing confounds related to language and motor planning.

Implications. Findings from the study contribute to the broader effort to evaluate and validate outcome measures for use with children with DS. This broader effort has the potential to not only improve treatment trial study designs, but also to characterize within-DS heterogeneity in STM performance with much greater specificity. Although measures of STM exist in the literature for use in the general population of children, the administration of those measures often involves using skills beyond STM that are known areas of vulnerability in DS, like language and motor planning. Adjustments to the classic span tasks in ways that minimize receptive language confounds but preserve the STM span component of the task make it possible to more accurately evaluate child performances and ultimately child skill acquisition in intervention and treatment studies. Future psychometric evaluations should seek to extend the developmental range for this modified Corsi task even further, perhaps by reducing the number of locations to decrease the information processing demands of the task in its current form.

Advancing intervention work in this area is especially promising, in that some studies have also found moderate or far-transfer effects for broader memory and

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cognitive training on untrained measures of visuospatial STM in both general and clinical populations, including children with ADHD, children with poor working memory skills, and adolescents with mild to borderline intellectual disabilities (Klingberg et al., 2005; Norris et al., 2019; Van der Molen et al., 2010). In particular, the computer-based training program Cogmed has demonstrated well-established transfer effects to measures of verbal and visuospatial STM (for a meta-analytic review, see Bharadwaj et al., 2022). Transfer effects to visuospatial STM have even been observed in the DS population, with Cogmed demonstrating feasibility and effectiveness in enhancing visuospatial STM among children 7 to 12 years old with DS (Bennett et al., 2013). Collectively, these findings underscore the promising modifiability of visuospatial STM in clinical populations, including individuals with DS. Capturing these potential effects with validated measures in this population could facilitate an exciting and potentially impactful line of intervention studies for young children with DS.

Limitations. This study contributes new knowledge regarding the measurement of STM and the distribution of performances in children with DS. However, findings from this study must be interpreted within the context of several methodological limitations. First, the data analyzed were cross-sectional, aside from a two-week testretest time window. Therefore, age-related performances can only be interpreted within a cross-sectional data context, and age-related differences should not be construed as having been derived from longitudinal performances and change over time. In addition, the estimations generated by the Quasi-Poisson analyses are preliminary and will require replication for more precise prediction of performances in this population based on CA, MA, and CSS. To truly interpret child performances in this population, future

studies should seek to establish CA-based norms for this measure with a larger sample of participants with DS.

An important limitation of this study relates to the homogeneity of participant characteristics. Although there was substantial heterogeneity in child dimensions like MA, participants from the study came from primarily white and non-Hispanic racial and ethnic backgrounds, and as such cannot generalize to the broader population of children within the United States from racially, ethnically, and language of origin minoritized groups. Future work must implement more tailored outreach and engagement approaches to all segments of the DS population to establish true representation in study samples across many different identity groups.

Another limitation is the establishment of convergent validity mainly by comparing the scores of the *CMCS* STM task with a recently evaluated measure of working memory in young children with DS. Although STM and working memory are thought to be related, they are also recognized as distinct cognitive components (Aben et al., 2012). Thus, the modest correlation between the two tasks serves as evidence of the expected result. As more measures of STM are assessed and validated for young children with DS, the construct validity of the *CMCS* task can be further ascertained.

It is also important to note that only a subgroup of children returned within a two-week window to assess test-rest reliability and practice effects. Replication of these effects in a larger sample would provide further evidence for these important psychometric properties. Finally, due to the CA and MA range for this study, both the Bayley-4 Cognitive Scale and the SB-5 ABIQ were used to derive MA scores. This approach allowed for participants with scores at the floor of the SB- 5 (\leq 2-year age equivalent) to have values that better approximated their overall cognitive status.

However, not all participants were administered both assessments for time- and agerelated reasons, and as a result, a precise MA could not be captured for a subgroup of participants who performed at the floor of the SB-5. Ideally, one measure would have been available for the entire sample of participants, but to date, no such measure exists to assess cognitive status across this chronological and developmental age range. For this reason, AE scores were utilized in data analysis for evaluating developmental sensitivity across the full sample. While each type of score has its own strengths and weaknesses, ability scores that preserve the variability of raw scores while allowing for reliable differentiation at extreme values may be preferable in samples with neurodevelopmental disorders (Farmer et al., 2020). Thus, we also modeled developmental sensitivity using CSS from the SB-5 ABIQ when available, but these scores were only available for participants in the appropriate age range for the SB-5.

Future Directions. Despite the limitations described above, the promising psychometric findings presented in this study contribute to the growing effort to validate measures of early cognitive processes in DS. Future work should seek to increase the sample size for performance on the *CMCS* task, particularly for individuals ages 5 to 8 years, or perhaps even older than 8 years, to facilitate the establishment of DS group-level norms, which can then be used to characterize and contextualize individual performances and capture treatment effects.

References

- Aben, B., Stapert, S., & Blokland, A. (2012). About the distinction between working memory and short-term memory. *Frontiers in Psychology*, *3*. https://doi.org/10.3389/fpsyg.2012.00301
- Antonarakis, S. E., Skotko, B. G., Rafii, M. S., Strydom, A., Pape, S. E., Bianchi, D. W., Sherman, S. L., & Reeves, R. H. (2020). Down syndrome. *Nature Reviews Disease Primers*, *6*(1). https://doi.org/10.1038/s41572-019- 0143-7
- Bayley, N., & Aylward, G. P. (2019). Bayley Scales of Infant and Toddler Development fourth edition (Bayley-4). *Bloomington, MN: NCS Pearson*.
- Bennett, S. J., Holmes, J., & Buckley, S. (2013). Computerized memory training leads to sustained improvement in visuospatial short-term memory skills in children with Down syndrome. *American Journal on Intellectual and Developmental Disabilities*, *118*(3), 179–192.
- Berg, D. H. (2008). Working memory and arithmetic calculation in children: The contributory roles of processing speed, short-term memory, and reading. *Journal of Experimental Child Psychology*, *99*(4), 288–308. https://doi.org/10.1016/j.jecp.2007.12.002
- Bharadwaj, S. V., Yeatts, P., & Headley, J. (2022). Efficacy of cogmed working memory training program in improving working memory in school-age children with and without neurological insults or disorders: A metaanalysis. In *Applied Neuropsychology: Child* (Vol. 11, Issue 4, pp. 891– 903). Routledge. https://doi.org/10.1080/21622965.2021.1920943
- Blankenship, T. L., Slough, M. A., Calkins, S. D., Deater-Deckard, K., Kim-Spoon, J., & Bell, M. A. (2019). Attention and executive functioning in infancy: Links to childhood executive function and reading achievement. *Developmental Science*, *22*(6). https://doi.org/10.1111/desc.12824
- Brock, J., & Jarrold, C. (2005). Serial order reconstruction in Down syndrome: Evidence for a selective deficit in verbal short-term memory. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, *46*(3), 304–316. https://doi.org/10.1111/j.1469-7610.2004.00352.x
- Bull, R., Espy, K. A., & Wiebe, S. A. (2008). Short-term memory, working memory, and executive functioning in preschoolers: Longitudinal predictors of mathematical achievement at age 7 years. *Developmental Neuropsychology*, *33*(3), 205–228. https://doi.org/10.1080/87565640801982312
- Caviola, S., Mammarella, I. C., Cornoldi, C., & Lucangeli, D. (2009). A metacognitive visuospatial working memory training for children. *International Electronic Journal of Elementary Education*, *2*(1). https://www.iejee.com/index.php/IEJEE/article/view/261
- Corsi, P. M. (1972). *Human memory and the medial temporal region of the brain*. McGill University.
- Cowan, N. (2022). Working memory development in childhood. In M. L. Courage & N. Cowan (Eds.), *The Development of Memory in Infancy and Childhood* (3rd ed., pp. 111–145). Psychology Press. https://doi.org/10.4324/9781003016533
- Daunhauer, L. A., & Fidler, D. J. (2011). The Down syndrome behavioral phenotype: Implications for practice and research in occupational therapy. *Occupational Therapy in Health Care*, *25*(1), 7–25. https://doi.org/10.3109/07380577.2010.535601
- Devine, R. T., Ribner, A., & Hughes, C. (2019). Measuring and predicting individual differences in executive functions at 14 months: A longitudinal study. *Child Development*, *90*(5), e618–e636. https://doi.org/10.1111/cdev.13217
- Diamond, A., Prevor, M. B., Callender, G., & Druin, D. P. (1997). *Prefrontal cortex cognitive deficits in children treated early and continuously for PKU*. *62*(4), 1–206. https://www.jstor.org/stable/1166208
- Enabling Devices. (2019). *Big talk assistive technology communicator [Equipment]*. https://enablingdevices.com/product/big-talk/
- Esbensen, A. J., Hooper, S. R., Fidler, D., Hartley, S. L., Edgin, J., D'Ardhuy, X. L., Capone, G., Conners, F. A., Mervis, C. B., Abbeduto, L., Rafii, M., Krinsky-Mchale, S. J., Urv, T., Dykens, E., Esbenson, A., Hartlay, S., Keller, S., & Weir, S. (2017). Outcome measures for clinical trials in down syndrome. *American Journal on Intellectual and Developmental Disabilities*, *122*(3), 247–281. https://doi.org/10.1352/1944-7558- 122.3.247
- Farmer, C. A., Kaat, A. J., Thurm, A., Anselm, I., Akshoomoff, N., Bennett, A., Berry, L., Bruchey, A., Barshop, B. A., Berry-Kravis, E., Bianconi, S., Cecil, K. M., Davis, R. J., Ficicioglu, C., Porter, F. D., Wainer, A., Goin-Kochel, R. P., Leonczyk, C., Guthrie, W., … Miller, J. S. (2020). Person ability scores as an alternative to norm-referenced scores as outcome measures in studies of neurodevelopmental disorders. In *American Journal on Intellectual and Developmental Disabilities* (Vol. 125, Issue 6, pp. 475– 480). American Association on Mental Retardation. https://doi.org/10.1352/1944-7558-125.6.475
- Fidler, D. J. (2005). The emerging Down syndrome behavioral phenotype in early childhood. *Infants & Young Children*, *18*(2), 86–103. https://doi.org/10.1097/00001163-200504000-00003
- Frenkel, S., & Bourdin, B. (2009). Verbal, visual, and spatio-sequential shortterm memory: Assessment of the storage capacities of children and teenagers with Down's syndrome. *Journal of Intellectual Disability Research*, *53*(2), 152–160. https://doi.org/10.1111/j.1365- 2788.2008.01139.x
- Gade, M., Zoelch, C., & Seitz-Stein, K. (2017). Training of visual-spatial Working memory in preschool children. *Advances in Cognitive Psychology*, *13*(2), 177–187. https://doi.org/10.5709/acp-0217-7
- Garon, N., Bryson, S. E., & Smith, I. M. (2008). Executive function in preschoolers: A review using an integrative framework. *Psychological Bulletin*, *134*(1), 31–60. https://doi.org/10.1037/0033-2909.134.1.31
- Godfrey, M., & Lee, N. R. (2018). Memory profiles in Down syndrome across development: A review of memory abilities through the lifespan. In *Journal of Neurodevelopmental Disorders* (Vol. 10, Issue 1). BioMed Central Ltd. https://doi.org/10.1186/s11689-017-9220-y
- Grieco, J., Pulsifer, M., Seligsohn, K., Skotko, B., & Schwartz, A. (2015). Down syndrome: Cognitive and behavioral functioning across the lifespan. *American Journal of Medical Genetics, Part C: Seminars in Medical Genetics*, *169*(2), 135–149. https://doi.org/10.1002/ajmg.c.31439
- Harrison, T. L., Shipstead, Z., Hicks, K. L., Hambrick, D. Z., Redick, T. S., & Engle, R. W. (2013). Working memory training may increase working memory capacity but not fluid intelligence. *Psychological Science*, *24*(12), 2409–2419. https://doi.org/10.1177/0956797613492984
- Hessl, D., Sansone, S. M., Berry-Kravis, E., Riley, K., Widaman, K. F., Abbeduto, L., Schneider, A., Coleman, J., Oaklander, D., & Rhodes, K. C. (2016). The NIH Toolbox Cognitive Battery for intellectual disabilities: Three preliminary studies and future directions. *Journal of Neurodevelopmental Disorders*, *8*(1), 1–18. https://doi.org/10.1186/s11689-016-9167-4
- Hick, R. F., Botting, N., & Conti-Ramsden, G. (2007). Short-term memory and vocabulary development in children with Down syndrome and children with specific language impairment. *Developmental Medicine & Child Neurology*, *47*(8), 532–538. https://doi.org/10.1111/j.1469- 8749.2005.tb01187.x
- Holmes, J., Gathercole, S. E., & Dunning, D. L. (2009). Adaptive training leads to sustained enhancement of poor working memory in children. *Developmental Science*, *12*(4). https://doi.org/10.1111/j.1467- 7687.2009.00848.x
- Holmes, J., Gathercole, S. E., Place, M., Dunning, D. L., Hilton, K. A., & Elliott, J. G. (2010). Working memory deficits can be overcome: Impacts of training and medication on working memory in children with ADHD. *Applied Cognitive Psychology*, *24*(6), 827–836. https://doi.org/10.1002/acp.1589
- Jaeggi, S. M., Buschkuehl, M., Jonides, J., & Shah, P. (2011). Short-and longterm benefits of cognitive training. *Proceedings of the National Academy of Sciences*, *108*(25), 10081–10086. https://doi.org/https://doi.org/10.1073/pnas.1103228108
- Jarrold, C., & Baddeley, A. (2001). Short-term memory in Down syndrome: Applying the working memory model. *Down Syndrome Research and Practice*, *7*(1), 17–23. https://doi.org/10.3104/reviews.110
- Jarrold, C., & Baddeley, A. D. (1997). Short-term memory for verbal and visuospatial information in Down's syndrome. *Cognitive Neuropsychiatry*, *2*(2), 101–122. https://doi.org/10.1080/135468097396351
- Kessels, R. P. C., van Zandvoort, M. J. E., Postma, A., Kappelle, L. J., & de Haan, E. H. F. (2000). The Corsi block-tapping task: Standardization and normative data. *Applied Neuropsychology*, *7*(4), 252–258. https://doi.org/10.1207/S15324826AN0704_8
- Klingberg, T., Fernell, E., Olesen, P. J., Johnson, M., Gustafsson, P., Dahlström, K., Gillberg, C. G., Forssberg, H., & Westerberg, H. (2005). Computerized training of working memory in children with ADHD - A randomized, controlled trial. *Journal of the American Academy of Child*

and Adolescent Psychiatry, *44*(2), 177–186. https://doi.org/10.1097/00004583-200502000-00010

Koo, T. K., & Li, M. Y. (2016). A guideline of selecting and reporting intraclass correlation coefficients for reliability research. *Journal of Chiropractic Medicine*, *15*(2), 155–163.

https://doi.org/https://doi.org/10.1016/j.jcm.2016.02.012

Lanfranchi, S., Cornoldi, C., & Vianello, R. (2004). Verbal and visuospatial working memory deficits in children with Down syndrome. *American Journal on Mental Retardation*, *109*(6), 456–466.

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https://doi.org/10.1352/0895-8017(2004)109<456:VAVWMD>2.0.CO;2
Määttä, T., Tervo-Määttä, T., Taanila, A., Kaski, M., & Iivanainen, M. (2006).
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- Mental health, behaviour and intellectual abilities of people with Down syndrome. *Down Syndrome Research and Practice*, *11*(1), 37–43. https://doi.org/10.3104/reports.313
- Martin, G. E., Klusek, J., Estigarribia, B., & Roberts, J. E. (2009). Language characteristics of individuals with Down syndrome. *Topics in Language Disorders*, *29*(2), 112–132.
- https://doi.org/10.1097/TLD.0b013e3181a71fe1 Norris, D. G., Hall, J., & Gathercole, S. E. (2019). Can short-term memory be

trained? *Memory and Cognition*, *47*(5), 1012–1023. https://doi.org/10.3758/s13421-019-00901-z

- Onnivello, S., Schworer, E. K., Prince, M. A., Daunhauer, L. A., & Fidler, D. J. (2022). Early developmental profiles among infants with Down syndrome. *Journal of Intellectual Disability Research*. https://doi.org/10.1111/jir.12997
- Pennington, B. F., Moon, J., Edgin, J., Stedron, J., & Nadel, L. (2003). The neuropsychology of Down syndrome: Evidence for hippocampal dysfunction. *Child Development*, *74*(1), 75–93. https://doi.org/10.1111/1467-8624.00522
- Petrides, M. (1995). Impairments on nonspatial self-ordered and externally ordered working memory tasks after lesions of the mid-dorsal part of the lateral frontal cortex in the monkey. *Journal of Neuroscience*, *15*(1), 359– 375.
- Pinks, M. E., Van Deusen, K., Prince, M. A., Esbensen, A. J., Thurman, A. J., Patel, L. R., Abbeduto, L., Walsh, M. M., Daunhauer, L. A., Feigles, R. T., Nguyen, V., & Fidler, D. J. (2023). Psychometric evaluation of a working memory assessment measure in young children with Down syndrome. *Research in Developmental Disabilities*, *139*, 104564. https://doi.org/10.1016/j.ridd.2023.104564
- Purser, H. R. M., & Jarrold, C. (2005). Impaired verbal short-term memory in Down syndrome reflects a capacity limitation rather than atypically rapid forgetting. *Journal of Experimental Child Psychology*, *91*(1), 1–23. https://doi.org/10.1016/j.jecp.2005.01.002
- R Core Team. (2022). *R: A language and environment for statistical computing*. R Foundation for Statistical Computing. https://www.Rproject.org/
- Rasmussen, C., & Bisanz, J. (2005). Representation and working memory in early arithmetic. *Journal of Experimental Child Psychology*, *91*(2), 137– 157. https://doi.org/10.1016/j.jecp.2005.01.004
- Roid, G. H. (2003a). *Stanford Binet intelligence scales, technical manual* (5th ed.). Riverside Publishing.
- Roid, G. H. (2003b). Stanford-Binet Intelligence Scales–Fifth Edition. *Itasca, IL: Riverside Publishing*.
- Schworer, E. K., Esbensen, A. J., Fidler, D. J., Beebe, D. W., Carle, A., & Wiley, S. (2022). Evaluating working memory outcome measures for children with Down syndrome. *Journal of Intellectual Disability Research*, *66*(1– 2), 195–211. https://doi.org/10.1111/jir.12833
- Schworer, E. K., Soltani, A., Altaye, M., Fidler, D. J., & Esbensen, A. J. (2023). Cognitive flexibility assessment in youth with Down syndrome: Reliability, practice effects, and validity. *Research in Developmental Disabilities*, *133*, 104416. https://doi.org/10.1016/j.ridd.2022.104416
- Tungate, A. S., & Conners, F. A. (2021). Executive function in Down syndrome: A meta-analysis. *Research in Developmental Disabilities*, *108*, 103802. https://doi.org/10.1016/j.ridd.2020.103802
- Van der Molen, M. J., Van Luit, J. E. H., Van der Molen, M. W., Klugkist, I., & Jongmans, M. J. (2010). Effectiveness of a computerised working memory training in adolescents with mild to borderline intellectual disabilities. *Journal of Intellectual Disability Research*, *54*(5), 433–447. https://doi.org/10.1111/j.1365-2788.2010.01285.x
- Ver Hoef, J. M., & Boveng, P. L. (2007). Quasi-poisson vs. negative binomial regression: How should we model overdispersed count data? *Ecology*, *88*(11), 2766–2772. https://doi.org/10.1890/07-0043.1
- Vicari, S., & Carlesimo, G. A. (2006). Short-term memory deficits are not uniform in Down and Williams syndromes. *Neuropsychology Review*, *16*(2), 87–94. https://doi.org/10.1007/s11065-006-9008-4
- Wedderburn, R. W. M. (1974). Quasi-Likelihood Functions, Generalized Linear Models, and the Gauss-Newton Method. *Biometrika*, *61*(3), 439–447. https://www.jstor.org/stable/2334725
- Yang, Y., Conners, F. A., & Merrill, E. C. (2014). Visuo-spatial ability in individuals with Down syndrome: Is it really a strength? In *Research in Developmental Disabilities* (Vol. 35, Issue 7, pp. 1473–1500). Elsevier Inc. https://doi.org/10.1016/j.ridd.2014.04.002

Figures

Figure 1. Photo of the *Early Childhood Modified Corsi Span* materials.

Figure 2. Boxplot of *Early Childhood Modified Corsi Span* score by chronological age.

Figure 3. Boxplot of *Early Childhood Modified Corsi Span* score by mental age.

Figure 4. Relationship between *Early Childhood Modified Corsi Span* score and chronological

age, with a Quasi-Poisson regression line.

Figure 5. Relationship between *Early Childhood Modified Corsi Span* score and mental age, with a Quasi-Poisson regression line.

Table 1. Adaptations made to the original Corsi Block-Tapping Task for the *CMCS*.

Table 2. Demographic information (n=110).

Chronological Age	Corsi Total Score
	0.81
$\overline{4}$	1.49
	2.73
$\mathfrak b$	5.00
	9.17
8	16.82

Table 3a. Predicted Corsi score values by chronological age based on the current sample.

Table 3b. Predicted Corsi score values by mental age based on the current sample.